

Facultad de Ciencias de la Educación Departamento de Psicología Programa de Doctorado de Ciencias Sociales y Jurídicas

### **DOCTORAL DISSERTATION**

Early Differential Diagnosis in Infant Population with Neurodevelopmental Disorders. Psychophysiological Markers Based on Eye Tracking Methodology

### **TESIS DOCTORAL**

Diagnóstico Diferencial Temprano en Población Infantil con Trastornos del Neurodesarrollo. Marcadores Psicofisiológicos Basados en Metodología de Seguimiento Ocular

Córdoba, 21 de diciembre de 2021

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### **Informe del director**



TÍTULO DE LA TESIS: Early Differential Diagnosis in Infant Population with Neurodevelopmental Disorders. Analysis of Psychophysiological Markers Based on Eye-Tracking Methodology.

DOCTORANDO/A: Julia Vacas Ruiz

#### INFORME RAZONADO DEL/DE LOS DIRECTOR/ES DE LA TESIS

(se hará mención a la evolución y desarrolio de la tesis, así como a trabajos y publicaciones derivados de la misma).

La Tesis Doctoral: "Early Differential Diagnosis in Infant Population with Neurodevelopmental Disorders. Analysis of Psychophysiological Markers Based on Eye-Tracking Methodology", que presenta la doctoranda **Julia Vacas Ruiz** es un trabajo de investigación original que cuenta con los indicios de calidad científicoacadémicos necesarios para poder defender su Tesis Doctoral por compendio de artículos y mención Internacional.

Consideramos que esta tesis aborda un tema relevante socialmente y hace una aportación significativa al conocimiento de los trastornos del neurodesarrollo, concretamente los Trastornos del Desarrollo del Lenguaje (TEDL) y los Trastornos del Espectro Autista (TEA).

La revisión de la literatura incluida en la tesis, sobre la competencia emocional y el uso de metodología de seguimiento ocular en la investigación de los trastornos abordados, ha sido amplia y rigurosa.

La utilización en la tesis de la metodología de seguimiento ocular con población infantil con trastornos del neurodesarrollo ha supuesto el reto más importante para el desarrollo de esta tesis, y ha sido resuelto por la doctoranda de forma muy satisfactoria. Este tipo de metodología aporta una nueva forma de abordar la investigación sobre el procesamiento de información socio-emocional en población infantil, y supera las limitaciones que tienen otras medidas.

Los resultados de la tesis pueden ser de enorme ayuda a la práctica clínica, y a largo plazo el desarrollo de esta línea de investigación podría llegar a transferirse implementándose en el contexto asistencial real.

Las conclusiones alcanzadas pueden ayudar al desarrollo de instrumentos de evaluación objetivos y fiables, que permitan la detección temprana y el diagnóstico diferencial de trastornos especialmente difíciles de diferenciar en estas edades. Además, teniendo en cuenta la importancia de una intervención temprana específica y sus consecuencias sobre el pronóstico de estos trastornos, desarrollar este tipo de instrumentos de evaluación puede tener un impacto sobre la salud y la calidad de vida de esta población y sus familias, además de un beneficio para la sociedad en general. El trabajo de investigación desarrollado ha sido publicado en revistas científicas sometidas a procesos de revisión por pares, indexadas en las principales bases de datos. Como resultado de esta tesis se han publicado cinco artículos:

Vacas, J., Antolí, A., Sánchez-Raya, A., & Pérez-Dueñas, C. (2021) Eye Tracking Methodology for Studying Emotional Competence in Children with Autism Spectrum Disorder (ASD) and Specific Language Impairment (SLI): a Comparative Research Review. Review Journal of Autism and Developmental Disorders.

Journal Citation Report (JCR) Índice de impacto: 2.600 (Q2). Posición en área temática: 35/78 (Psicología del Desarrollo).

Vacas, J., Antolí, A., Sánchez-Raya, A., Pérez-Dueñas, C., & Cuadrado, F. (2021) Visual preference for social vs. non-social images in young children with autism spectrum disorders. An eye tracking study. *PLoS ONE*, 16(6), e0252795.

Journal Citation Report (JCR) Índice de impacto: 3.240 (Q2). Posición en área temática: 26/73 (Ciencias Multidisciplinarias).

Vacas, J., Antolí, A., Sánchez-Raya, A., & Pérez-Dueñas, C. (2021) Emotional Competence in Children with Autism Spectrum Disorders and Specific Language Impairment: A Comparative Research Review. Education and Training in Autism and Developmental Disabilities, 56(3), 306–327.

Journal Citation Report (JCR) Indice de impacto: 1.578 (Q3). Posición en áreas temáticas: 54/74 (Rehabilitación) y 32/44 (Educación Especial).

Vacas, J., Antolí, A., Sánchez-Raya, A., & Cuadrado, F. (2020) Análisis de Perfiles Cognitivos en Población Clínica Infantil con Trastornos del Neurodesarrollo. *Revista Iberoamericana de Diagnóstico y Evaluación – e Avaliação Psicológica (RIDEP)*, 54(1), 35-46.

Journal Citation Report (JCR) Índice de impacto: 1.000 (Q4). Posición en área temática: 115/131 (Psicología Clínica)

Vacas, J., Antolí, A., Sánchez-Raya, A., Pérez-Dueñas, C., & Cuadrado, F. (2021) Migración diagnóstica entre Trastorno Específico del Lenguaje y Trastorno del Espectro Autista: Estudio exploratorio del impacto en los Centros de Atención Infantil Temprana. *Revista de Investigación en Logopedia*, 11(Special Issue), 77-88. Emerging Sources Citation Index (ESCI). Posición en áreas temáticas en Journal Citation Indicator (JCI): 231/259 (Lingüística) y 438/911 (Lenguaje y Lingüística).

Scopus. Índice de impacto de la revista: 0.19 (Q4).

Además, un artículo está en fase de revisión:

Vacas, J., Antolí, A., Sánchez-Raya, A., Pérez-Dueñas, C., & Cuadrado, F. (under review) Social attention and autism in early childhood: Evidence on psychophysiological markers based on visual scanning of emotional faces with eye tracking methodology.

También, un artículo en preparación, incluido en la tesis, y que añade la comparación de las tres poblaciones (TEA, TEDL y de desarrollo típico) en la metodología de seguimiento ocular y que contribuye con un posible patrón diferenciador entre los distintos trastornos estudiados.

Los resultados de la tesis han sido difundidos en congresos y jornadas en formato de ponencias, comunicaciones y pósteres. Algunas de las más relevantes: XIV Encuentro de Servicios Psicológicos y Psicopedagógicos Universitarios (2017), III International Congress of Clinical and Health Psychology on Children and Adolescents (2017) y VII International Congress of Clinical and Health Psychology on Children and Adolescents (2021).

La doctoranda ha realizado una estancia de investigación internacional de tres meses en el Departamento de Psicología Social de la University of Amsterdam (Holanda) desde 01/10/2019 a 31/12/2019; colaborando en el proyecto 'The role of face processing strategies in emotion recognition across cultures'.

Especialmente valorable es el esfuerzo realizado por la doctoranda para redactar la tesis doctoral en idioma inglés con el objetivo de facilitar su difusión internacional.

Durante su periodo formativo, la doctoranda ha participado en diversas acciones formativas dirigidas a aumentar su competencia científica e investigadora que han sido un complemento coherente con los estudios y tema desarrollado en la presente tesis.

Por todo esto, recomendamos su aceptación y autorización para realizar el acto de defensa pública para la obtención del título de doctora por compendio de artículos y mención internacional.

Y como directoras autorizamos la presentación de la tesis doctoral.

Córdoba, 20 de diciembre de 2021

Firma de las directoras

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A Helena, aunque nos dejaste muy pronto tu *Hamor* y tu Huella se quedarán con nosotros siempre. *Panta rei*.

> "Someday I'll wish upon a star And wake up where the clouds are far behind me Where troubles melt like lemon drops Away above the chimney tops That's where you'll find me.

> > Somewhere over the rainbow Bluebirds fly Birds fly over the rainbow Why then, oh why can't I."

(The Wizard of Oz, Somewhere over the rainbow, 1939)

### Agradecimientos

En estas primeras líneas me gustaría hacer un reconocimiento especial a todas las personas que han hecho posible este trabajo.

A Adoración Antolí y a Araceli Sánchez, que vieron en mí cualidades que ni yo misma sabía que tenía y me animaron a embarcarme en esta aventura que estaba completamente fuera de mi mente. Gracias por inspirarme, guiarme y animarme en cada paso. Tanto Dori como Araceli han puesto a mi disposición todos sus recursos y su conocimiento para que esta tesis saliera adelante, pero sin duda, la aportación que más valoro por su parte es la calidad humana y los valores de trabajo cooperativo y constructivo que me han enseñado, no desde la teoría sino desde la práctica individual, convirtiéndose así en una inspiración tanto a nivel personal como profesional. A pesar de haber estado inmersa en este proyecto durante tanto tiempo y haber enfrentado situaciones muy difíciles, el clima de trabajo, respeto y unión que habéis creado lo ha hecho mucho más fácil. Gracias por todo.

A Vicente Sánchez, quien ya abrió el camino de esta tesis incluso antes de que naciera la idea. Él, sin saberlo, encendió mi curiosidad por la atención infanto-juvenil y por la investigación. Sin esa primera toma de contacto en el máster, no habríamos llegado hasta aquí. Ha sido un auténtico privilegio haberle conocido.

A Carolina Pérez, que ha sido una directora más de este proyecto. Aunque no conste en documentos oficiales, quiero dejar plasmado en papel mi más sincero agradecimiento hacia ella por poner a mi disposición su amplia experiencia sin esperar nada a cambio, por animarme a embarcarme en aventuras internacionales y servirme siempre como modelo de referencia.

A Fátima Cuadrado, ¿compañera, investigadora, supervisora? Sobre todo, amiga. Fátima me enseñó que la investigación también puede ser divertida y que, cuando no lo es, resulta un alivio poder compartirlo con otras personas. He aprendido de ella en cada paso del camino, nos hemos visto crecer como investigadoras pre y post doctorales e incluso hemos unido líneas de investigación. Gracias, amiga.

A la Dra. Disa Sauter de la Universidad de Ámsterdam, quien, junto a todo su equipo, me acogió con los brazos abiertos en su laboratorio poniendo a mi disposición todos sus recursos y su entusiasmo y haciéndome sentir una más desde el primer momento. Gracias a la Dra. Sauter y a su equipo no sólo aprendí nuevas formas de trabajo y me desarrollé como investigadora, sino que también se consolidó mi deseo de asumir retos internacionales.

Al departamento de Psicología de la Universidad de Córdoba, formado por grandes profesionales con ganas de emprender nuevos proyectos y crear redes de cooperación, apostando siempre por una política de plena inclusión. Me he sentido muy cómoda trabajando en él y compartiendo proyectos y actividades divulgativas con grandísimos profesionales.

A todos los centros que nos abrieron sus puertas para desarrollar nuestros estudios: CAIT-UCO, Asociación Autismo Córdoba, CEIP Al-Andalus, CAIT-Fundación PRODE... Así como a todos los y las profesionales que facilitaron nuestra labor en sus centros y a todas las familias que aceptaron participar en los estudios con la convicción de que la investigación conduce al avance social. Gracias por su confianza y por apostar por la ciencia como motor de desarrollo.

A mis padres y hermanos, para quienes no tengo suficientes palabras de agradecimiento. Gracias por enseñarme el valor de lo diferente, ahora entiendo que la diversidad no solo es deseable sino necesaria. También os agradezco vuestra apuesta por el humor como medio de afrontar la vida; si bien no resuelve todos los problemas, sí ayuda a sobrellevarlos. Pero, sobre todo, gracias por existir y estar ahí.

En definitiva, me siento muy afortunada por todas las personas que forman parte de mi vida, a nivel profesional y personal. A todas ellas, las que aparecen en estas líneas y las que por falta de espacio no están, gracias.

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## **List of Abbreviations**

Abbreviation	Explanation		
ADFES	Amsterdam Dynamic Facial Expression Set		
ADHD	Attention-Deficit/Hyperactivity Disorder		
AE	Autism Europe		
AOI	Area of Interest		
APA	American Psychiatric Association		
ASD	Autism Spectrum Disorders		
ASD-LD	Autism Spectrum Disorders with Language Delay		
ASD-NL	Autism Spectrum Disorders with Normal Language		
BESD	Behavioral, Emotional, and Social Difficulties		
CD	Communication Disorders		
CD-CU	Conduct Disorder with traits of antisocial behavior		
CI	Circumscribed Interests		
CIO	CI-related Objects		
non-CIO	non-CI-related Objects		
СТ	Contextual Theory of Mind		
DEC	Division for Early Childhood		
DEFSS	Developmental Emotional Faces Stimulus Set		
DLD	Developmental Language Disorder		
DSM	Diagnostic and Statistical Manual of Mental Disorders		
EBP	Evidence-Based Practices		
EC	Emotional Competence		
ECI	Early Childhood Intervention		
ECIC	Early Childhood Intervention Center		
EEG	Electroencephalograms		
EI	Emotional Intelligence		
EIH	Empathy Imbalance Hypothesis		
ER	Emotion Recognition		
ERC	Emotion Regulation Checklist		
FER	Facial Emotion Recognition		
fMRI	Functional Magnetic Resonance Imaging		
ICD	International Statistical Classification of Diseases and Related Health Problems		
ID	Intellectual Disability		
IQ	Intelligence Quotient		
JĂ	Joint Attention		
KDEF	Karolinska Directed Emotional Faces		
М	Mean		
NDD	Neurodevelopmental Disorders		
NEPSY	Developmental Neuropsychological Assessment		
NMD	Neurodevelopmental Motor Disorders		
NOS	Not Otherwise Specified		
PPTV	Peabody Picture Vocabulary Test		

PTFD	Proportion of Total Fixation Duration
SA	Social Attention
SD	Standard Deviation
SDQ	Strengths and Difficulties Questionnaire
SLD	Specific Learning Disorder
SLI	Specific Language Impairment
SPCD	Social (Pragmatic) Communication Disorder
TABS	Temperament and Atypical Behavior Scale
TD	Typically Developing
TFF	Time to First Fixation
ТоМ	Theory of Mind
TPV	Time per Visit
TRSAF	Total Raw Score Affect Recognition
TRST	Total Raw Score Theory of Mind
VSP	Visual Scanning Pattern
VT	Verbal Theory of Mind
WISC	Wechsler Intelligence Scale for Children
WOS	World Health Organization

# **Spanish Abbreviations**

Abbreviation	Explanation		
AT	Atención Temprana		
CAIT	Centro de Atención Infantil Temprana		
CI	Coeficiente Intelectual		
CIE	Clasificación Estadística Internacional de Enfermedades y Problemas		
	Relacionados con la Salud		
CIT	Coeficiente Intelectual Total		
CV	Comprensión Verbal		
DAL	Dificultades en el Aprendizaje de la Lectura		
DT	Desviación Típica		
ET	Error Típico		
GAT	Grupo de Atención Temprana - Federación Estatal de Asociaciones		
	de Profesionales de Atención Temprana		
ICC	Índice de Competencia Cognitiva		
ICG	Índice de Capacidad General		
M	Media		
MT	Memoria de Trabajo		
PBE	Prácticas Basadas en la Evidencia		
ODAT	Organización Diagnóstica de Atención Temprana		
OMS	Organización Mundial de la Salud		
RF	Razonamiento Fluido		
RP	Razonamiento Perceptivo		
SA	Síndrome de Asperger		
TDAH	Trastorno por Déficit de Atención e Hiperactividad		
TDAH-C	Trastorno por Déficit de Atención e Hiperactividad de tipo		
	Combinado		
TDAH-I	Trastorno por Déficit de Atención e Hiperactividad		
	predominantemente Inatento		
TEA	Trastorno del Espectro Autista		
TEL	Trastorno Específico del Lenguaje		
TND	Trastornos del Neurodesarrollo		
UAIT	Unidad de Atención Infantil Temprana		
USMIJ	Unidad de Salud Mental Infantojuvenil		
VP	Velocidad de Procesamiento		
VS	Visuoespacial		

#### **Extended Abstract**

Early diagnosis and intervention have a positive impact on prognosis and the quality of life of children with neurodevelopmental disorders (NDDs) and their families. In this sense, the aim of this thesis project is to contribute to the early differential diagnosis of children with NDDs. To do so, we firstly investigated the cognitive profiles of five NDD with some kind of language disruption using the Wechsler Intelligence Scale for Children (see Appendix 1). From this study, we realized two issues: (1) That cognitive profiles were not highly conclusive or helpful for the purpose of differential diagnosis and (2) That we needed to bring forward the age of detection of the disorder to make real progress in early diagnosis. Thus, we carried out a comprehensive review on the issue and we focused the project on the analysis of the emotional competence (EC) in young children with Autism Spectrum Disorders (ASD) and Developmental Language Disorder (DLD).

On the one hand, EC has proved to be more conclusive in yielding differences between disorders at early ages. On the other hand, ASD and DLD are two different NDDs which have demonstrated many similarities in behavioral, cognitive, and linguistic profiles at early ages. This inevitably hampers early diagnosis and, consequently, early intervention as revealed in our study on the prevalence and impact of the 'Diagnostic Migration' phenomenon between ASD and DLD on early intervention (see Appendix 2). Thus, the present doctoral dissertation was conceived to contribute to differential diagnosis of these conditions at early ages regarding their abilities in EC. With this purpose, we firstly conducted a thorough review of the state of the art and we understood that the construct of EC was too broad to be fully covered in this project; however, we found some key abilities which had been revealed as promising in differentiating disorders at early ages. These abilities were related to the visual processing of socialemotional images, whose analysis required eye tracking methodology. Then, we designed three intertwined experimental eye tracking studies to yield new evidence on the matter.

Therefore, the first part of this work comprises three chapters which unfold the current knowledge on early diagnosis of NDDs, the evaluation of the EC in children with ASD and those with DLD, and the potential of eye tracking methodology to contribute to the definition of both conditions as well as to differential diagnosis.

The second part of this work includes four chapters describing the experimental studies carried out. The former one explains the rationale of the three studies as well as their goals, the participants involved in them, the main hypotheses, and the designs. The other three chapters present a detailed reproduction of each study. As it was previously mentioned, we applied an eye tracking methodology along with a paired preference paradigm in all studies to describe the way in which young children with ASD and DLD observe and process social-emotional images. In this sense, the eye tracking methodology allowed us to describe children eye movements during images visualization; while the paired preference paradigm (consisting of the presentation of pairs of images to analyze their saliency and the competing effect of each one on each other) enabled us to identify which stimuli were more visually salient for children and which were able to capture or prevent their attention. Thus, by applying these methodological considerations in our three studies we have unveiled some psychophysiological markers that may contributed to the early identification of children with ASD and DLD (e.g., late orientation to angry and child faces, emotional sensitivity -visual preference for emotional faces with respect to the neutral ones-, and more superficial facial processing compared to their typical controls).

Finally, the third part of this work consists of one chapter discussing the main results derived from the whole project, stating its limitations, providing some guidelines for future research, and remarking some final conclusions.

Hence, this doctoral dissertation scientifically contributes to the current knowledge on the field in several ways: (1) Providing an exhaustive revision of the insight into the social-emotional competence in child populations with ASD and DLD; (2) Developing and consolidating a specific methodology based on the eye tracking technology and the paired preference paradigm, which allows a thorough study of these clinical populations boosting their comprehension and differentiation; (3) Revealing psychophysiological markers in young children with ASD and potential descriptors of visual scanning of faces in young children with DLD; and (4) Indicating new pathways for addressing the issue in future studies.

#### Introduction

The present doctoral dissertation arose from a concern on the impact of language difficulties on children's everyday functioning and wellbeing. As Reilly et al. (2014, p. 419) postulated: "Language skills have become critically important for the workforce and for economic prosperity. However, these skills have their origins early in life". This statement reflects the idea that society has changed in the last decades in such a way that nowadays language abilities are crucial for a successful social performance and personal realization. Following the line of Reilly et al. (2014), nobody with language difficulties would have been considered to be impaired or have a disorder in the first decades of the previous century, since social and working demands were different from current ones. However, communication skills have gained ground on modern society until becoming a requirement for personal, social, academic, and professional development. Moreover, according to the definition of health of the World Health Organization (WOS, 1948), being healthy implies a global state of wellbeing, not just the lack of disease or impairment. These are just some reasons why language disorders must be attended. Getting back to the statement of Reilly et al. (2014), language is acquired during the infancy, thus that is the pivotal moment to detect any linguistic deviation or delay and to take actions to reduce its impact on children's lives.

In order to better understand the myriad of language difficulties appearing during infancy and childhood and how they affect to the child development, we carried out a preliminary study on cognitive profiles of school-aged children with several neurodevelopmental disorders (NDDs) with a common affection in different aspects of language (see Appendix 1). Particularly in this study, we focused on Specific Learning Disorders, Specific Language Impairment, Asperger Syndrome and Attention-Deficit/Hyperactivity Disorder in two dimensions (Predominantly Inattentive and Combined). We used data from the Wechsler Intelligence Scale for Children 4<sup>th</sup> edition (WISC-IV) and we found interesting data regarding the within heterogeneity of some profiles. We also unveiled some clues which contributed to the understanding of these disorders; however, regarding differential diagnosis, we were only able to distinguish children with Specific Language Impairment from those with other disorders, and the verbal comprehension index of the WISC-IV did not yield any relevant data to disentangle the linguistic profiles of these populations.

After this attempt to seek into cognitive profiles of children with NDDs as a way of contributing to differential diagnosis, we made a thorough literature review on language disorders, their characterization, screening, and diagnosis, and we found that (at least at early ages) children with language disorders and those with autism spectrum disorders (ASD) present similar behavioral, cognitive, and linguistic profiles. This gave rise to our study on 'diagnostic migration' (see Appendix 2), which draws attention to the difficulties that professionals face to distinguish between both kinds of disorders at early ages and to assign an early diagnosis. Furthermore, this literature review provided us some clues to guide the experimental part of this thesis project.

In this sense, it revealed us all the possibilities that the study of the emotional competence (EC) could bring to us for comparing clinical populations. Particularly, research including psychophysiological measures of any construct related to the EC seemed to be the most promising one. Thus, among all the options available, we focused our work on the analysis of the visual processing of social-emotional content in young children with language disorders and those with ASD by applying an eye tracking methodology. This kind of methodology is based on monitoring eye movements during task performance or image visualization; thus, it is a psychophysiological technique that provides objective indexes on individuals visual behavior, which may be understood as specific markers of these clinical conditions. These advantages alongside the fact that this is a non-invasive technique makes it highly suitable for the purposes of this project. Therefore, we may say that the initial literature review set the ground for the actual purposes of this doctoral dissertation: (1) To analyze similarities and differences between young children with DLD and those with ASD in terms of social-emotional abilities, (2) To make use of the potential of the eye tracking technology to fill the gaps of the behavioral tests regarding the description and differentiation of these disorders, (3) To contribute to early differential diagnosis by providing specific markers of each condition.

With the mentioned aims, this work is organized in three different parts. Part one presents a literature review to put the reader into context on the state of the art. This part comprises three chapters: chapter one comments issues related to the reconceptualization of NDDs, the controversies around diagnosis, the difficulties of differential diagnosis, and the relevance of early diagnosis and intervention; chapter two shows a review on the EC in children with language disorders and those with ASD; finally, chapter three adds

to the previous review some knowledge derived from the implementation of the eye tracking methodology in the study of social-emotional abilities in both clinical populations.

On the other hand, part two describes an array of three intertwined experiments that we carried out to disentangle visual processing of social-emotional images in the target populations. Thus, chapter four defines the purposes of these experiments and some common methodological considerations; chapter five introduces our first experimental study on attention to social vs. non-social images in young children with ASD and typical peers; chapter six brings forward our second experimental study on attention to emotional faces comparing young children with ASD with their typical counterparts; and chapter seven presents a comparative study again on attention to social vs. non-social images, which included a sample of children with language disorders. This last study came to yield some hints about differential psychophysiological markers and paved the way for future replications studies.

Finally, it is worthy to point that the content of chapters two, three and five, alongside both Appendixes included at the end of this document were published in sound scientific journals (see the Report on Papers Resulting from this Dissertation, pages 213-220). Likewise, the content of chapter six is currently under review in another high-impact journal. On the other hand, it is also noteworthy that this doctoral dissertation embraces the language principle of "person-first", which implies that when referring to someone's impairment or disability, we firstly name the person and then we mention the condition (e.g., children with ASD instead of ASD children or autistic children). That is the reason why some chapters were rewritten from their published version and adapted to this principle.

# First Part.

### **Review of Literature**

### Chapter 1. The State of the Art. The Importance of Differential Diagnosis and Early Intervention in Neurodevelopmental Disorders

#### **1.1 Introduction**

Infancy and childhood are crucial stages of human development. What happens during the early years of life may be determinant for the future of individuals. Thus, the appropriate growth and maturity of the main developmental areas (physical, affective, social-emotional, cognitive, and personality) during the first years after the birth becomes a relevant aspect to monitor in order to detect any threat to the child's development. When an alteration, deviation, or delay from the typical course of development emerges, it is critical to identify it and intervene on it as soon as possible with the aim of reducing its impact on the child's development and the family dynamic. Likewise, the early response to a developmental condition has proved to enhance the quality of life of all the stakeholders (not only the child, but also the family and the community) and to increase the child's potential of development.

In this chapter, we are going to discuss the current standpoint of the major authorities in the field on neurodevelopmental disorders (NDDs). Moreover, we will describe the main difficulties of differential diagnosis at early ages as well as the relevance of early diagnosis and intervention for the stakeholders. Thus, the purpose of this chapter is to offer the reader a general view of the state of the art, raise awareness about the importance of these issues for individuals' development, and lay the foundations for the present doctoral dissertation.

### **1.2** Considerations on Neurodevelopmental Disorders. New approach in Current Diagnostic Manuals

The term NDDs was coined in the latest version of the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-5; American Psychiatric Association [APA], 2013) and it came to substitute the outdated 'Developmental Disorders', term applied in the previous versions of the manual (e.g., DSM-4-TR; APA, 2000) and widespread to the whole

clinical context. Thereafter, the most recent version of the International Statistical Classification of Diseases and Related Health Problems (ICD-11; World Health Organization [WHO], 2019) went even further with respect to its previous version (ICD-10; WHO, 1992) and included that term in the name of the chapter addressing psychological disorders (Chapter 06-Mental, Behavioral, or Neurodevelopmental Disorders), reinforcing the relevance of this new concept. The mentioned classification systems are the two major references for the clinical and research community: the DSM-5 is the prevailing diagnostic system in the United States, whilst the ICD-11 is the worldwide mainstream medical classification system. According to some academics, each one serves for slightly different purposes: DSM-5 aims at clinical and research utility, while ICD-11 extends these goals to achieve universal applicability, to unify diagnostic language, and to address insurance policies and legal issues (Baird, 2013; Doernberg & Hollander, 2016; Gaebel et al., 2020; Reed et al., 2019; Stein et al., 2020). Despite this difference, APA and WHO have tried to harmonize the latest versions of their classification systems and both reference manuals have made a similar classification of NDDs, including in this category six main conditions: (1) Intellectual Disability (ID), (2) Communication Disorders (CD), (3) Autism Spectrum Disorders (ASD), (4) Attention-Deficit/Hyperactivity Disorder (ADHD), (5) Neurodevelopmental Motor Disorders (NMD), and (6) Specific Learning Disorders (SLD) (Gaebel et al., 2020; Morris-Rosendahl & Crocq, 2020; Reed et al., 2019; Stein et al., 2020). To better understand the relevance of this reconceptualization and to put into context the amendments made in the current classifications with respect to the previous ones, Tables 1.1 and 1.2 show both comparisons between the categorization and codification of NDDs applied in current manuals: DSM-5 (APA, 2013) and ICD-11 (WHO, 2019), with regard to their previous versions: DSM-4-TR (APA, 2000) and ICD-10 (WHO, 1992).

DSM-4-TR	DSM-5
Disorders Usually First Diagnosed in Infancy,	Neurodevelopmental Disorders
Childhood, or Adolescence         Mental Retardation         -       F70 Mild Mental Retardation         -       F71 Moderate Mental Retardation         -       F72 Severe Mental Retardation         -       F73 Profound Mental Retardation         -       F79 Mental Retardation, Severity         Unspecified	<ul> <li>Intellectual Disabilities</li> <li>Intellectual Developmental Disorder</li> <li>F70 Mild</li> <li>F71 Moderate</li> <li>F72 Severe</li> <li>F73 Profound</li> <li>F88 Global Developmental Delay</li> <li>F79 Unspecified Intellectual Disability (Intellectual Developmental Disorder)</li> </ul>
<ul> <li>Communication Disorders</li> <li>F80.1 Expressive Language Disorder</li> <li>F80.2 Mixed Receptive-Expressive Language Disorder</li> <li>F80.0 Phonological Disorder</li> <li>F98.5 Stuttering</li> <li>F80.9 Communication Disorder NOS</li> </ul>	<ul> <li>Communication Disorders</li> <li>F80.2 Language Disorder</li> <li>F80.0 Speech Sound Disorder</li> <li>F80.81 Childhood-Onset Fluency Disorder (Stuttering)</li> <li>F80.89 Social (Pragmatic) Communication Disorder</li> <li>F80.9 Unspecified Communication Disorder</li> </ul>
<ul> <li>Pervasive Developmental Disorders (PDD)</li> <li>F84.0 Autistic Disorder</li> <li>F84.2 Rett's Disorder</li> <li>F84.3 Childhood Disintegrative Disorder</li> <li>F84.5 Asperger's Disorder</li> <li>F84.9 PDD NOS</li> </ul>	<ul><li>Autism Spectrum Disorders</li><li>F84.0 Autism Spectrum Disorder</li></ul>
<ul> <li>Attention-Deficit and Disruptive Behavior Disorders</li> <li>() Attention-Deficit/Hyperactivity Disorder (ADHD) <ul> <li>F90.0 Combined Type</li> <li>F90.8 Predominantly Inattentive Type</li> <li>F90.0 Predominantly Hyperactive- Impulsive Type</li> </ul> </li> <li>F90.9 ADHD NOS</li> <li>F91.8 Conduct Disorder <ul> <li>Childhood-Onset Type</li> <li>Adolescent-Onset Type</li> <li>Unspecified Onset</li> </ul> </li> <li>F91.3 Oppositional Defiant Disorder NOS</li> </ul>	<ul> <li>Attention-Deficit/Hyperactivity Disorder</li> <li>() ADHD <ul> <li>F90.2 Combined Presentation</li> <li>F90.0 Predominantly Inattentive Presentation</li> <li>F90.1 Predominantly Hyperactive-Impulsive Presentation</li> </ul> </li> <li>F90.8 Other Specified ADHD</li> <li>F90.9 Unspecified ADHD</li> </ul>
<ul> <li>Learning Disorders</li> <li>F81.0 Reading Disorders</li> <li>F81.2 Mathematics Disorder</li> <li>F81.8 Disorder of Written Expression</li> <li>F81.9 Learning Disorder NOS</li> </ul>	<ul> <li>Specific Learning Disorder</li> <li>() Specific Learning Disorder</li> <li>F81.0 with Impairment in Reading</li> <li>F81.2 with Impairment in Mathematics</li> <li>F81.81 with Impairment in Written Expression</li> </ul>
Motor Skills Disorder - F82 Developmental Coordination Disorder	Motor Disorders - F82 Developmental Coordination Disorder

**Table 1.1.** Comparison between the classification and codification systems in DSM-4-TR and DSM-5

	- F98.4 Stereotypic Movement Disorder
DSM-4-TR	DSM-5
Disorders Usually First Diagnosed in Infancy, Childhood, or Adolescence	Neurodevelopmental Disorders
<ul> <li>Tic Disorders</li> <li>F95.2 Tourette's Disorder</li> <li>F95.1 Chronic Motor or Vocal Tic Disorder</li> <li>F95.0 Transient Tic Disorder</li> <li>F95.9 Tic Disorder NOS</li> </ul>	<ul> <li>Tics Disorders</li> <li>F95.2 Tourette's Disorder</li> <li>F95.1 Chronic Motor or Vocal Tic Disorder</li> <li>F95.0 Transient Tic Disorder</li> <li>F95.8 Other Specified Tic Disorder</li> <li>F95.9 Tic Disorder NOS</li> </ul>
<ul> <li>Feeding and Eating Disorders of Infancy or Early</li> <li>Childhood</li> <li>F98.3 Pica</li> <li>F98.2 Rumination Disorder</li> <li>F98.2 Feeding Disorder of Infancy or Early Childhood</li> </ul>	
<ul> <li>Elimination Disorders</li> <li>() Encopresis</li> <li>R15 With Constipation and Overflow Incontinence</li> <li>F98.1 Without Constipation and Overflow Incontinence</li> <li>F98.0 Enuresis (Not due to a General Medical Condition)</li> </ul>	
<ul> <li>Other Disorders of Infancy, Childhood, or Adolescence</li> <li>F93.0 Separation Anxiety Disorder</li> <li>F94.0 Selective Mutism</li> <li>F94.2 Reactive Attachment Disorder of Infancy or Early Childhood</li> <li>F98.4 Stereotypic Movement Disorder</li> <li>F98.9 Disorder of Infancy, Childhood, or Adolescence NOS</li> </ul>	<ul> <li>Other Neurodevelopmental Disorders</li> <li>F88 Other Specified Neurodevelopmental Disorder</li> <li>F89 Unspecified Neurodevelopmental Disorder</li> </ul>

NOS, Not otherwise specified

ICD-10	ICD-11
Mental Retardation	Mental, Behavioral, or Neurodevelopmental Disorders
<ul> <li>F70 Mild Mental Retardation</li> <li>F71 Moderate Mental Retardation</li> <li>F72 Severe Mental Retardation</li> <li>F73 Profound Mental Retardation</li> <li>F78 Other Mental Retardation</li> <li>F79 Unspecified Mental Retardation</li> </ul>	<ul> <li>6A00 Disorders of Intellectual Development</li> <li>(DID)</li> <li>6A00.0 DID Mild</li> <li>6A00.1 DID Moderate</li> <li>6A00.2 DID Severe</li> <li>6A00.3 DID Profound</li> <li>6A00.4 DID Provisional</li> <li>6A00.Z DID Unspecified</li> </ul>
Behavioral and Emotional Disorders with Onset Usually Occurring in Childhood and Adolescence	Mental, Behavioral, or Neurodevelopmental Disorders
<ul> <li>F90 Hyperkinetic Disorders</li> <li>F90.0 Disturbance of Activity and Attention</li> <li>F90.1 Hyperkinetic Conduct Disorder</li> <li>F90.8 Other Hyperkinetic Disorders</li> <li>F90.9 Hyperkinetic Disorder, Unspecified</li> </ul>	<ul> <li>6A05 Attention Deficit Hyperactivity Disorder (ADHD)</li> <li>6A05.0 ADHD, Predominantly Inattentive Presentation</li> <li>6A05.1 ADHD, Predominantly Hyperactive- Impulsive Presentation</li> <li>6A05.2 ADHD, Combined Presentation</li> <li>6A05.Y ADHD, Other Specified Presentation</li> <li>6A05.Z ADHD, Presentation Unspecified</li> </ul>
Disorders of Psychological Development	Mental, Behavioral, or Neurodevelopmental Disorders
<ul> <li>F80 Specific Developmental Disorders of Speech and Language</li> <li>F80.0 Specific Speech Articulation Disorder</li> <li>F80.1 Expressive Language Disorder</li> <li>F80.2 Receptive Language Disorder</li> <li>F80.3 Acquired Aphasia with Epilepsy (Landau-Kleffner)</li> <li>F80.8 Other Developmental Disorders of Speech and Language</li> <li>F80.9 Developmental Disorder of Speech and Language, Unspecified</li> </ul>	<ul> <li>6A01 Developmental Speech or Language Disorders</li> <li>6A01.0 Developmental Speech Sound Disorder</li> <li>6A01.1 Developmental Speech Fluency Disorder</li> <li>6A01.2 Developmental Language Disorder (DLD)</li> <li>6A01.20 DLD with Impairment of Receptive and Expressive Language</li> <li>6A01.21 DLD with Impairment of Mainly Expressive Language</li> <li>6A01.22 DLD with Impairment of Mainly Pragmatic Language</li> <li>6A01.23 DLD, with Other Specified Language Impairment</li> <li>6A01.Y Other Specified Developmental Speech or Language Disorders</li> <li>6A01.Z Developmental Speech or Language Disorders, Unspecified</li> </ul>
<ul> <li>F81Specific Developmental Disorders of Scholastic Skills</li> <li>F81.0 Specific Reading Disorder</li> <li>F81.1 Specific Spelling Disorder</li> <li>F81.2 Specific Disorder of Arithmetical Skills</li> <li>F81.3 Mixed Disorder of Scholastic Skills</li> </ul>	<ul> <li>6A03 Developmental Learning Disorder</li> <li>6A03.0 Developmental Learning Disorder with Impairment in Reading</li> <li>6A03.1 Developmental Learning Disorder with Impairment in Written Expression</li> <li>6A03.2 Developmental Learning Disorder with Impairment in Mathematics</li> </ul>

**Table 1.2.** Comparison between the classification and codification systems in ICD-10
 and ICD-11

<ul> <li>F81.8 Other Developmental Disorders of Scholastic Skills</li> <li>F81.9 Developmental Disorder of Scholastic Skills, Unspecified</li> <li>ICD-10</li> <li>Disorders of Psychological Development</li> <li>F82 Specific Developmental Disorder of Motor Function</li> </ul>	<ul> <li>6A03.3 Developmental Learning Disorder with Other Specified Impairment of Learning</li> <li>6A03.Z Developmental Learning Disorder, Unspecified</li> <li>ICD-11</li> <li>Mental, Behavioral, or Neurodevelopmental Disorders</li> <li>6A04 Developmental Motor Coordination Disorder</li> </ul>
F83 Mixed Specific Developmental Disorders	
<ul> <li>F84 Pervasive Developmental Disorders</li> <li>F84.0 Childhood Autism</li> <li>F84.1 Atypical Autism</li> <li>F84.2 Rett Syndrome</li> <li>F84.3 Other Childhood Disintegrative Disorder</li> <li>F84.4 Overactive Disorder Associated with Mental Retardation and Stereotyped Movements</li> <li>F84.5 Asperger Syndrome</li> <li>F84.8 Other Pervasive Developmental Disorders</li> <li>F84.9 Pervasive Developmental Disorder, Unspecified</li> </ul>	<ul> <li>6A02 Autism Spectrum Disorder (ASD)</li> <li>6A02.0 ASD without Disorder of Intellectual Development and with Mild or no Impairment of Functional Language</li> <li>6A02.1 ASD with Disorder of Intellectual Development and with Mild or no Impairment of Functional Language</li> <li>6A02.2 ASD without Disorder of Intellectual Development and with Impaired Functional Language</li> <li>6A02.3 ASD with Disorder of Intellectual Development and with Impaired Functional Language</li> <li>6A02.5 ASD with Disorder of Intellectual Development and with Impaired Functional Language</li> <li>6A02.5 ASD with Disorder of Intellectual Development and with Absence of Functional Language</li> <li>6A02.Y Other Specified ASD</li> <li>6A02.Z ASD, Unspecified</li> </ul>
	<ul> <li>6A06 Stereotyped Movement Disorder (SMD)</li> <li>6A06.0 SMD without Self-Injury</li> <li>6A06.1 SMD with Self-Injury</li> <li>6A06.Z SMD, Unspecified</li> </ul>
F88 Other Disorders of Psychological Development	6A0Y Other Specified Neurodevelopmental Disorders
F89 Unspecified Disorder of Psychological Development	6A0Z Neurodevelopmental Disorders, Unspecified

As the reader may infer from the previous tables, the new term NDDs is not just a simple label change but a real makeover, as it implies an up-to-date reconceptualization of those disorders starting during the early development and impairing functioning in different areas of individuals' daily live. The construct of NDDs is supported by the recognition of a recurrent phenotypic overlapping between neurodevelopmental conditions, which has led researchers and clinicians to advocate for continua instead of discrete categories when describing NDDs (Morris-Rosendahl & Crocq, 2020). In line with that, DSM-5 and ICD-11 are moving from a categorical approach to a dimensional one in which some disorders are viewed as a continuum of impairment along which individuals may be located; thus, differences between subjects rest on specifiers of the severity of symptoms and difficulties, the level of functioning, the course, or the amount of support required, among others variables (Baird, 2013; Doernberg & Hollander, 2016; Gaebel & Kerst, 2019; Gaebel et al., 2020; Reed et al., 2019). The concept of NDD also illustrates some nuances: (1) A lifespan approach according to which these conditions appear in the birth but they persist throughout life, and (2) A neurological cause (Doernberg & Hollander, 2016; Gaebel & Kerst, 2019; Reed et al., 2019; Reed et al., 2019).

Focusing on the target disorders comprised in this doctoral dissertation, ASD is a paradigmatic example of the mentioned reconceptualization of disorders as spectra in which there are not distinct categories but a continuum of impairment with specific levels of symptoms severity and required support (see Tables 1.1 and 1.2). This concept seems to increase specificity and sensitivity of diagnoses and fits better with the heterogeneity associated to this disorder, although the unification of the previous five conditions (Autistic Disorder, Rett' s Disorder, Childhood Disintegrative Disorder, Asperger' s Disorder, and Pervasive Developmental Disorder-Not Otherwise Specified) into one single spectrum in the DSM-5 has been controversial (Wills, 2014). Another major and polemic change relative to the redefinition of ASD was the reduction of core domains of autism from three to two. This change resulted from the combination of the social and communication domains as a panel of experts agreed that both domains were closely related and their distinction responded to arbitrary criteria; however, its implementation has also created debate (Doernberg & Hollander, 2016). At this respect, the fact that current ASD diagnosis relies on the fulfilment of two criteria (social and communicative difficulties and the presence of repetitive and restricted behaviors) led the DSM-5 to include a new diagnostic alternative for those children who only fulfil the first criterium, but not the second one: the Social (Pragmatic) Communication Disorder (SPCD), which was embodied in the 'Communication Disorders' category (see Table 1.1). However, this alternative has demonstrated to have unclear boundaries with respect to ASD or Language Disorder (Taylor & Whitehouse, 2016) and it was not supported by the panel of experts who worked on the conception of the ICD-11 due to the lack of empirical support (First et al., 2021), which reflects the lack of agreement even between the main referential classification system.

Regarding Developmental Language Disorders (DLD), the issue is even more complicated. The term DLD is relatively new as it was coined in the mid-1970s but it was brought out and widespread after a Delphi Consensus Study developed by the CATALISE consortium (Bishop et al., 2016, 2017). It came to replace the previous and more generalized term Specific Language Impairment (SLI), but the issue of the terminology on language disorders has been troublesome from the first time they were described. The evolution of the nomenclature regarding language disorders has been marked by the constant change (not only in terms of labels, but also in their definitions) and the lack of consensus within the research and clinic communities (for a more detailed discussion on the issue see Reilly et al., 2014). In fact, the reader may have already noted that the diversity of terms applied in the clinic (Expressive or Mixed Receptive-Expressive Language Disorder in the DSM-4-TR; Expressive or Receptive Language Disorder in the ICD-10, Language Disorder in the DSM-5, and Developmental Language Disorder in the ICD-11) highly differ from the myriad of terms applied in the research field in the last decades (developmental aphasia, deviant language, delayed language, developmental language impairment, SLI or DLD, among others, -see Figure 1.1-). That was one of the main reasons why the CATALISE consortium (Bishop et al., 2016, 2017) was so welcomed by the stakeholders, because it brough the issue to light and managed to reach some consensus among professionals from different fields in some key points regarding DLD (e.g., the issue of terminology).

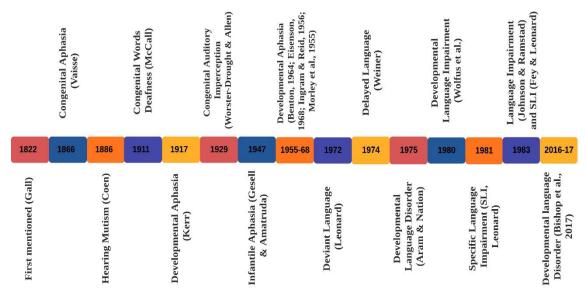


Figure 1.1. Evolution of the terminology on language disorders

*Note*. Adapted from "Specific language impairment: a convenient label for whom?" by S. Reilly et al., 2014, *International Journal of Language & Communication Disorders*, 49(4), 416–451. Copyright 2014 by John Wiley & Sons, Inc.

Hence, similar to what happened with the term NDDs, the introduction of the name DLD was not just a label substitution but it implied a reconceptualization of what had traditionally been considered language disorders. Language disorders comprise those language difficulties emerging at early ages but enduring into adulthood which have a serious impact on individuals' everyday life. The CATALISE consortium promoted a new approach that, instead of basing diagnostic decisions about language disorders on exclusionary criteria, proposes a three-way differentiation: (1) Differentiating conditions (if the language disorder is associated to a more global biomedical condition), (2) Risk factors, and (3) Comorbid conditions (Bishop et al., 2017; Paul, et al., 2020). Therefore, languages difficulties of those children who also present a broader disorder (named "differentiating condition") would be recognized with terms like "language disorder associated with X condition", while the term DLD would be applied to children previously diagnosed as having SLI plus those with language problems and low nonverbal IQ and those with language impairment and comorbid conditions such as ADHD or motor disorders.

In this sense, the conceptualization of DLD entails three main differences with respect to SLI in terms of scope: (1) The removal of the low non-verbal IQ as exclusionary criterium since research has substantially demonstrated that children show similar performance, course, and prognosis regardless of this ability (Bishop et al., 2016, 2017; Castro-Rebodello et al., 2004; Martos & Ayuda, 2004; McGregor et al., 2020; Norbury et al., 2016; Paul, 2020; Reilly et al., 2014); (2) The inclusion of children with comorbid conditions whose relation with the language difficulties is undetermined; (3) The stress on the impact of the language problems on the individual's daily functioning, instead of on the scores of standardized tests. In other words, this reconceptualization implies that language disorders will not be concealed by broader conditions or by the results of standardized tests in overstated and unrelated cognitive indexes.

Among the main advantages of embracing the term DLD, McGregor et al. (2020) highlighted: (1) Advocacy, despite being seven times more prevalent than ASD according to some studies, it is a fact that the authorities and the general public are not aware of the dimension of language disorders; (2) Research, it opens new possibilities for disentangling causality, specific phenotypes, and potential associations between cognitive abilities; and (3) Clinical practice, it better reflects the reality of practitioners

who deal with heterogeneous language and cognitive profiles whose common features are the presence of language difficulties and a serious impairment in daily life functioning. In line with that, Lancaster and Camarata (2019) demonstrated through cluster analyses that the high variability observed in DLD fits better with a continuum/spectrum model similar to that of ASD rather than with those models based on subtypes or individual differences. On the other hand, this new view of language disorders, specifically DLD, promoted by researchers and practitioners seems to have reached the main classification systems (e.g., the DSM-5 removed the criterium of low non-verbal IQ from the diagnosis of language disorders), which implies that clinical practice is currently immerse in a change process.

All evidence provided in this section comes to highlight the late changes in the conception and approach of NDDs, particularly those targeted in this doctoral dissertation. Following sections are aimed at underpinning the importance of diagnoses, highlighting the difficulties in distinguishing between ASD and DLD, and stressing the benefits of initiating intervention and treatment at early ages.

### **1.3 Difficulties on Differential Diagnosis in Clinical Practice**

As referred in Reilly et al. (2014, p. 417): "when clinicians diagnose diseases or disorders their intention is to assign an individual's symptoms to a particular category which is distinct from others and which is informative with respect to etiology, treatment and prognosis". In this sense, it is inevitable to mention that the validity and convenience of diagnostic labels has been called into question by some academics and professionals from the research, educational, and clinical community. Thus, while some claim that diagnoses can stigmatize children with NDDs and may be more damaging than beneficial (see Lauchlan & Boyle, 2007), others highlight the fact that the lack of diagnosis prevent children and families from receiving assistance (Reilly et al., 2014). Bishop (2014) summarizes very comprehensively the main pros and cons derived from the use of diagnostic labels, which have been synthesized in Table 1.3.

Drawbacks	Benefits
Stress on the child when addressing the disorder, disregarding the potential effect of the environment on it.	Reinforcement of the cause and validity of the construct.
Lack of responsibility of parents.	Lack of blame of parents.
Pygmalion effect: the child thinks that s/he is not able to improve, so s/he fails at every trial and, eventually, s/he does not even try.	Lack of blame of the child.
Avoidance of responsibility by educators and professionals related to the child.	Lack of blame of educators and other professionals involved in the life of the child.
Stigma and social alienation.	Advocacy, visibility, protection and support, sense of belonging, community involvement, and demystification.
Exclusion of those children who do not fit into specific criteria and/or abuse of diagnoses to increase funding.	Gateway to assistance and supplies.
Emphasis on the label instead of the particular difficulties of the child and trend toward generalizing, neglecting individual differences.	Identification and validation of shared features among children with identical handicaps.
Non-necessity of other intervention beyond that of well-trained educators.	Openness to individualized interventions, therapies, and treatments.
Misunderstanding provoked by the use of same diagnostic labels in different context and in different ways.	Bridge-building between professionals, improving the flow of information.
Questionable criteria in some cases, particularly those related to IQ.	Provision of unbiased and standard criteria for developmental evaluations.
Over-medicalization of conditions that do not need a medical approach and medical attribution for environmental difficulties.	Acknowledgement of the causal effect of biological as well as environmental factors.
Organization according to statistics, not to the actual difficulties and needs of children and families.	Usefulness of statistics to arrange resources and intervention, as well as for measure and register improvements.
Unnatural groups involved in studies with clinical samples, which hinders results generalization.	Utility for research in terms of favoring continuity and follow-up.

**Table 1.3.** Benefits and drawbacks of applying diagnostic labels

*Note*. Adapted from "Ten questions about terminology for children with unexplained language problems" by D. V. M. Bishop, 2014, *International Journal of Language & Communication Disorders*, 49(4), 381–415. Copyright 2014 by John Wiley & Sons, Inc.

After going through the main arguments in favor and against the use of diagnostic labels, nowadays it seems that most research agree that these labels entail more benefits than detriments (Baird, 2014; Bishop, 2014; Clark & Carter, 2014; Hansson et al., 2014; Reilly et al., 2014). From a practical point of view, receiving a diagnosis enables children

and families to access to resources, legal protection, and professional and financial assistance. From a personal perspective, it also allows not only to name a set of suspicions about atypical behaviors observed in the child, which is the basis for acceptance and usually provides a great relieve to families according to Hüneke & Lascelles (2014) and Karst & Vaughan van Hecke (2012), but also to have the opportunity to get involved in support groups and raise awareness and visibility inside the community.

Regarding the diagnosis of NDDs, there is a threefold difficulty: (1) Etiologies are still poorly understood, (2) There is a high heterogeneity within clinical infant populations, and (3) Developmental courses may vary but those variables involved in them are largely unknown (Reilly et al., 2014). Following the line of these authors, current tools for diagnosing NDDs are unspecific and can be classified into three groups depending on the condition:

Syndromic conditions with a known etiology and, hence, a biological diagnostic test (e.g., Williams and Fragile X syndrome); non-syndromic conditions with no known etiology, but which are diagnosed through objective testing (e.g., SLI, reading disorder); and non-syndromic conditions diagnosed through the use of subjective rating scales or clinical judgments (e.g., ADHD, autistic spectrum conditions). (Reilly et al., 2014, p. 418)

According to this classification, both disorders involved in this doctoral dissertation belong to non-syndromic conditions but they differ in the kind of tools used during diagnosis (objective tests for DLD and subjective ones for ASD). This may partially explain the imbalance of diagnoses of each disorder; there are much more diagnoses of ASD than DLD, despite the fact that DLD is seven times more prevalent than ASD according to some studies (for a thorough discussion see our work on diagnostic migration, Appendix 2).

In addition to difficulties associated to general diagnostic process in NDDs, the distinction between ASD and DLD at early ages is even more complex as most cases are far from textbook descriptions and present very similar cognitive and behavioral markers (Bishop 2000, 2010, 2014; Taylor & Whitehouse, 2016). For example, traditionally it has been considered that ASD and DLD showed distinct language deficits; however, recent

evidence has refuted that statement since several studies have found analogous linguistic profiles (Bishop, 2000; Durrleman & Delage, 2016; Modyanova et al., 2017; Ramírez-Santana et al., 2019; Wittke et al., 2017). This has led to question a possible shared etiology, which would be translated into a unique continuum of impairment in which we could find ASD at one end of the spectrum and DLD at the other one (Bishop, 2000, 2003, 2010; Bishop & Norbury, 2002; Conti-Ramsden et al., 2006; Georgiou & Spanoudis, 2021; Leyfer et al., 2008; Taylor & Whitehouse, 2016; Tomblin, 2011). The high variability of linguistic profiles found in both disorders impairs inevitably differential diagnosis at early ages (Durrleman & Delage, 2016; Kjelgaard & Tager-Flusberg, 2001; Taylor & Whitehouse, 2016; Williams et al., 2008). That is the reason why many researchers and practitioners have argued in favor of more research comparing disorders in order to elucidate differences in clinical profiles and developmental trajectories, consequently contributing to improving differential diagnosis (Bishop et al., 2016; Rice, 2016; Weismer, 2013). This issue will be discussed more thoroughly in the following chapters; however, so far, the reader must be aware of the similarities between disorders and reflect on how this fact may impact on differential diagnosis blurring the profiles and hampering accurate early diagnoses.

# 1.4 The Relevance of Early Childhood Intervention

Once stablished the benefits of early diagnosis and the obstacles for differential diagnosis between ASD and DLD, it is convenient to highlight the link between diagnosis and intervention. As mentioned in the previous section, one of the main advantages of diagnosis is that it provides access to professional support and resources. Research as well as practice have substantially proved the benefits of the first provisional diagnoses assigned to children in the initial stages of early childhood intervention (ECI), since they have been related to early-onset interventions which drove to better prognosis (Bennett et al., 2014; Bishop, 2000; Charman & Baird, 2002; Eadie et al., 2014; Germain et al., 2015; Mandell et al., 2005; Martos & Ayuda, 2004; Nicholas et al., 2009; Özerk, 2017; Reilly et al., 2014; Zablotsky et al., 2017).

ECI was boosted between the late 1960s and the 1970s when the focus was put on working with infant and child population with detected difficulties or at-risk of having any developmental condition (Dunst, 2018b). Since then, research and practice in ECI have grown exponentially. Current knowledge on NDDs and best practice has led to

radically shift the approach in ECI towards the implementation of evidence-based practices (EBP). Dunst (2018b) operationalized EBP as those practices whose components are endorsed by sound results coming from other interventions and by research based on practice and replicated systematically. In 2014, the Division for Early Childhood (DEC) released a set of recommended practices for practitioners arranged in eight broader categories: (1) Assessment, (2) Environment, (3) Family, (4) Instruction, (5) Interaction, (6) Leadership, (7) Teaming and collaboration, and (8) Transition practices. From these recommended practices, a set of performance checklists were developed with the aim of systematizing the format and the addressing of the mentioned eight categories and supporting the practice indicators with findings from research based on practice (Dunst, 2018a). Thus, the trend in ECI nowadays involves the early identification of the disorder, the assessment of the particular strengths and difficulties of the child, the design of individualized programs based on evidence from practice and research, and the involvement of the family and the community in the intervention process.

#### **1.5 Conclusions**

In this chapter, we have put into context the current approach towards NDDs. Thus, we have highlighted that evidence suggests that ASD and DLD are better explained as spectra, instead of tight and discrete categories, due to the heterogeneity of both profiles. Regarding the ongoing controversy about the potential shared etiology of ASD and DLD, we are going to try to disentangle some key issues that may help shed light into the phenotypic definition of both profiles in the following chapters.

Moreover, we have discussed the benefits and drawbacks of using diagnostic labels and we have referred to the main contributions of research to support the valuable effort of early diagnosis and ECI to improve the wellbeing of children with NDDs and the quality of life of all the stakeholders. Thus, the validity and utility of differential diagnosis and early interventions in NDDs like ASD and DLD have been evidenced. Keeping these ideas in mind, the reader is now able to further the comparative analysis between children with ASD and those with DLD in order to solve some questions:

 How are the specific profiles of children with ASD and those with DLD at early ages?

- 2) Is there any specific area that may contribute to establish differences between these disorders?
- 3) Is there enough evidence to support the existence of a unique continuum between both disorders or is there a clear distinction between them?
- 4) Can new technologies and methodologies help in early differential diagnosis? If the answer is affirmative, which ones and in what ways may they help?

# Chapter 2. Emotional Competence in Children with Autism Spectrum Disorders (ASD) and Specific Language Impairment (SLI). A Comparative Research Review

#### 2.1 Preamble

This chapter includes the first review that we performed in order to be acquainted with the characterization of children with ASD and DLD (in that time more commonly referred as SLI) in terms of emotional competence (EC). As the reader will note, this chapter and the following one contain the term SLI instead of DLD; that is because by the time when both literature searches were conducted there was not enough content on DLD as the term had not been sufficiently widespread yet. On the other hand, it is also important to state that we focused on EC after realizing that cognitive profiles did not yield sufficient significant differences between profiles to establish a differential diagnosis; thus, we decided to explore different developmental areas in search of early distinctive traits. In this line, we considered that it was essential to start this doctoral dissertation by digging into these abilities of our target populations with the purpose of describing them and contextualize our future experiments.

To do so, we performed a thorough review in the most commonly used databases, and we found out such an amount of literature that we resolved to divide the report of results into two different papers. On the one hand, which is the subject of this chapter, we presented results of studies applying mostly behavioral measures to assess the different abilities comprised in the construct of EC. On the other hand, which is the object of the next chapter, we put forward the outcomes of studies using primarily physiological measures, specifically eye tracking methodology. Thus, in this chapter the reader is going to find a comprehensive review of the impaired and conserved abilities related to EC in children with ASD and those with DLD. The aim of this chapter is, therefore, to have an overview of (1) How research has addressed this issue in both populations; (2) How children with ASD and those with DLD identify, understand, express, and regulate Early Differential Diagnosis in Infant Population with Neurodevelopmental Disorders. Psychophysiological Markers Based on Eye Tracking Methodology emotions; and (3) Which abilities they share, and which can be useful for distinguishing between disorders.

Vacas, J., Antolí, A., Sánchez-Raya, A., & Pérez-Dueñas, C. (2021). Emotional Competence in Children with Autism Spectrum Disorders and Specific Language Impairment: A Comparative Research Review. *Education and Training in Autism and Developmental Disabilities,* 56(3), 306-327. https://www.proquest.com/scholarly-journals/emotional-competence-childrenwith-autism/docview/2570258568/se-2?accountid=14520.

#### Abstract

Emotional competence (EC) refers to a set of skills to identify, understand, and respond to one's own emotions and those of others. It plays a fundamental role in socialization processes, where children with autism spectrum disorders (ASD) and specific language impairment (SLI) show marked deficits. However, due to the similarities between these two conditions, it is difficult to establish a differential diagnosis at early ages. A literature search of the WOS, SCOPUS, and Proquest databases was conducted. Year of publication (from 2000 to 2019), type of paper (reviews, meta-analyses, and experimental studies), language (English), and age of participants (children up to 13 years) were applied as inclusion criteria. Based upon these criteria, 34 papers were included in this review. Results revealed that children with ASD showed impairments in facial emotion recognition, whereas those with SLI showed difficulties in emotion recognition in auditory and audiovisual modalities. Results also highlighted that both children with ASD and SLI showed biases towards the recognition of positive emotions.

**Keywords:** Emotional competence, childhood, autism spectrum disorders, specific language impairment, differential analysis.

#### 2.2 Introduction

Autism spectrum disorders (ASD) and specific language impairment (SLI) have traditionally been regarded as distinct conditions given that the compromised language areas were often well-differentiated: Whereas pragmatic language function was impaired in ASD, structural language functions (e.g., phonology, morphology, or syntax) were compromised in SLI. However, given the significant heterogeneity of both populations, ample evidence has suggested a potential overlap between disorders, either because of Chapter 2. Emot ional Competence in Children wi th Autism Spectrum Disorders (ASD) and Specific Language Impairment (SLI). A Comparative Research Review the greater functional language disorders in individuals with ASD that resemble SLI or the marked social difficulties in individuals with SLI that resemble ASD traits (Taylor et al., 2014, 2015). Furthermore, according to the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association [APA], 2013) the high frequency with which ASD and SLI co-occur indicates a high comorbidity between conditions, which makes it difficult to differentiate them.

There are two main lines to explain the co-occurrence of ASD and SLI. Some evidence supports a shared etiology, while a second theory holds that similarities between disorders are superficial and cannot support the unification of diagnoses (for a review, see Taylor et al., 2012). Given its impact on clinical practice, more research is needed to obtain substantive evidence of the relationship and differences between conditions (Taylor et al., 2012, 2014, 2015).

#### **2.3 Emotional Competence**

Traditionally, studies that involved these clinical populations have focused on their language ability and, to a lesser extent, their cognitive profile, while the socialemotional domain has received less attention. However, this trend seems to be changing considering the increasing number of papers addressing the study of emotion (Jeanneret et al., 2015). According to the bibliometric study of Jeanneret et al. (2015), for example, there has been an exponential growth in studies on facial emotion recognition (FER) in recent years. The number of collaborations and indexed publications on this subject has also grown, which indicates that the study of the different domains of emotional competence (EC) is gaining relevance.

There is consensus regarding the complexity and multidimensionality of EC and the many skills involved in it. EC comprises the processes of identifying, understanding, expressing, and regulating emotions both within oneself and toward others (Beck et al., 2012; Denham, 1998). Saarni (1999) described eight basic skills that constituted the EC construct: (1) Awareness of one's own emotions; (2) Ability to recognize others' emotions; (3) Ability to use the vocabulary of emotion and expressions; (4) Ability to empathize with others' emotional experiences; (5) Ability to differentiate internal, subjective emotional experience from external, emotional expression; (6) Capacity for adaptive coping with aversive emotions and distressing circumstances; (7) Awareness of emotional communication within relationships; and (8) Capacity for emotional selfEarly Differential Diagnosis in Infant Population with Neurodevelopmental Disorders. Psychophysiological Markers Based on Eye Tracking Methodology efficacy. This classification has been widely adopted in recent research, which acknowledges that EC cannot be separated from individuals' social functioning, as its purpose is to achieve efficient management and performance in social situations (Beck et al., 2012; Begeer et al., 2008; Lau & Wu, 2012).

According to the developmental model of emotion understanding, EC is acquired in different phases: (1) Between the ages of 3 and 4, children learn external aspects such as FER, the impact of the situation on emotions, and the role of desires in emotions; (2) Between the ages of 6 and 7, children develop mental aspects such as the effect of beliefs and memory on emotions and distinguish between the outward expression of emotions and experienced emotions; (3) At the age of 8 to 9, reflective aspects such as the importance of morality, the awareness of emotion regulation through cognitive strategies, and the understanding of mixed feelings are learned (Pons et al., 2004, as cited in Ornaghi & Grazzani, 2013). Regarding the developmental trajectory of emotion recognition (ER), it has been found that happiness, sadness, and anger are the first emotions to be developed and distinguished. Thus, they undergo the least changes over the years, while the ability to recognize fear, disgust, and surprise (the latter to a lesser extent) is improved throughout childhood until adolescence (Lawrence et al., 2015).

Given the importance of EC in development, this review aimed to provide a stateof-the-art understanding of this competence in child populations with ASD and SLI. The main aim was to determine whether there are differences between these neurodevelopment disorders in any domain of EC to facilitate their diagnosis and guide interventions. For that purpose, the following questions were raised:

1. Which EC domains have been addressed in each population?

2. To date, what is it known about EC in each condition?

3. Which are the main similarities and differences between ASD and SLI in terms of EC?

# 2.4.1 Search Descriptors

The initial search was conducted in November 2018 and updated until December 2019 on the Web of Science (WoS), Scopus, and Proquest databases using the following descriptors: ('emotional competence' OR 'emotional profile' OR 'emotion') AND ('autism spectrum disorders') AND ('specific language impairment').

# 2.4.2 Study Selection

Given the developmental nature of EC and the fact that neurodevelopmental disorders are a set of conditions that emerge in the earliest stages of development and seriously impair individuals' personal, social, academic, or occupational functioning (APA, 2013), our bibliographic search focused on studies with child samples.

We reviewed the publications resulted from the search according to the following inclusion criteria: Year of publication (from 2000 to 2019), type of paper (reviews, metaanalyses, and experimental studies), language (only English papers), and age of the study participants (children up to 13 years of age).

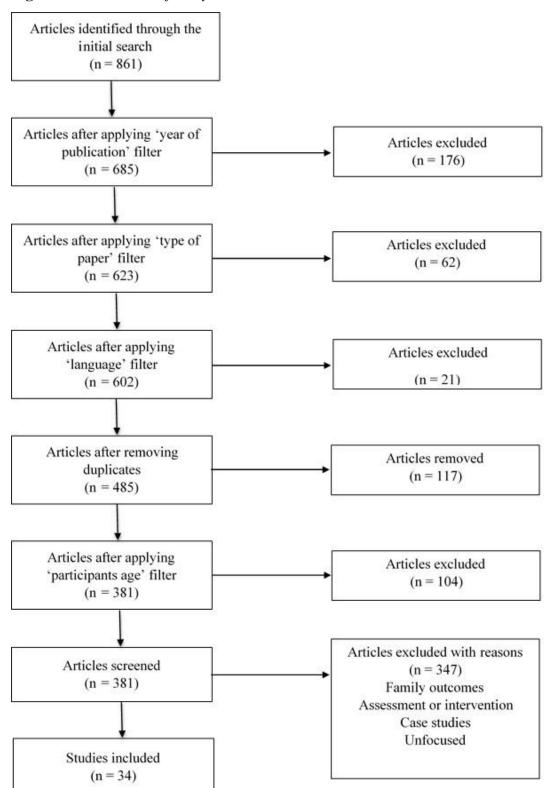


Figure 2.1. Flowchart of study selection

Chapter 2. Emot ional Competence in Children wi th Autism Spectrum Disorders (ASD) and Specific Language Impairment (SLI). A Comparative Research Review

#### 2.5 Results

After applying inclusion criteria and excluding duplicates, 381 articles were screened (Figure 2.1). Thirty-four articles were selected as they fitted the purpose of the review, the remaining 347 papers were ruled out after the screening process for being focused on assessment tools, interventions, case studies, relatives or parental outcomes, or being unrelated studies (considering ASD and SLI as comparison groups or EC as secondary measure). Five papers were reviews, two meta-analyses, and 27 experimental studies. Likewise, 20 articles referred to children with ASD, 10 to population with SLI, and four compared conditions. A total of 1,971 participants were included in these studies (482 with ASD, 427 with SLI, 962 neurotypical controls, and 100 with other conditions) with an age range from 1 to 13 years. Most studies were focused on the emotion identification domain (also called ER in literature, more specifically they were focused on FER, and emotion perception), while a few addressed the emotion understanding (e.g., emotion inference, empathy, and Theory of Mind [ToM]), expression (e.g., conscious and unconscious response to emotional stimuli), and regulation domains. One review focused on the four domains and some papers addressed other emotional aspects such as general social-emotional functioning and behavioral, emotional, and social difficulties (BESD). Finally, most works applied identification, labeling, and matching tasks, whereas others used open questions, observation tasks, imitation tasks, questionnaires, and standardized tests. Results from these articles are presented in the following according to the EC domain addressed and the target population included in each case.

# 2.5.1 Emotion Identification

# 2.5.1.1 Emotion Identification in Children with ASD

Results involving population with ASD are summarized in Table 2.1. There is consensus that children with ASD show poorer FER than their typically developing (TD) peers. However, the specific findings on FER were mixed (for a review, see Harms et al., 2010). Some studies argued that children with ASD have global difficulties in FER. Thus, the meta-analysis of Lozier et al. (2014) concluded that individuals with ASD of all ages showed global FER deficits whose severity varies between emotions, with the poorest recognition rates for anger, fear, and surprise. Individuals with ASD also demonstrated an atypical developmental recognition pattern in which they did not improve their ability

to recognize emotions over time. Some data supported the thesis that children with ASD present a general difficulty to recognize low-intensity, negative emotions (Evers et al., 2015; Uljarevic & Hamilton, 2013), although deficits in FER have also been found in these children in medium and high expression intensities (Griffiths et al., 2019). Similarly, a greater deficit has been identified for the recognition of surprise and anger in a sample of children with low-functioning ASD compared to a control group. Greater difficulties in matching tasks (pairing faces with the same emotional expression) than in tasks using emotional cues provided in writing or verbally have also been found in population with ASD (Golan et al., 2018). These authors suggested that this population may have a disadvantage in generalizing emotional expressions in different faces and could potentially benefit from verbal support. Interestingly, FER deficits in population with ASD have been found exclusively for human faces but not for canine ones, which may suggest that emotion identification is not completely impaired in this population, but it is related to social stimuli (Davidson et al., 2019). According to Lacroix et al. (2016), FER deficits in children with ASD may be associated to poor visual strategies and problems with pragmatics and lexicon (mainly in relation to emotional vocabulary).

On the other hand, two reviews pointed to the lack of deficits in recognition of basic emotions under simple and prototypical situations but identified difficulties regarding FER of complex emotions and those displayed subtly or incongruently as well as in other aspects such as emotional expressions processing in children with ASD (Beeger et al., 2008; Harms et al., 2010). These papers also highlighted the wide variability within the disorder and the methodologies applied. Likewise, Tell and Davidson (2015) reported that children with ASD performed similarly to their peers in FER when facial emotional expressions coincided with the context in which they were framed; otherwise, when facial emotional expressions were presented in incongruent contexts, children with ASD performed differently from their peers, relying more on facial cues than on contextual ones.

Finally, some studies have reported the absence of differences in FER between children with ASD and their peers (Lacroix et al., 2009, 2014). Thus, similar rates of accuracy between ASD and TD groups in tasks to label, match, and identify basic emotions were found in Lacroix et al. (2009), and a common recognition pattern between groups was yielded in Lacroix et al. (2014). This pattern consisted of more difficulties in

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and Specific Language Impairment (SLI). A Comparative Research Review identifying expressions of surprise and performing in labeling tasks in both groups. Similarly, Farran et al. (2011) did not find differences between children with ASD and their peers in accuracy in an emotion identification task; they also reported a similar pattern in which happy faces were detected more quickly, while fear, anger, and sad expressions were processed more slowly. In this study greater accuracy was also found in all groups for the identification of happiness and disgust, while recognition of fear and anger was less accurate. Similarly, Schwenck et al. (2012) reported comparable levels of accuracy in the ASD and TD groups for angry, happy, fear, and disgusted faces. Li et al. (2011) also reported similar FER performance in toddlers with ASD and their TD peers after analyzing the first-look time towards emotional faces in both groups. This measure has been proved to be sensitive to stimulus differences; thus, in this study both toddlers with and without ASD gazed longer at happy and angry faces than at fearful ones.

Finally, children with ASD have also shown deficits in identification of basic and complex emotions from body language and gestures when removing facial cues, which implies that their ER difficulties are not circumscribed to facial expressions but are more pervasive (Metcalfe et al., 2019).

Table 2.1. Summary of conserved and impaired EC domains in children with ASI	Table 2.1. Summary of	f conserved and	impaired EC	' domains in	children with ASL
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Study	Main Features
Begeer et al. (2008)	Systematic review.
	EC domains assessed: Emotion expression, perception, response, and understanding.
	<u>Conserved skills</u> : 1) Appropriate expression, understanding and response for basic emotions; 2) Adequate attention and perception of facial expressions.
	<u>Impaired skills</u> : Large variability in complex skills (greater problems in ASD + intellectual disability, late acquisition of complex skills in high-functioning ASD).
Ben Shalom et al. (2006)	<u>Groups</u> : High functioning ASD/Asperger syndrome ( $n = 10$ ; Mage = 13.0, SD = 2.5) and TD ( $n = 10$ ; Mage = 11.8, SD = 3.4).
	EC domains assessed: Physiological reaction (skin conductance) and conscious response (self-report).
	Tasks: Observation task and self-report of emotions.
	Conserved skills: Adequate autonomic response to pleasant and unpleasant stimuli.
	Impaired skills: Deficits in the cognitive dimension of emotion.
Cibralic et	Systematic review.
al. (2019)	EC domains assessed: Emotion regulation.
	Conserved skills: None.
	<u>Impaired skills</u> : 1) Poorer emotion regulation abilities (use of simpler and less efficient strategies); 2) More reliance on others to regulate their emotions.

5 1 5	forogrouf Murkers Bused on Eye Hueking Methodology
Davidson et al.	<u>Groups</u> : High functioning ASD ( $n = 23$ ; Mage = 11.10, SD = 1.1) and TD ( $n = 23$ ; Mage = 11.06, SD = 2).
(2019)	EC domains assessed: Emotion identification (FER).
	<u>Tasks</u> : Labeling task (choosing emotion labels for both human and canine faces from a forced-choice list).
	Conserved skills: Adequate performance in FER for canine faces.
	Impaired skills: Poorer performance in FER for human faces.
Evers et al.	<u>Groups</u> : ASD (n = 45; Mage = 10.6, SD = 2.3) and TD (n = 50; Mage = 10.3, SD = 2.4).
(2015)	EC domains assessed: Emotion identification (FER).
	Tasks: Labeling task (naming emotions observed in visual dynamic stimuli).
	<u>Conserved skills</u> : 1) Adequate performance in FER for happiness and anger; 2) Better performance in fear recognition.
	Impaired skills: Poorer performance in FER for sadness, surprise, and disgust.
Farran et al. (2011)	<u>Groups</u> : High functioning ASD/ Asperger syndrome ( $n = 20$ ; Mage = 12.3, SD = 1.6), TD matched on age ( $n = 20$ ; Mage = 12.3, SD = 1.5) and TD matched on verbal and non-verbal ability ( $n = 20$ ; Mage = 10.9, SD = 1.6).
	EC domains assessed: Emotion identification (FER).
	Tasks: Identification task.
	<u>Conserved skills</u> : 1) Adequate FER accuracy in basic emotions; 2) Typical FER patterns faster discrimination of happy faces and slower discrimination of frightened, angry, or sac faces; 3) Greater accuracy in identifying happiness and disgust, and less accuracy in identifying fear and anger.
	Impaired skills: Longer response time in FER for negative emotions (fear, anger, sadness)
Glaser & Shaw (2011)	<u>Groups</u> : ASD ( $n = 19$ ; Mage = 9.5, SD = 3.8) and 22q13 deletion syndrome ( $n = 18$ ; Mage = 12.6, SD = 3.2).
	EC domains assessed: Emotion regulation.
	Tasks: Temperament and Atypical Behavior Scale (TABS).
	Conserved skills: Adequate developmental and daily life skills.
	<u>Impaired skills</u> : Severe difficulties in emotion regulation and undeveloped social emotional skills.
Golan et al. (2018)	<u>Groups</u> : Low functioning ASD ( $n = 29$ ; Mage = 9.1, SD = 1.2) and TD matched on verba mental age ( $n = 34$ ; Mage = 4.0, SD = 1.1).
	EC domains assessed: Emotion identification (FER).
	Tasks: Matching task (pairing emotions from facial, vocal and verbal cues).
	Conserved skills: None.
	<u>Impaired skills</u> : 1) Deficit in the identification of surprise and anger; 2) More difficulty in matching tasks than in tasks where emotional cues are provided by the written or spoker word (although all domains are affected).
Griffiths et al. (2019)	<u>Groups</u> : High functioning ASD ( $n = 66$ ; Mage = 11.24, SD = 2.91) and TD matched or age ( $n = 70$ ; Mage = 11.24, SD = 2.49).
	EC domains assessed: Emotion identification (FER).
	<u>Tasks</u> : Labeling task (choosing a label from a forced-choice list to identify basic emotions at different intensities).
	<u>Conserved skills</u> : Adequate FER performance in low intensity expressions due to floor effect.

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	<u>Impaired skills</u> : Deficits in FER for all basic emotions in medium and high intensity expressions.
Harms et	Systematic review.
al. (2010)	EC domains assessed: Emotion identification (FER).
	<u>Conserved skills</u> : Ability to recognize basic emotions in normal situations and with prototypical facial expressions in high-functioning ASD.
	<u>Impaired skills</u> : Deficit in FER (only for complex emotions/situations) and in processing of emotional expressions.
Lacroix et al. (2009)	<u>Groups</u> : ASD (n = 12; Mage = 6.1, SD = 1.2), Williams syndrome (n = 12; Mage = 8.1, SD = 2.4), and TD matched on verbal mental age (n = 12; Mage = 5.7, SD = 1.1).
	EC domains assessed: Emotion identification (FER).
	<u>Tasks</u> : Facial discrimination task (identifying gender), facial movement task (identifying facial movement), and FER tasks (labeling, matching and identification emotional expressions).
	<u>Conserved skills</u> : Adequate performance in tasks to label, match, and identify basic emotions.
	Impaired skills: None.
Lacroix et al. (2014)	<u>Groups</u> : ASD (n = 22; Mage = 6.2, SD = 1.3), TD matched on chronological age (n = 22; Mage = 6.7, SD = 1.3) and TD matched on mental age (n = 20; Mage = 5.5, SD = 1.3).
	EC domains assessed: Emotion identification (FER).
	Tasks: Labeling, matching, and identification tasks (naming and pairing emotions).
	<u>Conserved skills</u> : Typical FER pattern for basic emotions: greater difficulties in recognizing expressions of surprise and performing in labeling tasks.
	Impaired skills: None.
Lacroix et	Review.
al. (2016)	Groups: ASD and Williams syndrome.
	EC domains assessed: Emotion identification (FER).
	Conserved skills: None.
	<u>Impaired skills</u> : 1) Global FER deficit when language is compromised; 2) Poor visual strategies; 3) Difficulties with pragmatics and lexicon.
Li et al. (2011)	<u>Groups</u> : ASD ( $n = 37$ ; Mage = 2.8, SD = 0.45) and TD ( $n = 40$ ; Mage = 1.8, SD = 0.22).
	<u>EC domains assessed</u> : Emotion identification (FER and emotion perception -visual attention in terms of time spent on looking at faces) and emotion expression (emotional response in terms of facial expression display after viewing each stimulus and behavioral response in terms of joint attention to emotional faces).
	Tasks: Observation task.
	<u>Conserved skills</u> : 1) Adequate discrimination and recognition of emotional expressions; 2) Adequate levels of response to positive emotions.
	Impaired skills: Less social and joint attention, and emotional reaction to negative emotions.
Lozier et	Meta-analysis.
al. (2014)	EC domains assessed: Emotion identification (FER).
	Conserved skills: None.
	<u>Impaired skills</u> : 1) Global deficit in FER (mainly for fear, anger, and surprise); 2) Inverse developmental pattern, poorer performance over the years (mainly in the recognition of fear, sadness, disgust, and happiness to a lesser extent).

Metcalfe et al. (2019)	<u>Groups</u> : ASD ( $n = 27$ ; Mage = 10.9, SD = 3) and TD matched on gender and age ( $n = 27$ ; Mage = 10.9 SD = 3).
	EC domains assessed: Emotion identification (ER from body language and gestures).
	Tasks: Naming task (inferring emotions from body language and gestures, no facial cues).
	Conserved skills: Benefits from contextual cues to the same extent than controls.
	Impaired skills: Deficits in recognition of all basic emotions from body movements.
Schwenck et al.	<u>Groups</u> : ASD (n = 55; Mage = 11.9, SD = 2.8), CD-CU+ (n = 36; Mage = 12.7, SD = 2.5), CD-CU- (n = 34; Mage = 11.9, SD = 2), and TD (n = 67; Mage = 12.0, SD = 2.7).
(2012)	EC domains assessed: Emotion identification (FER), empathy.
	<u>Tasks</u> : Naming task (identifying emotions in morphing stimuli), perspective taking task (description of dynamic scenes), and affection induction task (guessing emotions of different characters after watching short films and rating participants' own affection).
	Conserved skills: 1) Adequate FER performance; 2) Adequate level of empathy.
	<u>Impaired skills</u> : 1) Slower response time for sad faces; 2) Deficit in cognitive dimension of empathy (taking others' perspective).
Tell &	<u>Groups</u> : ASD (n = 22; Mage = 10.14) and TD (n = 22; Mage = 9.8).
Davidson (2015)	EC domains assessed: Emotion identification (FER).
(2013)	<u>Tasks</u> : Naming task (identifying emotions in facial expressions in congruent and incongruent contexts).
	Conserved skills: Adequate performance in emotional expressions congruent with context.
	Impaired skills: Deficit in processing of emotional expressions in incongruent contexts.
Uljarevic	Meta-analysis.
& Hamilton	EC domains assessed: Emotion identification (FER).
(2013)	Conserved skills: Adequate FER for happiness.
	Impaired skills: Deficits in FER (poorer recognition of negative emotions).
Zantinge et al. (2019)	<u>Groups</u> : ASD ( $n = 21$ ; $M_{age} = 5$ , $SD = 0.78$ ) and TD matched on age ( $n = 45$ ; $M_{age} = 4.67$ , $SD = 1$ ).
	EC domains assessed: Emotion expression.
	Tasks: Observation task.
	<u>Conserved skills</u> : Adequate autonomic response and facial expression of negative emotions when facing fearful stimuli.
	Impaired skills: Low concordance between emotion experience and expression.

Mean age is expressed in years. Conversion was done when it was expressed in months TD, Typically Developing; FER, Facial Emotion Recognition; CD-CU+, Conduct Disorder with high traits of antisocial behavior; CD-CU-, Conduct Disorder with low traits of antisocial behavior

# 2.5.1.2 Emotion Identification in Children with SLI

Results involving population with SLI are summarized in Table 2.2. Six out of the ten articles found in this population were focused on FER. Only one of these studies reported poorer performance in recognition, labeling, and inference of basic emotions (except for happiness) in primary-school children with SLI with respect to their peers (Bakopoulou & Dockrell, 2016).

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On the other hand, four studies pointed to an adequate performance in ER when facial expressions were presented only in the visual modality (Creusere et al., 2004; Czaplewska & Sterczynski, 2015; Delaunay-El Allam et al., 2011; Ford & Milosky, 2003). Czaplewska and Sterczynski (2015) found an identical FER pattern between the SLI and control groups as demonstrated by similar recognition rates and increasing competence throughout childhood (developmental trajectory). However, less accuracy was found in labeling emotion tasks, as well as greater confusion between sadness and anger in individuals with SLI in middle childhood (Delaunay-El Allam et al., 2011). This last finding was particularly noteworthy, as it is common for very young TD children to confound these emotions. They do not become aware of the difference between these emotions until the age of 5–6 and only correctly distinguish them in middle childhood. This could imply that children with SLI show a slight delay in the development of emotional concepts. A delay in the maturation of FER skills has also been found in this population, although both SLI and TD groups showed a greater bias for recognizing positive rather than negative emotions (Merkenschlager et al., 2012).

Finally, difficulties have been reported in ER in auditory (Czaplewska & Sterczynski, 2015) and audiovisual modalities (Creusere et al., 2004) in children with SLI. The first authors reported differences between SLI and TD groups when presenting incongruent semantic and prosodic content. Children with SLI relied more on prosodic cues (intonation), while TD children relied more on semantic cues (content). Creusere et al. (2004) observed differences between SLI and TD groups in an audiovisual task that included facial expression and unfiltered speech (speech containing prosodic elements and semantic content). This suggested that children with SLI did not benefit from messages containing simultaneously verbal and visual content, as one kind of information impaired the perception of the other.

Study	Main Features
Bakopoulou & Dockrell (2016)	<u>Groups</u> : SLI ( $n = 42$ ; $M_{age} = 7.9$ , $SD = 1.7$ ), TD matched on cognitive ability ( $n = 42$ ; $M_{age} = 7.9$ , $SD = 1.8$ ) and TD matched on language ability ( $n = 42$ ; $M_{age} = 5.7$ , $SD = 0.5$ ).
	EC domains assessed: Emotion identification (FER).
	<u>Tasks</u> : Identification task, labeling task, inferring cause of emotion task, and conflict resolution strategies task.
	<u>Conserved skills</u> : Adequate performance in recognition, labeling, and inference of happiness.
	Impaired skills: Deficit in recognition, labeling, and inference of other basic emotions.
Brinton et al.	<u>Groups</u> : SLI $(n = 19; M_{age} = 9.1, SD = 1)$ and TD $(n = 19; M_{age} = 9.1, SD = 1)$ .
(2007)	<u>EC domains assessed</u> : Emotion understanding (emotion inference) and response to emotional stimuli (emotional response and emotion dissemblance).
	<u>Tasks</u> : Listening task (short stories about a character in different emotional situations) and open question task (questions about the understanding of the situation, emotion inference, emotion dissemblance, and understanding of the social-emotional rule).
	<u>Conserved skills</u> : Adequate understanding of the social desirability of an emotional response.
	Impaired skills: Different social objectives when responding emotionally.
Creusere et al.	<u>Groups</u> : SLI ( $n = 26$ ; $M_{age} = 5.1$ ) and TD ( $n = 26$ ; $M_{age} = 5.1$ ).
(2004)	EC domains assessed: Emotion identification (FER and ER from auditory and audiovisual cues).
	<u>Tasks</u> : Identification task (dynamic stimuli presented via visual, auditory -filtered speech, only prosodic elements-, and audiovisual modality -filtered and non-filtered speech-; forced-choice paradigm).
	<u>Conserved skills</u> : Adequate performance in FER, ER from intonation cues, and ER from audiovisual cues when speech was filtered.
	<u>Impaired skills</u> : 1) Poor performance in audiovisual tasks when speech was non- filtered; 2) Deficit in interpreting audiovisual information with verbal content; 3) Difficulty in integrating visual and verbal cues simultaneously.
Czaplewska & Sterczynski (2015)	<u>Groups</u> : SLI $(n = 76; Age \ range = 4.0-6.11)$ and TD $(n = 131; Age \ range = 4.0-6.11)$ .
	EC domains assessed: Emotion identification (FER and ER from auditory cues).
	<u>Tasks</u> : Identification task (from visual and auditory modality, in congruent and incongruent conditions).
	<u>Conserved skills</u> : 1) Adequate performance in FER; 2) Adequate performance in ER from intonation cues.
	<u>Impaired skills</u> : Atypical pattern for incongruent messages through auditory modality (more attention to intonation rather than content).
Delaunay-El	<u>Groups</u> : SLI ( $n = 12$ ; Mage = 8.0, SD = 1.7) and TD ( $n = 12$ ; Mage = 8.1, SD = 1.8).
Allam et al. (2011)	EC domains assessed: Emotion identification (FER) and emotion vocabulary.
(=011)	Tasks: Face-matching task and free labeling task (naming emotions without choices).
	Conserved skills: Adequate performance in FER.

**Table 2.2.** Summary of conserved and impaired EC domains in children with SLI

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	<u>Impaired skills</u> : Less precision in the use of emotional labels and greater confusion between sadness and anger.
Ford & Milosky	<u>Groups</u> : SLI ( $n = 12$ ; Mage = 5.9, SD = 0.5) and TD ( $n = 12$ ; Mage = 5.8, SD = 0.4).
(2003)	EC domains assessed: Emotion identification (FER) and emotion understanding (emotion inference).
	<u>Tasks</u> : Labeling task (naming emotions of characters of pictures and inferring emotions of characters of stories told via visual, auditory, and audiovisual modality).
	<u>Conserved skills</u> : 1) Adequate performance in FER; 2) Adequate performance in emotion understanding.
	Impaired skills: Deficit in inferring emotions.
Fujiki et al. (2004)	<u>Groups</u> : SLI young group (n = 21; Mage = 7.1, SD = 0.9), SLI old group (n = 22; Mage = 10.7, SD = 0.8), TD young group (n = 21; Mage = 7.2, SD = 1.0), and TD old group (n = 22; Mage = 10.7, SD = 0.8).
	EC domains assessed: Emotion regulation.
	Tasks: Questionnaire (Emotion Regulation Checklist - ERC-).
	Conserved skills: None.
	Impaired skills: 1) Poor performance in emotion regulation; 2) Social reticence behavior.
Merkenschlager	<u>Groups</u> : SLI ( $n = 24$ ; Age range = 7-11) and TD ( $n = 40$ ; Age range = 7-11).
et al. (2012)	EC domains assessed: Emotion identification (FER).
	<u>Tasks</u> : Identity recognition task and emotion identification task (forced-choice paradigm).
	Conserved skills: Bias for better recognition of positive emotions.
	Impaired skills: Delay in maturation of FER skills.
St Clair et al. (2011)	<u>Groups</u> : Longitudinal study. SLI group assessed at age 7 ( $n = 234$ ), at 8 ( $n = 203$ ), at 11 ( $n = 167$ ), and at 16 ( $n = 103$ ).
	EC domains assessed: Behavioral, emotional, and social difficulties (BESD).
	<u>Tasks</u> : Questionnaire: The Strengths and Difficulties Questionnaire (SDQ) teacher version.
	Conserved skills: None.
	Impaired skills: Emotional difficulties deceasing by adolescence.
Vissers &	Groups: Review.
Koolen (2016)	<u>EC domains assessed</u> : Social-emotional functioning, emotion identification, and emotion understanding (ToM, cognitive aspects such as imitation, joint attention, and false believe understanding).
	Tasks: None.
	Conserved skills: None.
	<u>Impaired skills</u> : 1) Less imitation of prosodic elements; 2) Greater deficits in joint attention and understanding of the meaning of emotions; 3) Serious difficulties in false-belief tasks.

Mean age is expressed in years. Conversion was done when it was expressed in months TD, Typically Developing; FER, Facial Emotion Recognition; ER, Emotion Recognition; ToM, Theory of Mind

2.5.1.3 Emotion Identification Comparing Samples

Research comparing conditions is summarized in Table 2.3. Three out of the four papers found addressed the emotion identification domain from a comparative perspective. This kind of research is scarce but still yielded mixed results. In a study using the Affect Recognition subtest of the Developmental Neuropsychological Assessment (NEPSY-II; Korkman et al., 2007) with only visual stimuli, a general FER deficit was observed in all basic emotions in children with ASD but not in the SLI group, which performed similarly to the controls in this domain (Loukusa et al., 2014). Conversely, Boucher et al. (2000) assessed auditory processing using affect vocal cues in a study involving children with low functioning ASD, SLI, and TD matched on verbal ability. Participants were asked to name emotions listened through audio cues and match them to photographs of emotional facial expressions. The SLI group showed the poorest performance in both tasks naming emotions and matching them to the appropriate facial expression, while the ASD group only performed poorer than the controls in the matching task.

In turn, Taylor et al. (2015) studied ER using visual and auditory stimuli in both clinical populations, distinguishing between children with ASD and language delay (ASD-LD) and with normal language (ASD-NL). They concluded that language played a fundamental role in ER (in terms of both accuracy and latency) in autistic disorder, as the ASL-LD group performed similarly to their counterparts with SLI. Specifically, the results revealed that the SLI and ADS-LD groups had difficulties in recognizing all types of emotions, while children with ASD-LN only showed deficits in more complex emotions such as disgust and surprise; this pattern was found for both the visual and the auditory modality. Likewise, children with SLI had longer latencies than the other groups in all modalities. These results may support the hypothesis of a shared etiology between SLI and the ASD-LD subgroup.

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Study	Main Features
Boucher et al. (2000)	<u>Groups</u> : Low functioning ASD ( $n = 19$ ; $M_{age} = 9.7$ , $SD = 1$ ), SLI ( $n = 19$ ; $M_{age} = 9.0$ , $SD = 1.4$ ), and TD matched on verbal ability ( $n = 19$ ; $M_{age} = 6.4$ , $SD = 0.8$ ).
	EC domains assessed: Emotion identification (auditory processing, ER from auditory cues).
	<u>Tasks</u> : Vocal affect labeling tasks (naming emotions from vocal cues) and vocal affect matching task (matching vocal and visual emotional cues).
	Similarities between disorders: None.
	<u>Differences between disorders</u> :1) Deficit in naming and matching basic emotions in SLI; 2) Adequate performance in naming basic emotions in ASD but deficit in matching emotions (even so better performance than SLI); 3) Deficit in voice processing in children with SLI.
Colle et al. (2007)	<u>Groups</u> : ASD ( $n = 12$ ; $M_{age} = 8.1$ , $SD = 1.9$ ), SLI ( $n = 15$ ; $M_{age} = 8.3$ , $SD = 1.8$ ), and TD matched on mental age ( $n = 15$ ; $M_{age} = 4.6$ , $SD = 0.9$ ).
	EC domains assessed: Emotion understanding (ToM).
	Tasks: False belief tests.
	Similarities between disorders: None.
	<u>Differences between disorders</u> : ToM impairment for tasks with low linguistic load only in ASD.
Loukusa et al. (2014)	<u>Groups</u> : ASD ( $n = 14$ ; $M_{age} = 7.2$ , $SD = 1.3$ ), SLI ( $n = 18$ ; $M_{age} = 6.2$ , $SD = 0.9$ ), and TD ( $n = 25$ ; $M_{age} = 6.7$ , $SD = 1.1$ ).
	EC domains assessed: Emotion identification (FER) and emotion understanding (ToM).
	<u>Tasks</u> : Neuropsychological assessment (ER and ToM subtests of the social perception domain of the of NEPSY-II; Korkman et al., 2007).
	Similarities between disorders: ToM deficits.
	Differences between disorders: Deficit in recognition of basic emotions in ASD.
Taylor et al. (2015)	<u>Groups</u> : ASD-LD ( $n = 12$ ; $M_{age} = 7.7$ , $SD = 1.9$ ), ASD-NL ( $n = 17$ ; $M_{age} = 9.7$ , $SD = 2.3$ ), SLI ( $n = 18$ ; $M_{age} = 7.4$ , $SD = 1.4$ ), and TD ( $n = 54$ ; $M_{age} = 8.5$ , $SD = 1.9$ ).
	EC domains assessed: Emotion identification (FER and ER from auditory cues).
	Tasks: Matching task (pairing emotions, forced-choice paradigm).
	<u>Similarities between disorders</u> : Poorer performance in recognition of all basic emotions (visual and auditory modality) in SLI and ASD-LD.
	<u>Differences between disorders</u> : 1) Deficit in recognition of surprise and disgust (visual and auditory modality) in ASD-NL; 2) Longer latencies in all modalities in SLI.

**Table 2.3.** Summary of comparison between ASD and SLI in EC

Mean age is expressed in years. Conversion was done when it was expressed in months TD, Typically Developing; ER, Emotion Recognition; ToM, Theory of Mind; ASD-LD, ASD with language delay; ASD-NL, ASD with normal language development

# 2.5.2 Emotion Understanding

# 2.5.2.1 Emotion Understanding in Children with ASD

None of the selected studies targeting population with ASD addressed specifically the emotion understanding domain. However, Begeer et al. (2008) reviewed the four EC domains in children with ASD and determined that innate emotional skills were not

completely absent in young children with autism, as they adequately expressed basic emotions, attended to faces, and perceived facial expressions. Likewise, school-age children with ASD responded to the others' emotions and understood causality in emotional situations like their peers. However, when examining more complex skills in these children, the expressiveness of those with autistic traits and intellectual disabilities was not socially oriented, they failed to integrate emotional information in complex social contexts, needed help in structuring social exchanges, responded poorly to others' emotions, and had a limited awareness of mental states. Conversely, children with highfunctioning ASD displayed more emotional skills than those with low-functioning, which suggested a late acquisition of some components of EC with respect to their peers, rather than an impairment. In sum, the wide heterogeneity within the autistic spectrum yields different EC levels depending on the impairment severity.

Likewise, a study on empathy in children with ASD and conduct disorder confirmed the empathy imbalance hypothesis (EIH) in individuals with ASD (Schwenck et al., 2012). EIH posits that the cognitive dimension of empathy (e.g., ER and perspective taking, related to the emotion identification and understanding domains) is impaired in individuals with ASD, but the emotional dimension (adoption of behaviors congruent with the perceived emotions, related to the emotional expression and regulation domains) remains intact.

#### 2.5.2.2 Emotion Understanding in Children with SLI

Three articles have addressed abilities related to emotion understanding in children with SLI. Thus, this population showed an adequate knowledge of emotions (e.g., emotion identification and understanding) but they failed to integrate this knowledge within the contextual information of scenes, which could account for the deficit found in their ability to infer emotions from stories according to the study of Ford and Milosky (2003).

Likewise, Brinton et al. (2007) examined the ability of children with SLI to judge emotional situations using stories. To do so, it was necessary to understand the stories, be able to distinguish between experienced and displayed emotion (emotion concealment) and know the cultural rules that condition emotional display. These authors concluded that both SLI and TD groups understood the social desirability of the emotional response Chapter 2. Emot ional Competence in Children with Autism Spectrum Disorders (ASD) and Specific Language Impairment (SLI). A Comparative Research Review in each situation, but they had different perspectives on the relevance of displaying them or not. This suggested that children with SLI had different social objectives than the controls (perhaps due to a lack of experience or social exposure).

Conversely, a moderate to severe social-emotional deficit modulated by the link between language and ToM has also been found in preschoolers with SLI (for a review, see Vissers & Koolen, 2016). Particularly, these authors pointed to a lower propensity to imitate prosodic elements, greater deficits in joint attention and in the understanding of the meaning of emotions, and notable difficulties in false-belief tasks.

# 2.5.2.3 Emotion Understanding Comparing Samples

The study of the ToM has been included in this domain as it involves the understanding of emotional situations and states to perform the task. In this sense, a deficit in ToM has been highlighted in both populations according to the results of Loukusa et al. (2014), although no association was found between this and ER in any disorder in this study. Conversely, Colle et al. (2007) suggested that only the ASD group was affected in ToM when low linguistic load tasks were performed. These results impede drawing conclusions on emotion understanding in each condition as they are scarce and inconsistent.

# 2.5.3 Emotion Expression

Studies on the autonomic activity in children with ASD have highlighted an intact physiological response toward emotional stimuli in this population (Ben Shalom et al., 2006; Zantinge et al., 2019). These studies used electrodes to measure skin conductance responses and heart rate in ASD. Ben Shalom et al. (2006) also examined and the conscious emotional response in their sample through self-reports. Hence, they found important deficits in individuals with ASD in the cognitive dimension of the emotion (awareness of experiencing a certain emotion), but not in the physiological dimension in which both groups showed similar skin responses. This means that their physiological emotional expression may be intact in this population, while the expression of conscious feelings and emotions seems to be impaired. In turn, Zantinge et al. (2019) analyzed emotional expression (facial expressiveness after viewing emotional stimuli) towards fearful situations in young children with ASD and control peers. They found that facial

Early Differential Diagnosis in Infant Population with Neurodevelopmental Disorders. Psychophysiological Markers Based on Eye Tracking Methodology response to fearful stimuli was identical between groups, which implies that children with ASD display facial expressions appropriately in fearful situation.

Conversely, in the study of Li et al. (2011) toddlers with ASD showed less emotional reaction to negative emotions, but similar levels of response to positive emotions compared to their peers. It means that they displayed positive emotional expressions when viewing happy faces, but they did not adapt their facial expression when viewing fearful, angry, or sad faces, remaining indifferent or unexpectedly positive to these expressions. It was hypothesized that this peculiarity could be attributed to a deficit in emotion recognition and categorization in toddlers with ASD, this means that identification and categorization of negative emotions may be more challenging for them, which impairs their understanding of the emotion and, consequently, their emotional response. Discrepancies between results regarding emotion expression in children with ASD could be due to the nature of stimuli used in each case; while Zantinge et al. (2019) used a robot toy which approached to participants to elicit fear, Li et al. (2011) presented children with faces displaying both positive and negative emotions.

None of the selected studies addressed specifically the emotion expression domain either in children with SLI or in both populations jointly.

# 2.5.4 Emotion Regulation

In a comparative study between children and adolescents with ASD and 22q13 deletion syndrome, Glaser and Shaw (2011) concluded that subjects with ASD had adequate developmental and daily living skills in the physical, cognitive, adaptive, and communicative domains, but were significantly impaired in emotion regulation (understood as the detection, evaluation, and modification of emotional reactions to achieve a goal) and showed poorer social-emotional skills than the comparison group. Likewise, young children with ASD have demonstrated to display atypical emotion regulation strategies as they tend to rely more on others than on themselves to regulate their emotional states (Cibralic et al., 2019).

Finally, poorer emotion regulation has also been reported in children with SLI which, along with language competence, could predict the social reticence behavior of the clinical sample (Fujiki et al., 2004). None of the selected studies tackled this domain from a comparative perspective.

A set of studies reported considerable social-emotional difficulties in children with SLI (Bakopoulou & Dockrell, 2016; St Clair et al., 2011; Vissers & Koolen, 2016); however, emotional difficulties apparently decreased from childhood to adolescence, where they were also present to a lesser extent (St Clair et al., 2011).

#### 2.6 Discussion and Conclusions

The aim of this review was to gain insight into the state-of-the-art research on EC in child populations with ASD and SLI from a comparative point of view which could lead to identify differences, facilitate diagnosis, and guide interventions. For that purpose, some questions were raised, and they are discussed in the following paragraphs.

#### 2.6.1 Which EC Domains Have Been Addressed in each Population?

Results in population with ASD show that emotion identification has received the greatest research attention (particularly in visual modality, FER), while other domains of EC such as emotion understanding, expression, and regulation have seldom been addressed. Results with SLI samples show a common bias to the study of emotion identification but these studies comprised a wider range of modalities (visual, auditory, and audiovisual). Other domains assessed in children with SLI are emotion understanding and, more superficially, emotion regulation. No study has addressed emotion expression in this population and the EC construct has been tackled more generally that in the population with ASD. Regarding comparative studies, research has exclusively focused on emotion identification in different modalities and ToM, which was related to the emotion understanding domain.

This common focus on emotion identification may be partially explained by the fact that there is a wide range of well-validated tools and tasks for the assessment of this domain, which make the experimental process much easier. Conversely, other domains such as emotion understanding, expression, and regulation can only be evaluated through questionnaires or their task validating process is highly complex. This issue not only impacts on the amount of research on each domain, but also prevents from reaching sound conclusions on the description and comparison of the EC in both populations.

#### 2.6.2 To Date, What Is It Known about EC in each Condition?

#### Autism Spectrum disorders

Research on children with ASD indicates that they have difficulties in some domains of EC. Regarding FER, the most consistent studies confirm significant impairments in this population (Beeger et al., 2008; Davidson et al., 2019; Evers et al., 2015; Golan et al., 2018; Griffiths et al., 2019; Harms et al., 2010; Lacroix et al., 2016; Lozier et al., 2014; Uljarevic & Hamilton, 2013). Imaging studies have highlighted an abnormally large amygdala and hippocampus volumes in children with ASD regardless of the disorder severity (Schumann et al., 2004) and an atypical activation in the subcortical and paralimbic/limbic regions (Aoki et al., 2015). These areas are closely related to the processing of facial emotional expressions, which could explain their FER difficulties. Specifically, the study of Schumann et al. (2004) found that amygdala and hippocampus atypicalities were less pronounced in children with Asperger syndrome than in those with high- and low-functioning autism, which is in line with FER difficulties related to the severity the autistic disorder. Likewise, children with ASD have shown less social attention (visual attention in terms of total looking time spent on looking at a target stimulus and joint attention in terms of behaviors like looking back at the adult and finger-pointing while viewing the stimuli) to emotional faces than the control group (Li et al., 2011). This could also explain FER difficulties found in this population in most studies.

On the other hand, while the identification of specific impaired emotions varies between studies, most agree that negative valence emotions are the most compromised in children with ASD (Evers et al., 2015; Farran et al., 2011; Lozier et al., 2014; Uljarevic & Hamilton, 2013). In this line, a pattern of higher facial response to positive valence emotions, less attention to social stimuli, and a limited facial expressiveness to negative valence emotions has been identified in this population (Li et al., 2011), which could partially explain these findings.

Emotion understanding in children with ASD seem to be impaired depending on the severity disorder, being children with low-functioning ASD and/or intellectual disabilities those who struggle the most with this domain (Begeer et al., 2008). Deficits in understanding others' emotions and intentions, awareness of own emotions, and Chapter 2. Emot ional Competence in Children wi th Autism Spectrum Disorders (ASD) and Specific Language Impairment (SLI). A Comparative Research Review perspective taking in children with ASD could be partially explained by the mirror neuron system theory suggested by Dapretto et al. (2006), according to which the lack of activation of this system in individuals with ASD could be impairing abilities such as imitation, intention and emotion understanding, and empathy.

Finally, emotion expression is partially compromised in this population as physiological expression seem to be intact, while the expression of conscious feelings and emotions and the emotional response to negative emotions are compromised (Ben Shalom et al., 2006; Li et al., 2011). Emotion regulation difficulties have also been reported in children with ASD (Glaser & Shaw, 2011).

#### Specific Language Impairment

Regarding children with SLI, the results are more homogeneous, concluding that this population shows a general social-emotional deficit that notably impacts on the quality of their social interactions (St Clair et al., 2011; Vissers & Koolen, 2016). There is also consensus that FER remains intact (Czaplewska & Sterczynski, 2015; Delaunay-El Allam et al., 2011; Ford & Milosky, 2003), although emotion identification is compromised in auditory and audiovisual modalities (Creusere et al., 2004; Czaplewska & Sterczynski, 2015). This latter finding suggests that those domains of emotion with higher linguistic load are the most impaired in this population, as the presence of verbal content hinders emotional information processing. This statement is supported by the fact that reducing the role of language drives these children to perform similarly to their peers. Similar to the population with ASD, children with SLI better recognize emotions with positive valence (Bakopoulou & Dockrell, 2016) and, similar to TD children, they show a bias towards them (Merkenschlager et al., 2012).

Emotion understanding is generally intact in children with SLI, although more complex abilities such as inference of emotions from stories or emotion regulation have proved to be impaired (Brinton et al., 2007; Ford & Milosky, 2003; Fujiki et al., 2004). Nevertheless, more research on these domains is needed to confirm these results as these domains and abilities have been addressed scarcely and no study has tackled emotion expression in this population.

# 2.6.3 Which Are the Main Similarities and Differences between ASD and SLI in Terms of EC?

Taken together, most studies agree that both conditions have a global deficit in EC, which significantly affects their social functioning. Emotion regulation seems to be impaired in both conditions (Fujiki et al., 2004; Glaser & Shaw, 2011); however, this domain has not been deeply analyzed and methodologies significantly differed between studies, which suggests caution when considering these results. Both groups align with their TD peers in showing a bias toward positive emotions, which have important implications for intervention as it encourages the use of positive emotions with these children to foster their attention and their goal achievement.

Although comparing disorders in particular domains is difficult, as the amount of research on each one is clearly imbalanced, this review yields some differential markers:

- Children with ASD show serious difficulties in FER (Beeger et al., 2008; Harms et al., 2010; Lozier et al., 2014), while impairments in emotion identification via auditory and audiovisual modality seem to be a marker of population with SLI (Boucher et al., 2000; Creusere et al., 2004; Czaplewska & Sterczynski, 2015; Taylor et al., 2015).

- Children with ASD find more difficulties with matching tasks (Boucher et al., 2000; Golan et al., 2018), while children with SLI struggle more with labeling tasks due to the linguistic nature of their impairments (Boucher et al., 2000; Delaunay-El Allam et al., 2011). Thus, the kind of task may play a crucial role in distinguishing these populations from a clinical perspective. Some kind of tasks (e.g., forced-choice) and the prototypical stimuli included on standardized tests could be so easy for some children that they excel over their real ability. These methodological issues may drive to ceiling or floor effects, which bias research findings either by hiding or by stressing deficits in both clinical populations (Delaunay-El Allam et al., 2011; Harms et al., 2010). Hence, this aspect must be carefully considered when assessing these populations either for research or for clinical purposes.

- Paying attention to the linguistic component is fundamental to disentangle the distinction between conditions due to its role as a moderating variable of EC in both populations. Therefore, children with ASD seem to benefit from verbal support during FER (Golan et al., 2018), while verbal content seems to impair visual processing

Chapter 2. Emot ional Competence in Children wi th Autism Spectrum Disorders (ASD) and Specific Language Impairment (SLI). A Comparative Research Review during FER tasks in children with SLI. Moreover, the results of Taylor et al. (2015) raise the issue of distinguishing an ASD subgroup with language impairment that may explain similarities between conditions and the difficulties clinicians often find when diagnosing one disorder or another.

- Children with severe ASD have difficulties in emotion understanding (Begeer et al., 2008), while those with SLI show no impairment in basic emotion understanding (Brinton et al., 2007; Ford & Milosky, 2003). Results in ToM are inconclusive as only two studies compared clinical populations (Colle et al., 2007; Loukusa et al., 2014) and they yielded inconsistent conclusions.

#### 2.6.4 Future Directions

To summarize, very few studies have compared these populations, which is particularly striking given the high co-occurrence rate reported in both clinical practice and the research (Boucher et al., 2000) and the overlapping behaviors highlighted in some studies (Taylor et al., 2012, 2014, 2015). Therefore, more research comparing populations with ASD and SLI is still needed to disentangle the potential causes that may hinder differential diagnosis. It would also be interesting to isolate the linguistic component in ASD to quantify the influence of language on EC performance in this population. This effect could also be explored through specific language interventions in which progressive improvements in the linguistic and emotional domains could be measured and compared in both disorders. Whichever option is chosen, this line of research should continue and include cutting-edge technologies and methodologies to provide answers to many unresolved questions. In this sense, imaging and eye tracking studies focused on neural functioning and visual emotion processing may contribute as they add empirical validity to behavioral outcomes.

Finally, it is equally important to foster research on the emotional behavior of young children with ASD, as early identification broadens intervention options and improves outcomes. Several studies have demonstrated a significant progress and decrease in autistic symptoms in the population with ASD after receiving intensive early intervention. Likewise, this reasoning is also applicable to research on population with SLI, in which greater knowledge of their emotional skills is key to preventing poor social adjustment, high levels of anxiety and depression, and the higher rates of bullying these

Early Differential Diagnosis in Infant Population with Neurodevelopmental Disorders. Psychophysiological Markers Based on Eye Tracking Methodology children experience in adolescence (see Wadman et al., 2011), which could be mitigated through early interventions in social-emotional deficits.

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# Chapter 3. Eye Tracking Methodology for Studying Emotional Competence in Children with Autism Spectrum Disorders (ASD) and Specific Language Impairment (SLI). A Comparative Research Review

### 3.1 Preamble

After having dug into the social-emotional profile of children with ASD and those DLD, in this chapter the reader is going to find out more about the abilities of these populations to perceive, process, and recognize emotions. The use of up-to-date technologies has allowed researchers to develop new methodologies to describe EC more thoroughly in clinical populations like those involved in this doctoral dissertation. As the reader will see throughout the following pages, the eye tracking methodology is one example of how the application of technologies in research may improve substantially the knowledge on the visual processing of social-emotional images in these clinical samples through the tracing of individuals' eye movements. Likewise, information derived from these technologies allows to draw conclusions on children's attention which contribute to better define and understand the target disorders. Therefore, the aims of this chapter are (1) To continue disentangling the similarities and differences in EC between children with ASD and those with DLD and (2) To explore how the eye tracking methodology can contribute to the definition and comprehension of both conditions and, even more important, to the differentiation of disorders. Please, note that in this chapter we again refer to SLI for the same reasons we argued in the previous one.

Vacas, J., Antolí, A., Sánchez-Raya, A., & Pérez-Dueñas, C. (2021). Eye Tracking Methodology for Studying Emotional Competence in Children with Autism Spectrum Disorders (ASD) and Specific Language Impairment (SLI): a Comparative Research Review. *Review Journal of Autism and Developmental Disorders, s.* Advance online publication. https://doi.org/10.1007/s40489-021-00261-y

# Abstract

Children with autism spectrum disorders (ASD) and specific language impairment (SLI) exhibit serious social-emotional difficulties from early infancy which hamper the differential diagnosis of these two conditions. This review aims at analyzing differences between ASD and SLI disorders in children from 0 to 12 years regarding emotional competence abilities using eye tracking methodology. The findings show specific markers in the population with ASD (e.g., difficulties in facial emotion recognition, atypical visual scanning patterns, or non-social attentional orientation) and a notable scarcity of studies involving children with SLI. Comparative research points at potential differences in visual scanning patterns. The conclusions support the use of this methodology for comparative studies between clinical populations.

**Keywords:** Eye tracking, emotional competence, child population, Autism Spectrum Disorders, Specific Language Impairment.

# **3.2 Introduction**

Neurodevelopmental disorders are defined as a set of conditions that typically occur in early development and which have a serious impact on individuals' personal, social, academic, or occupational functioning (American Psychiatric Association [APA], 2013). Among them, autism spectrum disorders (ASD) and specific language impairment (SLI)<sup>1</sup> have drawn the attention of researchers due to the social-emotional difficulties associated with these conditions.

According to the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; APA, 2013), ASD is a set of persistent traits that impair social communication and interaction, as well as restricted and repetitive patterns of behavior and interests. The manual also highlights a significant deficit in social skills such as reciprocity or social interaction understanding, and a wide variability within this population as core features of ASD. Likewise, the DSM-5 (APA, 2013) reports an ASD prevalence of around 1% in both children and adults.

<sup>&</sup>lt;sup>1</sup> It is important to note that although the DSM-5 removed the term 'specific language impairment' ('SLI') from the manual and uses the term 'language impairment' ('LI') instead, it is still widely used in the literature on the condition.

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Conversely, SLI falls under the scope of communication disorders and refers to a set of persistent difficulties in the acquisition and use of different language components and dimensions (DSM-5; APA, 2013). Following the DSM-5 criteria, SLI entails early onset; language abilities significantly below average for the age when no auditory, sensory, or motor dysfunction, intellectual disability, or neurological condition is present; and important functional constraints in individuals' daily life.

Although the distinction between ASD and SLI is generally accepted, both disorders show strong similarities, particularly at early ages. These similarities have opened the discussion on a potential shared etiology (see Taylor et al., 2012, for a review). On the one hand, compromised language abilities differ in each disorder. The pragmatic language function is more reduced in the ASD than in the population with SLI, while structural components (e.g., phonology, morphology, or syntax) are more affected in SLI than in ASD. On the other hand, previous studies have found that both disorders share common features, thus highlighting the need to revise their definitions. In this regard, it is possible to find either ASD profiles with low-functioning language similar to SLI, or individuals with SLI displaying remarkable social-withdrawal behaviors and socialization difficulties such as those observed in ASD (Taylor et al., 2014; Taylor et al., 2015). Furthermore, the DSM-5 (APA, 2013) provides statistics on the high comorbidity rates between these disorders, which contributes to the discussion and accounts for the constraints in differential diagnosis.

From a theoretical standpoint, there are two views concerning differential diagnoses. Whereas there is some evidence of a shared etiology between ASD and SLI, other studies argue that the similarities found between conditions cannot sufficiently account for the unification of diagnoses due to their inconsistency (Taylor et al., 2012). Given the crucial role of differential diagnosis in clinical practice and individuals' prognosis, comparative studies between populations are clearly needed to better understand ASD and SLI, improve their differentiation, and transfer theoretical knowledge to the real-life context of practitioners, individuals, and their families (Taylor et al., 2012; Taylor et al., 2014; Taylor et al., 2015).

# 3.2.1 Emotional Competence

Language and cognitive profiles involving individuals with ASD and SLI have traditionally captured most research attention, while little was known about their socialemotional functioning until recently. Nevertheless, research interest in emotion-related aspects has grown exponentially in the last decades (Jeanneret et al., 2015), including studies on clinical populations. For example, the bibliometric study of Jeanneret et al. (2015) reported that the number of papers on facial emotion recognition (FER) has taken a quantum leap. Likewise, the amount of collaborations and indexed publications on the field has risen significantly in recent years, thus suggesting that emotional competence (EC) is gaining recognition in the research.

There is broad agreement on the greater complexity and multidimensionality of the definition and extent of EC. The EC concept comprises the skills involved in emotion identification, understanding, expression, and regulation, either one's own or those of others (Beck et al., 2012; Denham, 1998). Saarni (1999) listed eight major skills to cover the broad scope of EC: (1) Awareness of one's own emotions; (2) Ability to discern others' emotions; (3) Ability to use the vocabulary of emotion and expressions; (4) Ability to empathize with others' emotional experiences; (5) Ability to differentiate internal, subjective emotional experience from external, emotional expression; (6) Capacity for adaptive coping with aversive emotions and distressing circumstances; (7) Awareness of emotional communication within relationships; and (8) The capacity for emotional self-efficacy. Given this broadly accepted taxonomy, EC is closely linked to the socialization process as its final aim is to provide individuals with strategies to cope efficiently in social situations (Beck et al., 2012; Begeer et al., 2008; Lau &Wu, 2012). Therefore, the social and emotional dimensions of EC cannot be considered independently of each other.

From a developmental approach, there is sound evidence supporting the link between EC and other cognitive domains such as language (Beck et al., 2012; Ornaghi & Grazzani, 2013), executive function (Riggs et al., 2006), or the theory of mind (Liebermann et al., 2007) in the typically developing (TD) population. All these domains also have a strong impact on social competence (Monopoli & Kingston, 2012) and prosocial orientation (Ornaghi et al., 2016), which consequently reinforce the link between all these aspects of individuals. Chapter 3. Eye Tracking Methodology for Studying Emotional Competence in Children with Autism Spectrum Disorders (ASD) and Specific Language Impairment (SLI). A Comparative Research Review

It is also important to note that the terms EC and emotional intelligence (EI) are often used indistinctly in the literature. However, this paper uses the term EC for a number of reasons. First, EC is closely related to learning, so it feeds on the wide variety of daily social exchanges (Lau & Wu, 2012). Secondly, according to these authors, EC involves a strong cultural component due to its deployment within a specific cultural background. Finally, the EI concept has not always been defined in the same way (see Neubauer & Freudenthaler, 2005). Depending on the model chosen, for example the ability model (Mayer & Salovey, 1997) or the mixed model (e.g., Bar-On, 1997; Goleman, 1995), EI can be interpreted in different ways, while the EC concept seems to be more integrative.

### 3.2.2 Psychophysiological Measures to Study Emotional Competence

In addition to self-reported and behavioral measures, it has become increasingly common in the last decades to record psychophysiological responses as they are considered a reliable way to study EC in infancy in greater depth. In this regard, new technological tools and methodologies, such as eye tracking, have significantly contributed to the developmental understanding of this construct. Specifically, eye tracking is used to analyze individuals' gaze behavior by tracing and monitoring their eye movements during image visualization. This methodology draws on corneal reflection technology, so it yields reliable attentional markers in a non-invasive way (Aslin & McMurray, 2004; Gredebäck et al., 2009; Hepach et al., 2015). Depending on the research aims, different eye tracking measures may be used. These include duration of visits (time spent looking at the screen); number and duration of fixations (specific looking locations at each specific time); the number, location, and latencies of saccades or saccadic movements (rapid looking shifts from one point of the scene to another); anticipatory looking (prediction of stimuli location shifts); visual scanning pattern (VSP; also known as gaze pattern or looking pattern, a measure of the sequence of looking shifts); and more recently pupil dilatation (an indicator of the autonomous nervous system activation based on changes in pupil diameter), among others (see Gredebäck et al., 2009 and Hepach et al., 2015 for a review). For purposes of research, it is also useful to create different areas of interest (AOIs) in the stimuli, as they provide specific data for comparisons not only between stimuli, but also within them.

Eye tracking methodology is particularly useful when working with children, as it allows following the development of critical aspects of social-emotional competence

and their relationship with cognitive mechanisms (Aslin, 2007; Aslin & McMurray, 2004; Falck-Ytter et al., 2013; Gredebäck et al., 2009; Hepach et al., 2015). Thus, gaze pattern and pupil dilatation analysis using eye tracking has proven to be a reliable and accurate source of information on the internal physiological mechanisms and their arousal from the age of 2 (Hepach et al., 2015). In this regard, eye tracking studies have concluded that the mouth and eye regions are the most trustworthy and recurrent areas for recognizing emotions in infancy. Furthermore, a specific visual profile for the observation of painful expressions has been found in children, that involves 'faster orientation, shorter fixation duration and less fixation count' (Yan et al., 2017, p. 9). Likewise, eye tracking is a versatile methodology which can be implemented with different experimental paradigms such as dot-probe or cueing tasks (see Pfeiffer et al., 2013). In fact, the adaptation of dot-probe tasks to eye tracking seems to be effective in identifying the presence of an attentional bias towards emotional facial expressions in infants between 9 and 48 months (Burris et al., 2017).

Eye tracking has also been used jointly with other psychophysiological measures of central nervous system activity and neuroimaging techniques such as electroencephalograms (EEGs) or functional magnetic resonance imaging (fMRI) to obtain a more holistic view of the construct. To cite some examples of these experimental combinations, studies with eye tracking and EEGs in young children have shown that the eye region plays an essential role in facial processing. Likewise, the time spent looking at this region significantly correlates with the activation of certain event-related potentials when viewing negative emotions (N290 for fearful faces and P400 for angry ones). This suggests that the different neural circuitries for processing fear and anger found in adults could emerge as early as 7 months of age (Vanderwert et al., 2015). Additionally, the combination of eye tracking and fMRI has been used to study the developmental trajectory of joint attention (JA), understood as shared attention performed by following others' gaze. This line of research has shown the extent of specialization of neural mechanisms related to JA from childhood to adolescence and demonstrated that the JA network is modulated by the effects of the person who initiates the interaction (oneself or others) and familiarity with the counterpart (Oberwelland et al., 2016).

Finally, eye tracking methodology has also been applied to the study of clinical groups such as those examined in this paper. The use of this methodology in clinical

Chapter 3. Eye Tracking Methodology for Studying Emotional Competence in Children with Autism Spectrum Disorders (ASD) and Specific Language Impairment (SLI). A Comparative Research Review populations has also grown exponentially in recent decades due to the sensitivity of eye tracking to detect subtle differences in visual emotion processing and provide insight into the underlying mechanisms of facial emotional expression decoding in the population with ASD (Bolte et al., 2016; Falck-Ytter et al., 2013; Harms et al., 2010).

In brief, the above studies show that the use of technological tools in research on emotions has enabled exploring core mechanisms which were once inaccessible and whose role in EC was unknown until recently. Furthermore, eye tracking methodology has proved to be a valuable source of information in vulnerable populations such as infants or clinical ones since it facilitates data access and collection for research purposes.

# 3.2.3 Aims and Methodology of this Review

The aim of this review is to examine the contribution of eye tracking methodology to the study of EC in children with ASD and SLI, and to assess its potential in distinguishing disorders at early ages. Specifically, we have focused on studies that used this methodology in populations with ASD and SLI either from a comparative approach or independently. We performed a differential analysis, highlighting aspects of EC that can better contribute to the early identification and differential diagnosis of ASD and SLI, as well as guide interventions. As neurodevelopmental disorders appear in the earliest stages of development, our review focuses on studies using samples of children.

# 3.3 Method

# 3.3.1 Search Strategy

A literature search of the WOS, SCOPUS, and Proquest databases was conducted in November 2018. The search equation applied to all the databases was (emotional competence OR emotions) AND (ASD OR autism) OR (SLI OR language impairment) AND (eye tracking OR eye movement OR social attention OR scanning pattern) AND (child\* OR toddl\* OR infan\* OR preschool\*). These terms were included in the titles, abstracts, and keywords of the articles. The second, complementary search stage, comprised the revision of references from articles selected. Thus, nine articles were added at this stage.

# 3.3.2 Inclusion and Exclusion Criteria

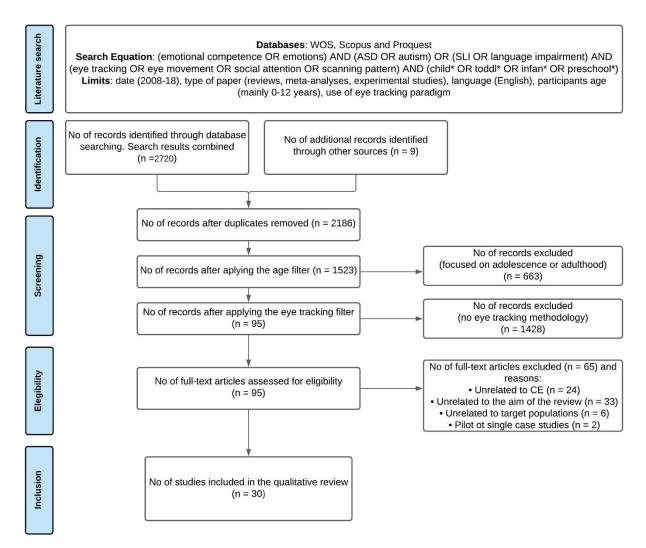
Filters applied to the search were (1) Date of publication, the search was limited to years 2008-2018; (2) Language, only studies written in English were included; and (3) Type of paper, only reviews, meta-analyses, and experimental studies were considered.

After the databases search and removing duplicates, papers were assessed according to the following inclusion criteria: (1) Participants' age, the target population were children from 0 to 12 years, and (2) The implementation of the eye tracking paradigm. Likewise, exclusion criteria comprised (1) The focus on adolescents or adults, (2) The application of paradigms or methodologies different from eye tracking, (3) The assessment of abilities unrelated to EC, (4) The unrelation to the aim of the review, (5) The inclusion of clinical groups different from the target populations, and (6) Pilot or single case studies. The literature search and results assessment were performed independently by two authors. Those publications whose fulfilment of inclusion criteria was unclear were discussed by all authors until reaching an inter-rater agreement.

# 3.4 Results

Figure 3.1 shows the results of searches carried out. After removing duplicates (543 results between the three databases), the 2186 results found were assessed according to the mentioned inclusion criteria. Thus, 663 papers were excluded for being focused mainly on adolescent or adult population and 1428 were rejected for not applying an eye tracking methodology. The resulting 95 publications were assessed for eligibility and 65 were excluded according to the exclusion criteria (see Figure 3.1); thus, 30 papers were included in the qualitative review (21 selected from the literature search and nine articles added from references screening). Likewise, 28 out of the 30 publications addressed the study of EC in ASD and two included a comparison between ASD and SLI.

Figure 3.1. Flowchart of the search procedure



# 3.4.1 Eye Tracking Research on Emotional Competence in Autism Spectrum Disorders

The results found specifically on EC in the population with ASD using eye tracking methodology, 28 studies in total (Table 3.1), can be classified according to the emotion dimension addressed<sup>2</sup>: emotion recognition (identification, matching, labelling, or naming emotions from facial or vocal prompts; n = 3), VSP of emotional images (the way individuals observe both emotional faces and scenes; n = 9), visual preference (time

 $<sup>^2</sup>$  Note that this is an approximated classification since some studies addressed more than one dimension (e.g., social attention and visual preference). In these cases, we classified papers according to the main dimension covered.

spent looking at different emotional stimuli or AOIs; n = 3), or attention to socialemotional stimuli (n = 13).

Concerning emotion recognition (n = 3), Black et al. (2017), Harms et al. (2010), and Tanaka and Sung (2016) agree in pointing out general deficits in core aspects of FER in individuals with ASD. Thus, Bal et al. (2010) highlighted longer reaction times for basic emotions and lower accuracy for anger in children with ASD compared to TD. The eye avoidance hypothesis has been suggested to account for the general impairment in FER and identity recognition in ASD (Tanaka & Sung, 2016). According to this hypothesis, individuals with ASD are reluctant to maintain direct eye contact since it is perceived as a threatening input and therefore develop eye avoidance as an adaptive mechanism. The continuous display of this mechanism impairs facial decoding and consequently their understanding of others' emotional and mental states, as well as social interactions in general.

Another possible explanation for the FER impairment found in ASD relates to visual processing. In this regard, some inconsistency across studies has been found. Four papers of those included in this review report greater differences between ASD and TD individuals in gaze behavior towards the mouth and eye regions (Chawarska & Shic, 2009; Chita-Tegmark, 2016a; de Wit et al., 2008; Falck-Ytter et al., 2013), whereas one fails to account for these differences (Guillon et al., 2014). Different results were found in studies included in Black et al. (2017), where four studies found similar visual behavior toward the eyes and the mouth in children with ASD compared to their TD peers (Bal et al., 2010; Falck-Ytter et al., 2010; Leung et al., 2013; Van der Geest et al., 2002), while two papers (Nuske et al., 2014a, b) reported reduced fixation to the eyes and mouth in this population. In this sense, Black et al. (2017) found higher abnormalities in the adolescent and adult population with ASD than in infant ones, thus suggesting a divergent course in the development of facial emotion processing in ASD compared to TD. This means that, instead of improving this skill over the years, the performance of individuals with ASD seems to deteriorate, presumably due to the lack of social exposure.

Following this line, among those studies addressing VSP of emotional images (n = 9), six papers reported an atypical scanning pattern of core facial features in children with ASD (Chawarska & Shic, 2009; Chita-Tegmark, 2016a, 2016b; de Wit et al., 2008; Falck-Ytter et al., 2013; Horlin et al., 2013). For example, a restricted scanning pattern

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of internal facial features has been found in children with ASD when attending to neutral facial expressions (Chawarska & Shic, 2009). This atypical scanning pattern seems to be more significant in 4-year-old children than in those of the age of 2, which also suggests that the cumulative lack of social experience may impair how these children explore faces over time. This scanning pattern is also related to a significant decrease in facial recognition among children with ASD compared to TD ones, indicating that this pattern affects face encoding and consequently alters these children's ability to identify familiar faces versus novel ones (Chawarska & Shic, 2009). A meta-analysis also found reduced scanning of the eyes and mouth areas and the whole face in favor of the body and nonsocial elements of scenes in children with ASD (Chita-Tegmark, 2016a). Likewise, children with ASD fail to adapt their visual scanning strategies to the social context (Horlin et al., 2013). Particularly, the time spent looking at the eye region plays an important role in social exchanges. The TD population tends to regulate this time depending on their role in an interaction (listeners or speakers). In contrast, children with ASD do not shift their VSP in response to static or dynamic emotional stimuli, which indicates difficulties to adapt to the requirements of the social situation. Similarly, Xavier et al. (2015) reported that ASD and TD children exhibit different face gaze patterns. In their study, children with ASD showed more saccades and shorter fixation duration while observing emotional faces, which seems to result in greater differences in recognition rates across emotions in the ASD sample. Specifically, happiness seems to be the most easily recognized emotion, while anger and neutral emotion are the hardest to recognize.

Conversely, similar visual strategies between ASD and TD children have been found when exposed to positive versus negative emotional faces (de Wit et al., 2008). Although children with ASD tend to look at the core facial features less than controls, ASD and TD children seem to increase the time spent looking at the eye region when faced with negative emotional expressions. Furthermore, time spent looking at the mouth seems to correlate negatively to the social and communicative deficit severity in the ASD sample, which implies that the more time looking at the mouth, the less severity of ASD symptoms, and vice versa. In a similar line, Guillon et al. (2014) did not find enough evidence in their systematic review to support a deficit of eyes gaze in favor of more mouth attention in children with ASD. Holistic face processing -a typical face processing strategy that allows people to recognize faces and to distinguish them from non-face objects- has also been reported in these children (Guillon et al., 2016). Similarly, equal

VSPs emerged between clinical and non-clinical groups in terms of the display of more visual attention to the upper side of pictures rather than the lower side across different stimuli type including human faces (both straight and inverted), monkey faces, geometric patterns, and Greebles (McPartland et al., 2011).

Regarding visual preference in the population with ASD (n = 3), studies following this paradigm usually present participants with pairs of stimuli simultaneously to analyze their VSPs mainly in terms of first fixation and duration and number of fixations on each stimulus or AOI, among other variables. Specifically, the three studies included in this category have compared geometric versus social images preference in populations with ASD and various control groups (Moore et al., 2018; Pierce et al., 2011; Pierce et al., 2016). These studies have found a significant correlation between geometric visual preference and symptoms severity in ASD, thus suggesting that geometric visual preference could be a phenotypical biomarker which could predict autism at early stages in this population. However, the salience of competing nonsocial objects seems to influence social preference in children with ASD (Sasson & Touchstone, 2014). This assumption implies that these children show the same attentional bias towards faces as TD controls when the competing non-social objects are unrelated to their circumscribed interests, as only objects belonging to their restricted interests are able to capture their attention more than faces.

Studies on visual preference are closely related to those on social attention (SA, n = 13), understood as the allocation of visual attention to social stimuli or socially relevant content of scenes, instead of non-social elements. It has been widely reported that children with ASD show reduced attention to social images or the social content of visual scenes in favor of an attention bias to non-social stimuli (Chevallier et al., 2015; Chita-Tegmark, 2016a, 2016b; Falck-Ytter et al., 2013; Guillon et al., 2014; Guillon et al., 2016; Zantinge et al., 2017). This atypical attention to social events seems to be a hallmark of autism from the first year of life and is usually accompanied by difficulties in attention disengagement (Falck-Ytter et al., 2013) and lack of arousal modulation towards others' emotions (Zantinge et al., 2017). A disperse gaze pattern when viewing social images has also been reported in children with ASD. That is, they display scattered eye movements across the scene when facing social situations (Liberati et al., 2017).

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Despite the previous research on SA deficits in infants with ASD, Chevallier et al. (2015) and Guillon et al. (2014) claim that difficulties in population with ASD are context-related, and caution should be taken before making generalized assumptions. Other variables that can modulate SA in children with ASD include the type of stimulus used, with interactive stimuli being those that better reflect differences between clinical and non-clinical groups (Chevallier et al., 2015); the social content of scenes, that is, the more people involved in the scene or the more complex the scene is, the less SA deployed by the population with ASD (Chita-Tegmark, 2016b; Guillon et al., 2014; Shi et al., 2015); the presence of speech during social interactions, which has been related to slower latencies and less social orientation to faces in children with ASD compared to their typical peers (Magrelli et al., 2013); the previously mentioned saliency of competing non-social objects (Sasson & Touchstone, 2014); and the social saliency of stimuli jointly with language ability (Stagg et al., 2014).

In line with the saliency of social stimuli, it is also noteworthy that children with ASD show increased attentional orientation to faces when they express emotionality rather than neutrality. Although children with ASD do so to a lesser extent than the controls, this pattern suggests that both ASD and TD children have emotional sensitivity (Nuske et al., 2014a, 2016; Vivanti et al., 2011). Furthermore, a study using eye tracking pupillometry identified familiarity as a potential trigger of pupil reactivity and visual attention to faces in children with ASD (Nuske et al., 2014a).

As part of the study of SA, eye tracking research in infants with ASD has analyzed JA through free observation tasks during dynamic social stimuli visualization and reported lower JA rates in children ASD (Franchini et al., 2017; Vivanti et al., 2011; Vivanti et al., 2014). More specifically, children with ASD fail to monitor others' gaze and pay less attention to facial cues regardless of gaze direction, which causes them to fail to predict others' behavior and respond accordingly (Vivanti et al., 2011; Vivanti et al., 2014). However, after reviewing the literature in the field, Guillon et al. (2014) considered the evidence supporting JA impairment in the infant population with ASD to be insufficient.

Finally, diminished social-emotional calibration has been reported in children with ASD (Nuske et al., 2016). According to these authors, this process can be defined as the regulation of one's own emotional reactions to a specific reference based on emotional

expression perceived in others. Social-emotional calibration is closely related to vicarious emotional learning or imitative learning and is a complex process which involves attention to others' emotional state, the understanding of that state, and the deployment of a coherent response.

In short, from the previous discussion we can infer that, despite the accuracy and sensitivity of eye tracking methodology, there are still a wide variety of reported outcomes which may be partially explained by the demographic features of the sample, the task requirements, and the dependent variables or measures considered in each case (Harms et al., 2010). These authors provide some clues that may help understand this inconsistency. For example, social-demographic features (e.g., age and verbal, nonverbal, or full IQ) have a greater impact on individuals with ASD and low-functioning than task requirements, as they cannot perform complex tasks. Conversely, task features have a major influence on people with ASD and high-functioning, as they can do more complex and varied tasks (e.g., matching, labelling, and imitating). In some cases, the ceiling effect may also account for the lack of differences between clinical and nonclinical samples, that is, some tasks are so dense that not even TD children can perform them accurately. Similarly, reported differences in the impaired emotions may be due to facial expression features, the type of stimulus used (dynamic vs. static), or the presentation modality (visual, auditory; unimodal, bimodal). In spite of these drawbacks, the evidence seems to widely support EC deficits in children with ASD, as well as the validity and usefulness of eye tracking methodology to better understand this issue.

Study	Main Features
Black et al.	Systematic review.
(2017)	Preserved skills: None.
	<u>Impaired skills</u> : 1) Higher deficits in FER in adolescent and adult populations than in infant ones; 2) Conclusion: divergent development of facial emotion processing.
Chawarska & Shic (2009)	<u>Groups</u> : ASD at age 1 (n = 14; Mage = 26.9 months, SD = 6.2), ASD at age 2 (n = 30; Mage = 46.4 months, SD = 6.4), TD at age 1 (n = 15; Mage = 26.3 months, SD = 6.5), and TD at age 2 (n = 15; Mage = 46.3 months, SD = 4.3).
	<u>Eye tracking measures</u> : Total time required to reach 10s of stimulus visualization, number of valid trials, time spent on each AOI, total time spent on familiar vs. novel faces, and novelty preference index (total time spent on novel stimuli divided by total time spent on all stimuli).
	<u>Type of stimuli and task</u> : Visual stimuli.(static pictures of neutral facial expressions); Free observation and face recognition task.
	Preserved skills: None.
	<u>Impaired skills</u> : 1) Less time spent on scanning internal core face features rather than external ones (significant decrease in this measure from 2 to 4 years old); 2) Impaired ability to recognize faces.
Chevallier et al.	<u>Groups</u> : ASD ( $n = 59$ ; Mage = 12.2 years, SD = 3.3) and TD ( $n = 22$ ; Mage = 14.9 years, SD = 1.7).
(2015)	Eye tracking measures: Duration of fixations per AOI.
	<u>Type of stimuli and task</u> : Visual stimuli (static pictures and dynamic short videos of objects, faces, and people interacting); Free observation task.
	<u>Preserved skills</u> : Similar SA to TD when using static and dynamic stimuli showing faces or individual people.
	Impaired skills: Reduced SA when viewing interactive social scenes.
Chita-	Meta-analysis.
Tegmark	Preserved skills: None.
(2016a)	<u>Impaired skills</u> : 1) Reduced visual scanning of eye, mouth, and whole face areas; 2) Increased attention to body and non-social elements; 3) Non-social attention bias.
Chita- Tegmark (2016b)	Meta-analysis.
	Preserved skills: None.
	<u>Impaired skills</u> : 1) High influence of social content of scenes (the more people in the scene, the less visual attention); 2) Non-social attention bias.
de Wit et al. (2008)	<u>Groups</u> : ASD (n = 13; Mage = 62 months, SD = 10.7) and TD (n = 14; Mage = 59.2 months, SD = 1.3).
	Eye tracking measures: Total looking duration and percentage of looking time in three AOIs (eye, mouth, and nose).
	<u>Type of stimuli and task</u> : Visual stimuli (static pictures of happy, calm, angry, and fearful facial expressions); Free observation task.
	<u>Preserved skills</u> : Display of different visual strategies when facing positive vs. negative emotional faces just like TD do.
	<u>Impaired skills</u> : 1) Less looking time spent on core facial features; 2) Negative correlation between looking time at the mouth and the severity of social and communicative deficit (the higher the severity, the less time spent on this area).
Falck- Ytter et al. (2013)	Review.
	Preserved skills: None.
	<u>Impaired skills</u> : 1) Reduced attention to people and their faces (particularly the eye and mouth regions) as early markers in ASD; 2) Difficulties in attention disengagement.

**Table 3.1.** Summary of eye tracking research on EC in ASD

<u>Groups</u> : ASD ( $n = 25$ ; $M_{age} = 34.2$ months, $SD = 9.6$ ) and TD ( $n = 21$ ; $M_{age} = 32.5$ months, $SD = 12.4$ ).
Eye tracking measures: Duration of fixations per AOI.
<u>Type of stimuli and task</u> : Visual stimuli (dynamic short videos of people looking at objects located either at their right or their left with surprised or neutral expressions); Free observation task.
<u>Preserved skills</u> : Typical response to JA in the most intense emotional expression condition.
<u>Impaired skills</u> : 1) Reduced response to JA (measure associated to the intensity of the emotional expression); 2) Reduced attention to faces.
Systematic review.
<u>Preserved skills</u> : 1) Lack of support for reduced eye attention and increased mouth gaze when viewing faces; 2) Non-conclusive results on deficits in JA.
Impaired skills: Reduced social orientation and attention in some contexts.
<u>Groups</u> : ASD ( $n = 17$ ; $M_{age} = 40.9$ months, $SD = 12.1$ ) and TD ( $n = 23$ ; $M_{age} = 43.5$ months, $SD = 14.3$ ).
Eye tracking measures: Total duration of fixations and duration of first fixation.
<u>Type of stimuli and task</u> : Visual stimuli (static pictures of non-emotional faces (straight and inverted); Free observation task.
Preserved skills: Holistic face processing.
Impaired skills: Reduced social orientation and attention.
Systematic review.
<u>Preserved skills</u> : Accuracy in facial recognition of basic emotions in congruent contexts and prototypical facial expressions in high-functioning ASD.
Impaired skills: 1) Abnormal emotional face processing; 2) Deficits in FER in ASD.
<u>Groups</u> : ASD ( $n = 19$ ; $M_{age} = 10.86$ years, $SD = 1.07$ ) and TD ( $n = 19$ ; $M_{age} = 10.65$ years, $SD = 1.04$ ).
Eye tracking measures: Number and duration of fixations.
<u>Type of stimuli and task</u> : Visual stimuli (static and dynamic pictures of facial emotional expressions); Emotion and face identification task.
Preserved skills: None.
<u>Impaired skills</u> : 1) Difficulties to adapt visual strategies to the social context; 2) Consistent pattern across stimuli type.
<u>Groups</u> : ASD ( $n = 20$ ; $M_{age} = 59.7$ months, $SD = 20.3$ ) and TD ( $n = 20$ ; $M_{age} = 57.6$ months, $SD = 20.6$ ).
<u>Eye tracking measures</u> : Latencies, amplitude, and duration of saccades, and number and duration of fixations.
<u>Type of stimuli and task</u> : Visual stimuli (static pictures of adults looking at one of two equal objects located at the right or left side of their heads); Free observation task.
Preserved skills: Attentional shift tendency identical to TD.
Impaired skills: 1) Disperse gaze pattern, scattered eye movements across the scene when
observing social images; 2) Higher frequency of long saccadic amplitudes in ASD compared to TD.
<u>Groups</u> : ASD ( $n = 14$ ; $M_{age} = 6.06$ years, $SD = 2.46$ ) and TD ( $n = 17$ ; $M_{age} = 3.99$ years, $SD = 1.27$ ).
<u>Eye tracking measures</u> : Reaction times to orient attention, duration of first fixations, and distance gaze-to-gaze.
Type of stimuli and task: Gambling session; Interactive playing task.
Preserved skills: Typical reaction times and fixation duration when looking at faces.

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McPartlan d et al.	<u>Groups</u> : ASD $(n = 15; M_{age} = 14.5 \text{ years}, SD = 1.7)$ and TD $(n = 17; M_{age} = 14.5 \text{ years}, SD = 1.3)$ .
(2011)	Eye tracking measures: Duration of fixations per AOI.
	<u>Type of stimuli and task</u> : Visual stimuli (static pictures of human faces -straight and inverted-, monkey faces, geometrical patterns, and Greebles); Free observation task. <u>Preserved skills</u> : Similar visual attentional pattern between clinical and non-clinical groups
	(more focus on the upper side than in the lower in all stimuli type). <u>Impaired skills</u> : Lower rates of facial recognition and social functioning (not assessed with eye tracking, but with behavioral measures).
Moore et al. (2018)	<u>Groups</u> : ASD (n = 76; Mage = 30.0 months, SD = 8.8), ASD traits (n = 11; Mage = 31.9 months, SD = 8.9), developmental delay (n = 56; Mage = 26.8 months, SD = 9.5), TD (n = 51; Mage = 29.7 months, SD = 9.5), other non-specified disorders (n = 22; Mage = 33.6 months, SD = 10.3), and typical siblings (n = 11; Mage = 27.8 months, SD = 11.2).
	Eye tracking measures: Number and specific and total duration of fixations per AOI and frequency of saccades.
	<u>Type of stimuli and task</u> : Visual stimuli (GeoPref test and Complex Social GeoPref test - dynamic short videos pairing social images with geometric patterns-); Free observation task.
	Preserved skills: None.
	<u>Impaired skills</u> : 1) Preference for geometric figures rather than social stimuli in a subtype of children with ASD; 2) This preference highly correlates with symptoms severity.
Nuske et al. (2014)	<u>Groups</u> : ASD ( $n = 21$ ; $M_{age} = 3.98$ years, $SD = 1.05$ ) and TD ( $n = 21$ ; $M_{age} = 4.27$ years, $SD = 0.60$ ).
	Eye tracking measures: Reaction of pupils (pupil diameter) and number and duration of fixations.
	<u>Type of stimuli and task</u> : Visual stimuli (dynamic short videos of familiar and unfamiliar people posing fearful expressions); Free observation task.
	<u>Preserved skills</u> : 1) Typical pupil reaction and attention to eyes when facing fearful expressions in familiar faces; 2) Increased attention to emotional expressions rather than neutral ones, regardless of the familiarity level.
	<ul> <li><u>Impaired skills</u>: 1) Reduced pupil reaction towards fearful expressions in unfamiliar faces;</li> <li>2) Reduced attention to neutral expressions in general; 3) Slower responses in all stimuli;</li> <li>4) Influence of familiarity in their response/reactivity towards others' emotions.</li> </ul>
Nuske et al. (2016)	<u>Groups</u> : ASD ( $n = 20$ ; $M_{age} = 4.03$ years, $SD = 1.09$ ) and TD ( $n = 20$ ; $M_{age} = 4.25$ years, $SD = 0.63$ ).
	Eye tracking measures: Reaction of pupils (pupil diameter) and number and duration of fixations.
	<u>Type of stimuli and task</u> : Visual stimuli (dynamic short videos of people reacting emotionally to hidden objects); Free observation task.
	<u>Preserved skills</u> : Typical visual attention to faces and coherent pupil reactivity towards others' emotional expressions.
	Impaired skills: Reduced social-emotional calibration.
Pierce et al. (2011)	<u>Groups</u> : ASD (n = 37; Mage = 26.7 months, SD = 7.7), developmental delay (n = 22; Mage = 22.7 months, SD = 8.5), TD (n = 51; Mage = 24.6 months, SD = 8.2).
	Eye tracking measures: Duration of fixations per AOI and number of saccades.
	<u>Type of stimuli and task</u> : Visual stimuli (dynamic short movie pairing social and geometric scenes); Free observation task.
	<u>Preserved skills</u> : None. <u>Impaired skills</u> : Preference for geometric figures rather than social stimuli in a subtype of
5	children with ASD.
Pierce et al. (2016)	<u>Groups</u> : ASD (n = 115; Mage = 28.0 months, SD = 8.4), ASD traits (n = 20; Mage = 22.2 months, SD = 9.3), developmental delay (n = 57; Mage = 22.0 months, SD = 8.3), TD (n = 64; Mage = 23.6 months, SD = 9.9), other non-specified disorders (n = 53; Mage = 22.0 months, SD = 8.7), and typical siblings (n = 25; Mage = 19.1 months, SD = 6.0). <u>Eve tracking measures</u> : Duration of fixations per AOI and number of saccades.

Preserved skills: None.Impaired skills: None.Impaired skills: 1) Preference for geometric figures rather than social stimuli in a subty of children with ASD; 2) This preference highly correlates with symptoms severity.Sasson & Groups: ASD (n = 15; $M_{age}$ = 45.7 months, $SD = 11.2$ ) and TD (n = 15; $M_{age}$ = 40.5 month $SD = 9.7$ ).e (2014)Eve tracking measures: Total duration of fixations per AOI (preference), individu duration of fixations (duration), and latency of first fixation (prioritization). Type of stimuli and task: Visual stimuli (still prictures pairing faces with non-social stimul (some from their circumscribed interests, some not); Free observation task. Preserved skills: Typical attention to faces when competing non-social object is, the less attention to faces).Shi et al.Groups: ASD (n = 13; $M_{age} = 60.85$ months, $SD = 16.29$ ) and TD (n = 20; $M_{age} = 58$ . months, $SD = 13.30$ ). Eve tracking measures: Number and duration of fixations (individually and per AOI) a time to first fixation. Type of stimuli and task: Visual stimuli (dynamic short movie pairing social and geomet scenes); Free observation task. Preserved skills: Reduced SA in complex situations (more than one person in interaction sormal language (n = 11; $M_{age} = 9.6$ , years, $SD = 1.9$ ), ASD wit (2014)Stage et al.Groups: ASD with delayed language (n = 12; $M_{age} = 9.6$ years, $SD = 1.9$ , ASD wit anumber of saccades.Type of stimuli and task: Visual stimuli (static pictures of people face-to-face or back-t back -each slide contained a pair of images.); Free observational task. Preserved skills: To reduced SA in children with ASD and delayed language; 2) Failtree distinguishing social astiency of stimuli; 3) SA related to language proficiency a saliency of social stimuli. <th></th> <th><u>Type of stimuli and task</u>: Visual stimuli (GeoPref test -dynamic short videos pairing social</th>		<u>Type of stimuli and task</u> : Visual stimuli (GeoPref test -dynamic short videos pairing social
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<ul> <li>back -each slide contained a pair of images-); Free observational task.</li> <li><u>Preserved skills</u>: Typical visual attention towards the most socially salient stimuli children with ASD and normal language.</li> <li><u>Impaired skills</u>: 1) Reduced SA in children with ASD and delayed language; 2) Failure distinguishing social saliency of stimuli; 3) SA related to language proficiency at saliency of social stimuli.</li> <li>Tanaka &amp; Review.</li> <li>Sung <u>Preserved skills</u>: None.</li> <li>(2016) <u>Impaired skills</u>: Eye avoidance hypothesis in autism.</li> <li>Vivanti et <u>Groups</u>: ASD (n = 18; M<sub>age</sub> = 13 years, SD = 17 months) and TD (n = 18; M<sub>age</sub> = 12 al. (2011) years, SD = 24 months).</li> <li><u>Eye tracking measures</u>: Number of fixations per AOI.</li> <li><u>Type of stimuli and task</u>: Visual stimuli (dynamic short videos of people performing eith complex actions unconventionally or incompletely, or simple actions varying ga direction); Free observation task and imitation task.</li> <li><u>Preserved skills</u>: 1) Typical understanding of logic of others' actions; 2) Typical ability</li> </ul>		Eye tracking measures: Number and duration of fixations, location of first fixation, and
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(2016)Impaired skills: Eye avoidance hypothesis in autism.Vivanti etGroups: ASD $(n = 18; M_{age} = 13 \text{ years}, SD = 17 \text{ months})$ and TD $(n = 18; M_{age} = 12 \text{ years}, SD = 24 \text{ months})$ .al. (2011)Eye tracking measures: Number of fixations per AOI.Type of stimuli and task: Visual stimuli (dynamic short videos of people performing eith complex actions unconventionally or incompletely, or simple actions varying ga direction); Free observation task and imitation task.Preserved skills: 1) Typical understanding of logic of others' actions; 2) Typical ability	Sung	Review.
Imparted skills: Dye avoidance hypothesis in autism.Vivanti etGroups: ASD $(n = 18; M_{age} = 13 \text{ years}, SD = 17 \text{ months})$ and TD $(n = 18; M_{age} = 12 \text{ years}, SD = 24 \text{ months})$ .Eye tracking measures: Number of fixations per AOI.Type of stimuli and task: Visual stimuli (dynamic short videos of people performing eith complex actions unconventionally or incompletely, or simple actions varying ga direction); Free observation task and imitation task.Preserved skills: 1) Typical understanding of logic of others' actions; 2) Typical ability		Preserved skills: None.
<ul> <li>al. (2011) years, SD = 24 months).</li> <li><u>Eye tracking measures</u>: Number of fixations per AOI.</li> <li><u>Type of stimuli and task</u>: Visual stimuli (dynamic short videos of people performing eith complex actions unconventionally or incompletely, or simple actions varying ga direction); Free observation task and imitation task.</li> <li><u>Preserved skills</u>: 1) Typical understanding of logic of others' actions; 2) Typical ability</li> </ul>		Impaired skills: Eye avoidance hypothesis in autism.
<u>Type of stimuli and task</u> : Visual stimuli (dynamic short videos of people performing eith complex actions unconventionally or incompletely, or simple actions varying ga direction); Free observation task and imitation task. <u>Preserved skills</u> : 1) Typical understanding of logic of others' actions; 2) Typical ability		<u>Groups</u> : ASD ( $n = 18$ ; $M_{age} = 13$ years, $SD = 17$ months) and TD ( $n = 18$ ; $M_{age} = 12.2$ years, $SD = 24$ months).
complex actions unconventionally or incompletely, or simple actions varying ga direction); Free observation task and imitation task. <u>Preserved skills</u> : 1) Typical understanding of logic of others' actions; 2) Typical ability		Eye tracking measures: Number of fixations per AOI.
		<u>Type of stimuli and task</u> : Visual stimuli (dynamic short videos of people performing either complex actions unconventionally or incompletely, or simple actions varying gaze direction); Free observation task and imitation task.
		<u>Preserved skills</u> : 1) Typical understanding of logic of others' actions; 2) Typical ability to infer others' intentions based on their actions and on emotional cues; 3) Increased attention to faces when expressing emotion; 4) Increased response to direct gaze (though still less
response than TD group).		
Impaired skills: 1) Reduced attention to referential facial cues (provided by people's he		<u>Impaired skills</u> : 1) Reduced attention to referential facial cues (provided by people's head and gaze) and, consequently, misinterpretation of their intentions based on these cues; 2)
		<u>Groups</u> : ASD ( $n = 24$ ; $M_{age} = 46.54$ months, $SD = 10.55$ ) and TD ( $n = 24$ ; $M_{age} = 44.30$
Eye tracking measures: Proportion of fixations per AOI.		

	I I I I I I I I I I I I I I I I I I I
	<u>Type of stimuli and task</u> : Visual stimuli (dynamic short videos of unfinished actions in which people pretended to pick up one of two objects -one on their right, the other on their left-); Free observation task.
	Preserved skills: None.
	<u>Impaired skills</u> : 1) Reduced attention to socially relevant information; 2) Difficulties in monitoring others' gaze, predicting their behavior, and responding accordingly.
Xavier et al. (2015)	<u>Groups</u> : ASD ( $n = 19$ ; $M_{age} = 9.95$ years, $SD = 1.75$ ) and TD ( $n = 19$ ; $M_{age} = 8.84$ years, $SD = 1.79$ ).
	Eye tracking measures: Number of saccades and duration of fixations.
	<u>Type of stimuli and task</u> : Visual stimuli (static pictures of facial expressions of basic emotions), auditory stimuli (non-verbal affective vocalizations), and bimodal stimuli (combinations of the previous visual and auditory stimuli); Labelling forced-choice task.
	Preserved skills: None.
	<u>Impaired skills</u> : 1) Large variability in rates of recognition accuracy across emotions (joy is the most easily recognized, while neutral and anger are the least recognized); 2) Different gaze pattern in ASD compared to TD (more saccades and less time spent looking at faces).
Zantinge et al. (2017)	<u>Groups</u> : ASD ( $n = 28$ ; $M_{age} = 57.96$ months, $SD = 10.06$ ) and TD ( $n = 45$ ; $M_{age} = 55.22$ months, $SD = 11.31$ ).
	<u>Eye tracking measures</u> : Duration of visits, duration of fixations, and non-eye tracker measures (ECG to monitor arousal).
	<u>Type of stimuli and task</u> : Visual stimuli (dynamic short videos of non-social and social- emotional content involving happiness, sadness, anger, and fear); Free observation task.
	Preserved skills: Similar attention to the screen as TD group.
	<u>Impaired skills</u> : 1) Reduced attention to faces and social content of videos; 2) Lack of arousal modulation across emotions; 3) Relation of both atypicalities with ASD symptoms severity.
	· · · ·

# 3.4.2 Eye Tracking Research on Emotional Competence in Specific Language Impairment

Regarding general studies on EC, unlike ASD and despite the evidence of atypical EC-related difficulties in this population (see St Clair et al., 2011; Vissers & Koolen, 2016), there is a greater gap in the research addressing this issue in children with SLI and the studies that do address it use a general approach rather than examining specific aspects. Furthermore, to the best of our knowledge, no eye tracking study has addressed EC in SLI independently, but some have instead focused on discourse ability (either expressive or receptive) through the observation of prompted images (Andreu et al., 2011; Andreu et al., 2013; Palmović & Jušić, 2011; Pons et al., 2013; Serra et al., 2017).

# 3.4.3 Eye Tracking Research on Emotional Competence in Autism Spectrum Disorders and Specific Language Impairment. Comparative Studies

In addition to the research that studies EC in children with ASD and SLI separately as described above, comparative studies on both conditions have also been performed

using eye tracking methodology. This is a relevant topic due to the high comorbidity rates reported in the literature (Boucher et al., 2000), the key role of language in EC in children with ASD (Stagg et al., 2014), and the current debate on the potential shared etiology between them (Taylor et al., 2012; Taylor et al., 2014; Taylor et al., 2015). Based on our search inclusion criteria, we found two papers covering the topic from a comparative approach (Table 3.2).

In this case, both studies coincide in finding similar VSPs in the SLI and TD groups using dynamic and ecological stimuli. This gaze pattern has been described as social oriented and focused. Conversely, gaze behavior in both ASD groups was atypical in terms of non-social orientation and reduced attention to people's faces, more specifically to the eye region (Hanley et al., 2014; Hosozawa et al., 2012). Furthermore, accurate attention shifts were observed in the SLI and TD samples, that is, both looked alternatively at the appropriate speaker, while children with ASD failed to follow the speakers with their gaze (Hosozawa et al., 2012).

Nonetheless, both studies differ in their outcomes regarding attentional bias to the mouth region in children with SLI. While Hosozawa et al. (2012) found this potential compensatory mechanism in their SLI sample, this value was marginal in the group of Hanley et al. (2014), thus suggesting a slight tendency for language compensation in SLI, but which is non-significant in fact. Interestingly, this last study found no significant between-group differences in time spent on looking at the mouth.

Study	Main Features
Hanley et al. (2014)	<u>Groups</u> : ASD ( $n = 17$ ; Mage = 121 months, SD = 25), SLI ( $n = 14$ ; Mage = 115.2 months, SD = 10), and TD ( $n = 16$ ; Mage = 119.6 months, SD = 8).
	Eye tracking measures: Percentage of fixations per AOI.
	<u>Type of stimuli and task</u> : Semi-structured interaction during a magician performance; Interaction task (free observation of tricks and semi-structured conversation).
	<u>Similarities between conditions</u> : 1) Similar time spent on looking at the mouth (no compensatory strategy in either group); 2) Similar performance in socio-cognitive measures.
	<u>Differences between conditions</u> : 1) Similar VSP between SLI and TD groups (social oriented gaze behavior), whereas increased attention to bodily and non-social elements rather than the counterpart face and eyes was found in children with ASD; 2) Lower proficiency in theory of mind in ASD.
Hosozawa et al. (2012)	<u>Groups</u> : ASD ( $n = 25$ ; $M_{age} = 58.3$ months, $SD = 21.9$ ), SLI ( $n = 16$ ; $M_{age} = 30.1$ months, $SD = 5.4$ ), and TD ( $n = 25$ ; $M_{age} = 37.7$ months, $SD = 23.4$ ).
	<u>Eye tracking measures</u> : Proportion of time spent on each AOI and full gaze pattern (distance between different fixations during the whole visualization).
	Type of stimuli and task: Audiovisual stimuli (dynamic short videos of people interacting);
	Free observation task.
	Similarities between conditions: None.
	<u>Differences between conditions</u> : 1) Similar full gaze pattern (social oriented and focused) between SLI and TD groups, while atypical pattern (context oriented and more scattered fixations) was observed in the ASD sample; 2) Greater fixation time on the mouth region in the SLI group compared to the ASD and TD samples; 3) Accurate attentional shifts between speakers in SLI and TD children, lack of speakers visual following of speakers was found in the ASD group.

**Table 3.2.** Summary of eye tracking research on EC in ASD and SLI. Comparative studies

# **3.5 Conclusions**

The purpose of this review was to examine how EC is addressed in children with ASD and SLI from an eye tracking approach, and to weigh its potential contribution to the differentiation between these disorders at early ages. Furthermore, we aimed at providing a comprehensive view that could aid in the differential analysis of ASD and SLI and guide detection, diagnosis, and interventions. In this respect, the imbalance between disorders in the literature has been a major drawback in the review. Unlike the large volume of papers addressing this issue in the population with ASD, the lack of research on SLI is striking. Similarly, we have noticed a remarkable lack of sound knowledge comparing both conditions, in spite of the evidence of a high co-occurrence rate (Boucher et al., 2000) and the overlap of certain behaviors (Taylor et al., 2012; Taylor et al., 2014; Taylor et al., 2015).

Despite these drawbacks, the inconsistent outcomes found in some studies, and the greater heterogeneity within the autism spectrum, we believe there is sufficient

evidence supporting difficulties in some aspects of EC in children with ASD as most reviews and meta-analysis reported conclude (Black et al., 2017; Chita-Tegmark, 2016a, 2016b; Falck-Ytter et al., 2013; Guillon et al., 2014; Harms et al., 2010; Tanaka & Sung, 2016). The most impaired aspects of EC in this population are deficits in FER (Black et al., 2017; Harms et al., 2010; McPartland et al., 2011; Tanaka& Sung, 2016; Xavier et al., 2015); atypical VSP to faces (Black et al., 2017; Chawarska & Shic, 2009; Horlin et al., 2013; Liberati et al., 2017; Xavier et al., 2015); reduced attention to socially relevant stimuli (Chevallier et al., 2015; Chita-Tegmark, 2016a, 2016b; Falck-Ytter et al., 2013; Guillon et al., 2014; Guillon et al., 2016; Magrelli et al., 2013; Sasson & Touchstone, 2014; Shi et al., 2015; Stagg et al., 2014; Zantinge et al., 2017); and, to a lesser extent, reduced JA (Franchini et al., 2017;Vivanti et al., 2011; Vivanti et al., 2014). We have also found some evidence of a link between the ability to understand and predict others' intentions and EC in infants with ASD (Vivanti et al., 2011; Vivanti et al., 2014), which supports the close relationship between social and emotional dimensions (Beck et al., 2012; Begeer et al., 2008; Lau & Wu, 2012).

Nevertheless, caution must be taken when interpreting these results and making assumptions about EC in ASD, as the research has also yielded critical variables which may be biasing the study outcomes by either concealing significant differences between clinical and non-clinical groups or increasing their size. In particular, the demographic features of the samples, the task requirements, and the dependent variables used in the studies seem to have a significant impact on the reported outcomes (Harms et al., 2010). In line with these authors, we believe that certain aspects should be controlled for in order to reduce these between-study differences. Some of these aspects include (1) The control of ceiling and floor effects in tasks; (2) The inclusion of as many control groups as possible to override the potential effect of such variables as verbal and non-verbal IQ or mental and chronological age; (3) The use of morphed images (dynamic stimuli changing from a neutral expression to a high-intensity emotional one), as they have more ecological validity due to the fact that they reflect the most subtle changes in facial emotion; and (4) The use of technologies and novel methodologies to gain greater insight on the emotional dimension, as such approaches provide more accurate and reliable data and allow analyzing more hidden behaviors. Controlling these aspects would lead to more homogeneous protocols in the study of EC in the population with ASD and hence to more sound and robust conclusions.

Chapter 3. Eye Tracking Methodology for Studying Emotional Competence in Children with Autism Spectrum Disorders (ASD) and Specific Language Impairment (SLI). A Comparative Research Review

Regarding the population with SLI, the lack of studies using eye tracking methodology to gain deeper insight into EC in this population is particularly striking, as previous research without this methodology has already shown a generalized emotional impairment in these children that notably affects the quality of their social relationships (St Clair et al., 2011; Vissers & Koolen, 2016). Conversely, to the best of our knowledge, at least two studies to date have used eye tracking to compare children with ASD and SLI in terms of EC. Their results coincide in pointing out a difference in gaze patterns between both conditions, although the scarce research in the field and the small samples in both studies call for caution when drawing conclusions in this respect. However, in light of these results, we consider that this is a promising line of research as it has yielded potential markers which may help distinguish disorders (Hanley et al., 2014; Hosozawa et al., 2012). Therefore, research can contribute to improving clinical practice by shedding light on questions such as the high rates of comorbidity (Boucher et al., 2000) and the overlap of behaviors between disorders (Taylor et al., 2012; Taylor et al., 2014; Taylor et al., 2015).

Likewise, we have found evidence of the critical influence of language on EC in both disorders. In this sense, language onset and development seem to be related to SA in children with ASD (Stagg et al., 2014) and some authors have suggested that language trajectory could aid in distinguishing a subtype of ASD with language impairment whose EC performance is close to that of individuals with SLI (Taylor et al., 2015). If that were the case, some overlapping behaviors between conditions and the difficulties practitioners face when attempting to identify them could be partially explained. Thus, we believe that this aspect must be thoroughly considered in the future through comparative studies between clinical populations.

Furthermore, we have confirmed that the recent introduction of technological tools and novel methodologies such as eye tracking has widened the scope of research on EC in infant clinical populations. Due to the specificity of eye tracking data, the aspects of EC that have benefitted most from these tools and methods are FER, VSP, SA, and to a lesser extent, JA and visual preference. This has also led to more accurate conclusions and broadened the knowledge of automatic responses which cannot be verbally reported by individuals. The accuracy of eye tracking to unveil subtle abnormalities in gaze microstructures has been highlighted as one of its main strengths for the study of clinical

populations (Thorup et al., 2017). Therefore, in line with Falck-Ytter et al. (2013) and Harms et al. (2010), we agree that eye tracking methodology should be used for assessing EC in individuals with ASD and other neurodevelopmental disorders.

Moreover, in line with Zantinge et al. (2017), we believe it is of critical importance to continue studying emotional behavior in infants with ASD, since early diagnosis leads to more intervention opportunities and substantially better prognoses. These benefits come from the significant improvement and alleviation of autism symptoms after intensive intervention as observed in clinical practice. Similarly, we consider that the same assumption regarding the benefits of early diagnosis also applies to SLI. For this reason, the social-emotional performance in this population should be studied in greater depth in order to reduce the significant rates of social difficulties, anxiety, depression, and bullying reported during adolescence (see Wadman et al., 2011), which could be substantially reduced with early intervention in the social impairments found.

In short, we conclude this review by highlighting the need for research targeted at the differential analysis of these clinical conditions in order to identify the roots of overlapping behaviors that affect diagnosis in the early stages of development. In this regard, the use of cutting-edge technologies and methodologies like eye tracking provide valuable support to investigate hidden aspects of individuals, work with vulnerable populations like infants or clinical ones, and to report consistent and robust data. Regarding the comparative study of ASD and SLI, it is essential to control for the effect of language on social-emotional performance given its potential to account for some similarities between both disorders. This effect could also be assessed by intervening in language and following up on improvements in linguistic and emotional aspects. Regardless of the preferred method, we believe that further research in this line should be carried out following the proposed guidelines to resolve important issues and have a direct impact on clinical practice as well as all the stakeholders involved.

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# Second Part. Empirical Studies

# Chapter 4. Introduction to the Empirical Studies Compounding this Doctoral Dissertation

#### **4.1 Rationale for Addressing this Topic**

So far, we have reviewed some key points about the importance of early diagnosis and intervention, the difficulties of differential diagnosis of DLD and ASD due to the many similarities between both profiles, and the potential of the eye tracking methodology for contributing to the comparative analysis between conditions. Once we revised previous literature and drew these conclusions, we designed a set of experiments whose main purpose was to explore early psychophysiological markers in each disorder in order to contribute to the particular definition of each condition and their potential distinction and, consequently, to have a direct effect on early differential diagnosis.

To do so, we carried out three experiments applying an eye tracking methodology to assess different attentional abilities directly related to SA. The original intention was to undertake two experiments comparing three groups: (1) Young children with DLD, (2) Age-matched peers with ASD, and (3) Age-matched neurotypical children. Nevertheless, the difficulties to access to the sample alongside the current pandemic caused a serious delay and forced us to restructure the initial plan without compromising the participation of children from both clinical groups. Thus, we first analyzed data from the groups of children with ASD and controls to have a preliminary approach (studies 1 and 2); then, we added data from a small group of children with DLD to explore the potential of our experimental design to disentangle differences between conditions. Therefore, the first experiment, called 'Social Preference Study', was focused on exploring visual attention to social versus non-social stimuli in a sample of preschoolers with ASD and neurotypical peers. The second one, named 'Emotional Preference Study', addressed the assessment of visual attention to faces considering the role of the emotion displayed, the poser's gender, and the poser's age in a group of young children with ASD and their TD counterparts. Finally, the third study, called 'Comparative Study', was intended to develop a differential analysis through the comparison between disorders regarding the

visual attention to social versus non-social stimuli. In this case, we applied the same task and protocol than in the 'Social Preference Study' but we added a third group of preschoolers with DLD. We consider that this work has a significant value within the field given the scarcity of comparative studies involving both disorders. Moreover, considering the particular aims of this doctoral dissertation, the inclusion of the 'Comparative Study' in this compilation is pertinent as its results made an important contribution to the purpose of this research project as the reader will note in due course. Thus, in the following sections, we outline the aims, hypotheses, participants, designs, and procedures applied in all the three studies.

# 4.2 General Aims and Starting Hypotheses

Study 1. Social Preference Study

**Aim 1.** To compare young children with ASD and their TD peers in terms of SA assessing the role of facial emotions and competing objects in both groups.

**Hypothesis 1.1.** Young children with ASD would show significantly reduced attention to faces compared with objects, which would differentiate them from the TD group.

**Hypothesis 1.2.** Both groups (children with ASD and controls) would pay more attention to emotional faces, particularly to happy faces.

**Hypothesis 1.3.** Young children with ASD would pay atypically less attention to faces when these competed with objects related to their circumscribed interests, but typical attention when these competed with uninteresting objects.

**Hypothesis 1.4.** Both groups (children with ASD and controls) would show a significant bias toward interesting objects with respect to uninteresting ones.

Study 2. Emotional Preference Study

**Aim 2.** To compare SA in young children with and without ASD regarding the role of emotional expressions, the poser's gender, and the poser's age.

**Hypothesis 2.1.** Both, ASD and TD groups, were expected to discriminate between emotional and neutral faces, orienting faster and deploying more attention to emotional than neutral ones.

**Hypothesis 2.2.** Both groups (children with ASD and controls) would show the same pattern of visual behavior, but it would differ quantitatively. This means that children with ASD would show less orientation to faces and a reduced scanning of them compared to controls.

**Hypothesis 2.3.** Children with ASD would display less attention to child faces regardless of the emotion compared to the TD group.

**Hypothesis 2.4.** The narrowed scanning of faces in children with ASD would be replicated in a second task comprising pictures of emotional child and adult faces.

Study 3. Comparative Study

**Aim 3.** To compare young children with ASD, those with DLD, and controls in terms of visual attention to faces and objects considering the role of emotional expressions and competing objects in their VSP.

**Hypothesis 3.1.** Young children from the ASD group would display significantly less attention to faces than their counterparts of the control group. We did not have sufficient grounds to predict the visual behavior of children from the DLD group in this regard.

**Hypothesis 3.2.** Emotional faces (happy and angry ones) would lead to a specific visual processing different from that of neutral ones in all groups.

**Hypothesis 3.3.** The type of object would have a major impact on children with ASD, with objects related to their circumscribed interests significantly reducing their attention to faces, but uninteresting objects leading to typical attention to faces. As circumscribed interests are a hallmark in ASD, we expected that children with DLD as well as controls were less distracted by objects related to autistic circumscribed interests than their counterparts with ASD, which would yield differences between conditions.

# 4.3 General Design and Procedure

Participants in all the three studies were recruited from centers of early childhood intervention (the clinical samples) and from mainstream schools (the control group), all of them belonging to the province of Cordoba (Spain). As the purpose of the research project was focused on early intervention, all participants were aged 32 to 72 months.

Early intervention comprises the assistance to children from 0 to 72 months, but the detection of some neurodevelopmental conditions like those included in this dissertation cause a delay in the age at which children get access to this service.

The inclusion and exclusion criteria were defined to be the same for all studies. The inclusion criteria regarding clinical groups were: 1) The regular attendance to a center of early childhood intervention, 2) Having received a thorough assessment and a formal diagnosis of ASD or DLD by an experienced team of licensed clinicians from the public health system, 3) Having some ability to maintain attention during the eye tracking tasks and being able to understand and follow instructions, and 4) The absence of any mental or medical concomitant condition. On the other hand, participants from the control group were included in the studies if they did not have a history of developmental disorder either now or in the past, and in accordance with the gender and chronological age instead of on developmental age as the low demands of the eye tracking tasks (passive-observational tasks) did not require high-level cognitive abilities, thus developmental age was not a determinant variable in these studies. We also decided to pair groups on gender to control its potential effect on SA in all samples and because ASD and DLD are more prevalent in boys than in girls.

All the three experiments applied an eye tracking methodology and followed a similar paired preference paradigm. This implies that in all studies we tracked eye movements of children while performing passive-observational tasks in which pairs of stimuli were matched. The aims of the paired preference paradigm are to compare visual attention to competing stimuli and to assess the weight of the influence of each one on each other.

Another particularity of our studies was the way how we processed data. In this sense, we collected raw eye tracking data and we designed three indexes to operationalize the complex VSP of our participants: (1) Prioritization index resulted from the eye tracking parameter 'Time to First Fixation', which is related to attentional orientation as it reflects the speed at which individuals focus on a stimulus once it appears in their field of view; (2) Preference index arose from dividing the eye tracking parameter 'Total Fixation Duration' to a specific AOI by the total time that a participant looked at the screen, it reflects the participants' visual preference for one stimulus compared to another

one; and (3) Duration index derived from dividing the eye tracking parameter 'Total Visit Duration' to a particular AOI by the 'Number of Visits' to this AOI, this relates to the kind of processing that participants deployed (the longer Duration index, the deeper visual processing). This last index is also associated to the ability of individuals to maintain attention to a stimulus. Thus, we may say that these three indexes thoroughly described participants' VSP in terms of attentional orientation (what kind of stimulus did participants attend earlier?), visual preference (what kind of stimulus did participants attend during more time?), and depth of processing/sustained attention (what kind of stimulus did participants process in more detail?). This approach had been previously applied in the study of Sasson and Touchstone (2014).

As part of the general procedure, alongside the eye tracking measures we also assessed receptive vocabulary, affect recognition, and ToM abilities in our participants in the three studies in order to complement information on the samples and to perform correlational analyses when possible. Therefore, we applied the Peabody Picture Vocabulary Test-Third Edition (PPTV-III; Dunn & Dunn, 1997) and both subtests comprised in the social perception domain of the Developmental Neuropsychological Assessment-Second Edition (NEPSY-II; Korkman et al., 2007). We chose these assessment tools on the basis of their extensive use by practitioners and researchers when working with these samples.

On the one hand, PPTV-III is a screening test which evaluates the level of comprehension of vocabulary in persons from 2.6 to 90 years. Individuals are asked to observe an array composed by four pictures and then to indicate which one matches with a target word. They are allowed to provide their answers by pointing the finger, thus reducing the high demands of the expressive language.

On the other hand, NEPSY-II is a neuropsychological assessment battery composed by 32 tests which evaluate six main domains (attention and executive functioning, language, memory and learning, sensorimotor, social perception, and visuospatial processing) in child and adolescent population from 3 to 16 years. For the purpose of our three studies, we only applied the social perception subscale. This subscale comprises the assessment of affect recognition (the ability to recognize emotions from children faces) with 16 items in 3-4 years old children and 35 items in 5-16 years old

individuals, and ToM (the ability to mentalize or understand others' intentions, thoughts, and feelings) with 21 items (15 including verbal content and 6 contextual clues).

Finally, in the following paragraphs we are going to introduce the specific design of each study. However, more information about it is provided in each respective chapter (chapter five for study 1, chapter six for study 2, and chapter seven for study 3).

#### Study 1. Social Preference Study

We applied a  $2 \times 2$  mixed design, where we conducted separate repeated measures ANOVAs on each index with Group (ASD, control) as between-groups factor and Type of Object (related to autistic circumscribed interests, not related to them) as within-group factor. We performed a twofold analysis in which we firstly assessed participants' VSP toward faces and then participants' VSP to objects. Moreover, we firstly conducted both general analyses using data of the total faces, then we performed a second set of analyses for each individual emotion.

#### Study 2. Emotional Preference Study

In this study, we carried out two different experiments. In Experiment 1, we tested the effect of emotion and poser's gender on participants' VSP toward faces. Thus, we applied a  $2 \times 3 \times 2$  mixed design, where we conducted separate repeated measures ANOVAs on each index with Group (ASD, control) as between-groups factor and Emotion (happiness, anger, neutral) and Poser's Gender (female, male) as within-group factors.

In Experiment 2, we examined the impact of poser's age on participants' VSP to faces. Hence, we applied a  $2 \times 3 \times 2$  mixed design, where we conducted separate repeated measures ANOVAs on each index with Group (ASD, control) as between-groups factor and Emotion (happiness, anger, neutral) and Poser's Age (child, adult) as within-group factors.

## **Study 3.** Comparative Study

As in study 1, we again analyzed participants' VSP of faces and objects separately. However, in this time we applied a  $3 \times 2 \times 3$  mixed design, where we conducted separate repeated measures ANOVAs on each index with Group (DLD, ASD, control) as betweengroups factor and Type of Object (related to autistic circumscribed interests, not related to them) and Emotion (happiness, anger, neutral) as within-group factors.

# Chapter 5. Visual Preference for Social Vs. Non-Social Images in Young Children with Autism Spectrum Disorders. An Eye Tracking Study

Vacas, J., Antolí, A., Sánchez-Raya, A., Pérez-Dueñas, C., & Cuadrado, F. (2021c). Visual preference for social vs. non-social images in young children with autism spectrum disorders. An eye tracking study. *PLoS ONE*, 16(6), e0252795. https://doi.org/10.1371/journal.pone.0252795

#### Abstract

Autism Spectrum Disorders (ASD) are associated to social attention (SA) impairments. A gaze bias to non-social objects over faces has been proposed as an early marker of ASD. This bias may be related to the concomitant circumscribed interests (CI), which question the role of competing objects in this atypical visual behavior. The aim of this study was to compare visual attention patterns to social and non-social images in young children with ASD and matched typical controls (N= 38; age range 41-72 months) assessing the role of emotion in facial stimuli and the type of competing object. A paired preference task was designed pairing happy, angry, and neutral faces with two types of objects (related or not related to autism CI). Eye tracking data were collected, and three indexes were considered as dependent variables: prioritization (attentional orientation), preference, and duration (sustained attention). Results showed that both groups had similar visual pattern to faces (prioritization, more attention and longer visits to faces paired with objects non-related to their CI); however, the ASD group attended to faces significantly less than controls. Children with ASD showed an emotional bias (late orientation to angry faces and typical preference for happy faces). Finally, objects related to their CI captured attention in both groups, significantly reducing SA in children with ASD. Atypical SA is present in young children with ASD regardless of the competing non-social object. Identifying strengths and difficulties in SA in this population may have substantial repercussion for early diagnosis, intervention, and ultimately prognosis.

**Keywords:** Social attention, Autism Spectrum Disorders, visual preference, eye tracking, early diagnosis.

## 5.1 Introduction

Autism Spectrum Disorders (ASD) comprise a set of neurodevelopmental and pervasive conditions in which social-emotional and communication impairments along with restricted and repetitive behaviors are core symptoms (American Psychiatric Association [APA], 2013; Autism Europe [AE], 2015; Charman & Baird, 2002; World Health Organization [WHO], 1992). The wide heterogeneity of the autism spectrum hampers early diagnosis, which is considered an important predictor of future outcomes and prognosis as it leads to the early intervention and the deployment of appropriate supports (AE, 2015; Bennett et al., 2014; Corsello, 2005; Mandell et al., 2005; Nicholas et al., 2009; Özerk, 2016; Zablotsky et al., 2017). In this sense, research is claiming for the study of new markers to improve the early identification of this disorder. The study of social attention (SA, the allocation of visual attention to social stimuli or socially relevant content of scenes instead of non-social elements) has received increasing attention of research on ASD in the last decades. Likewise, new approaches and methodologies such as eye tracking have become widespread due to their significant contribution to the developmental understanding of SA.

Eye tracking methodology allows to analyze individuals' gaze behavior by tracing and monitoring their eye movements during image visualization. This methodology draws on corneal reflection technology, so it yields reliable attentional markers in a noninvasive way (Aslin & McMurray, 2004; Gredebäck et al., 2009; Hepach et al., 2015). This asset alongside its high sensitivity to detect subtle biases in visual emotion processing and disentangle the core mechanisms of facial emotional expression decoding have increased the use of eye tracking in research on ASD (Bolte et al., 2015; Falck-Ytter et al., 2013; Harms et al., 2010; Klin et al., 2002a).

Most studies on SA in children with ASD with eye tracking have reported reduced attention to social images or the social content of visual scenes in favor of an attentional bias to non-social stimuli (Chevallier et al., 2015; Chita-Tegmark, 2016a, 2016b; Falck-Ytter et al., 2013; Guillon et al., 2014; Guillon et al., 2016; Nakano et al., 2010; Zantinge et al., 2017). This atypical attention to social events has been found in toddlers with ASD from the first year of life and is usually accompanied by difficulties in attention

disengagement (Falck-Ytter et al., 2013; Sasson et al., 2008) and lack of arousal modulation towards others' emotions (Zantinge et al., 2017). Moreover, visual preference for geometric patterns instead of social images has been suggested as a potential marker for early diagnosis of ASD (Franchini, Glaser, Wood de Wilde et al., 2017; Moore et al., 2018; Pierce et al., 2011; Pierce et al., 2016). The scanning pattern (also known as gaze or looking pattern, the sequence of looking shifts during images visualization) has also been studied in children with ASD. Findings in this area yielded a dispersed scanning pattern among individuals with ASD when viewing social images, that is, they displayed scattered eye movements across the scene when it involved a social situation (Liberati et al., 2017). In the study on SA, face deserves special attention as it is an important gateway to perceive, recognize, and understand others' inner emotional states (Frith & Frith, 2006; Klin, 2008). Likewise, attention to faces at early ages is a natural mechanism which leads to brain specialization and, consequently, to social expertise (Klin, 2008; Johnson, 2000; Nelson, 2001; Schultz, 2005). Children with ASD have showed reduced attention to faces (Franchini, Glaser, Gentaz et al., 2017; Vivanti et al., 2011; von Hofsten et al., 2009; Zantinge et al., 2017). However, increased attentional orientation to emotional faces rather than to neutral ones was found in this population (although to a lesser extent than the controls). This may suggest that children with ASD, similar to their typically developing (TD) peers, have emotional sensitivity (Nuske et al., 2014; Vivanti et al., 2011). Moreover, some studies have reported a bias toward positive emotions and better recognition rates of these emotions in children with ASD, as well as in TD children (Farran et al., 2011; Li et al., 2011; Uljarevic & Hamilton, 2013). Familiarity has also been yielded as a potential trigger of pupil reactivity and visual attention to faces in children with ASD (Nuske et al., 2014). These results suggest that attention to faces is not completely impaired in children with ASD, as it can be modulating considering some variables. Regarding the attention to the core facial features, specific results related to the eyes and the mouth are still inconsistent when considering studies independently; however, sound current reviews agree that children with ASD show atypical face processing, which means that they scan faces differently than TD children (Black et al., 2017; Harms et al., 2010; Sasson, 2006). This atypicality has been found in toddlers with ASD as young as two years (Jones et al., 2008), which have led some researchers to suggest that these children may have reduced their social learning input from early ages, which impact on their brain specialization and, consequently, on their social development

at later ages (Black et al., 2017; Jones et al., 2008; Klin, 2008; Sasson, 2006; Sasson et al., 2008; Sasson et al., 2011; Schultz, 2005).

#### 5.1.1 The Role of Competing Objects in Social Attention

The saliency of competing non-social objects plays a crucial role when assessing SA in children with ASD (Sasson et al., 2008; Sasson et al., 2011; Sasson & Touchstone, 2014). It has been suggested that the restricted and recurring bias to non-social elements reported in this population may be related to the same cognitive mechanisms involved in the repetitive behaviors associated to this disorder, mainly the circumscribed interests (CI; Pierce & Courchesne, 2001; Sasson et al., 2011; Sasson & Touchstone, 2014). Despite the relevance of the topic, few studies have addressed the role of competing objects and their interaction with social stimuli in SA in children with ASD. A restricted looking behavior toward CI-related objects (CIO, not to non-related ones or to social stimuli) has been found in young children (Sasson et al., 2011) and in school-aged children with ASD (Sasson et al., 2008). This behavior was described as circumscribed, perseverative, and detail oriented in both studies.

Additionally, Sasson and Touchstone (2014) assessed SA in young children with ASD (age range 24-62 months) who performed a paired preference task where faces with different emotions (happiness, sadness, fear, anger, neutral) were paired with CIO and non-CI-related objects (non-CIO). No difference between emotions was found in this study but their findings revealed that children with ASD showed the same attentional bias towards faces as their TD peers when the competing non-social objects were unrelated to their CI (non-CIO), thus only CIOs were able to capture their attention more than faces.

Given that the presence of CI is a hallmark of ASD and that this restricted looking behavior seems to be also specific of the autistic phenotype, the effect of the saliency of competing non-social objects on SA may help identify children with ASD at earlier ages.

## 5.1.2 Aims and Hypotheses

Taken together, previous results showed the high potential of studies on SA in this population as they have yielded some specific markers of the autism spectrum. Thus, the particular role of CIO in SA in children with ASD may also contribute to early diagnosis. Following this rationale and the approach applied in Sasson and Touchstone (2014), the

aim of this study was to compare young children with ASD and their TD peers in terms of SA assessing the role of facial emotions and competing objects in both groups. As in Sasson and Touchstone (2014), we designed an eye tracking paired preference task where a social stimulus (happy, angry, or neutral face) and a non-social one (CIO or non-CIO) competed for attracting participants' SA. This experiment was based on the approach applied in Sasson and Touchstone (2014) in terms of the application of the paired preference paradigm and the eye tracking dependent variables definition; however, both studies differs in aspects of design such as the number of emotions considered (the original research comprised happiness, sadness, anger, fear, and neutral, while this study included happiness, anger, and neutral), the manipulation of emotion intensity (Sasson and Touchstone manipulated that variable, while in this experiment intensity was not considered), and the number of trials per condition (the original study included one trial, while we repeated each condition three times).

Based on previous studies, we hypothesized that young children with ASD would show significantly reduced attention to faces compared with objects, which would differentiate them from the TD group (H1). We included faces with different emotional expressions (happiness, anger, and neutral) to see the effect of emotionality in SA. Thus, we expected that both groups would pay more attention to emotional faces, particularly to happy faces (H2), due to the reported emotional sensitivity (Nuske et al., 2014; Vivanti et al., 2011) and bias toward positive emotions in children with ASD (Farran et al., 2011; Li et al., 2011; Uljarevic & Hamilton, 2013). This would imply that positive emotions had an attracting effect which could be relevant to consider for clinical practice. We also predicted that young children with ASD would pay atypically less attention to faces when they competed with CIO, but typical attention when they competed with non-CIO (H3). Finally, we expected to find a significant bias toward CIO with respect to non-CIO in both groups (H4).

# 5.2 Method

# 5.2.1 Participants

Thirty-eight preschoolers: 19 with ASD (18 boys, 1 girl;  $M_{age} = 55.89$  months,  $SD_{age} = 9.40$ , range = 44-72 months) and 19 TD (18 boys, 1 girl;  $M_{age} = 53.53$  months,  $SD_{age} = 9.28$ , range = 41-72 months) participated in this study. This sample size could

detect between-groups differences with 85% of power and an effect size of 0.5, according to a sensitivity analysis in GPower 3.1.9.7 (Faul et al., 2007; Faul et al., 2009). The clinical group was recruited from centers of early childhood intervention in the province of Córdoba. For the clinical group, inclusion criteria comprised (1) The attendance to a center of early childhood intervention, (2) Having received a thorough assessment by a licensed experienced clinician who had determined the presence of ASD according to the DSM-5 criteria and following the protocol of the Infant Mental Health program at a community mental health service, and (3) The absence of any mental or medical condition.

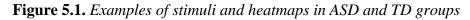
Children of the TD group were recruited from the preschool classes at a public school in Córdoba. Inclusion criteria for this group encompassed not having a history of developmental disorder either now or in the past, as well as the gender and chronological age matching with the clinical group. As in Sasson and Touchstone (2014), we matched groups on chronological age instead of on developmental age due to the low demand of the task applied, as passive-observational tasks do not require high-level cognitive abilities. We also matched groups on gender to control its potential effect on CI (Cho et al., 2017; DeLoache et al., 2007; Sasson et al., 2012; Sasson & Touchstone, 2014).

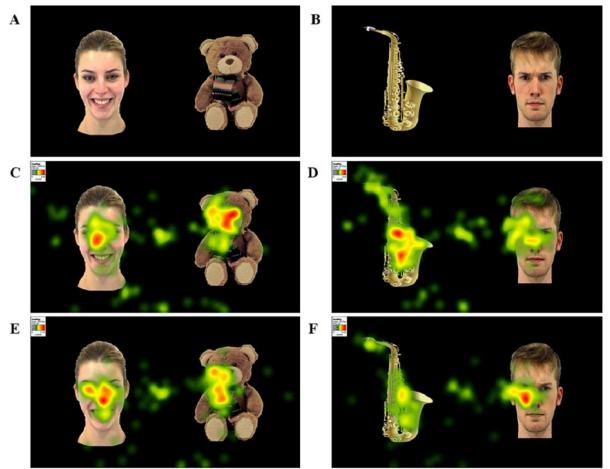
#### 5.2.2 Stimuli

Following the approach of Sasson and Touchstone (2014), the eye tracking paired preference task consisted of pairing social and non-social images to assess visual attention patterns for faces and objects. We paired faces displaying three different emotions (happiness, anger, and neutral) with two types of objects (CIO and non-CIO), which yielded a total of six experimental conditions that were repeated six times using different facial identities (a total of 36 trials). The gender of the faces and their location in the screen were counterbalanced to avoid potential effects of both variables. Thus, 36 pictures from 12 different identities (the same identities were taken for the three emotions, but these were not repeated for the same emotion) were taken from the Amsterdam Dynamic Facial Expression Set (ADFES; Van der Schalk et al., 2011). Faces were paired with 36 images of objects (18 CIO and 18 non-CIO, see Figures 5.1.A and 5.1.B). Some of these images were of our own creation by taking pictures of ordinary toys and puzzles used in the Early Childhood Intervention Center associated to the University of Córdoba, while others were taken from the Pixabay website and free of copyright under the Creative

Chapter 5. Visual Preference for Social Vs. Non-Social Images in Young Children with Autism Spectrum Disorders. An Eye Tracking Study

Commons CC0 license. Inclusion criteria for CIO were based on previous studies on CIrelated topics and objects in ASD (Sasson et al., 2008; Sasson & Touchstone, 2014; South et al., 2005). Thus, CIO belonged to the categories of toys, puzzles, means of transport, animals, and blocks, while non-CIO were plants, furniture, musical instruments, tools, school material, and clothes.





(A) Example stimulus pairing a happy face (F01-Joy-Apex) and a CIO. (B) Example stimulus pairing a non-CIO and an angry face (M02-Anger-Apex). (C and D). Heatmaps of the ASD group performance. (E and F) Heatmaps of the TD group performance.

Pairs were designed using PowerPoint to readjust images size (to 15 x 15 cm approximately), control the screen location, and remove pictures background to avoid contrast with the black background. The background removal forced us to rescale all pictures so that pairs looked similar in size. Final slides were resized to 1500 x 843 pixels to be correctly displayed in the laptop used to assess children and presentation order was counterbalanced.

## 5.2.3 Procedure

This study had the approval of the Research Ethics Committee of Córdoba. We contacted with potential candidate centers of early childhood intervention and those interested in participating distributed the project information and the written informed consent between families. Once we collected signed written informed consent from parents or guardians of children, we scheduled the assessment sessions according to families' and centers' availability. The assessment sessions took place in children's natural environment using a quiet room without distractions. In most cases, children were alone with the researcher during the assessment; however, there were some cases in which any parent was present, either for child requirement or for their own request. Assessment sessions were carried out in one single session in all cases, although the option of dividing the evaluation in two sessions had also been offered. The entire process lasted 45 minutes approximately. First, children performed the paired preference task, which lasted 3.6 minutes. Eye tracking data were recorded using Tobii X2-30 (Tobii Technology AB, Stockholm, Sweden) which tracks eye movements at a sampling rate of 30 Hz with a spatial accuracy of 0.4°. Children were seated at a deemed distance of 60 cm and 36° above a 15'screen of a laptop. They were given no other instruction but to look at the screen. The task started with a nine-points calibration with an animated stimulus as target. Calibration process was repeated in those cases where a child failed to complete it. After calibration, the task consisted of visualizing the set of 36 slides showing one face and one object. Each slide was displayed for 5 s. Prior to the presentation of each slide, children viewed an animated fixation point (a cartoon) for 1 s. to drive their attention to the center of the screen. After the eye tracking task, we assessed receptive vocabulary, affect recognition, and Theory of Mind (ToM) as described in the next section.

# 5.2.4 Measures

Along with the eye tracking task, receptive vocabulary, affect recognition, and Theory of Mind (ToM) abilities were assessed using the Peabody Picture Vocabulary Test-Third Edition (PPTV-III; Dunn & Dunn, 1997) and the two subtests comprised in the social perception domain of the Developmental Neuropsychological Assessment (NEPSY-II; Korkman et al., 2007) to analyze potential correlations among these domains and visual attention patterns in the sample. These scales were chosen due to their wide usability for clinical and research purposes, and the fact that both yield typification data with special groups (including ASD).

PPTV-III is a screening test which assesses vocabulary comprehension in individuals from 2.6 to 90 years. Subjects are presented with an array of four pictures and are asked to choose which one fits to a given word. Individuals are allowed to point the finger at their answer to reduce expressive language demands.

NEPSY-II is a neuropsychological assessment battery which comprises 32 tests to assess six domains (attention and executive functioning, language, memory and learning, sensorimotor, social perception, and visuospatial processing) in children and adolescents from 3 to 16 years. In the present study, only social perception domain was assessed. This domain encompasses affect recognition and ToM. Some participants of the ASD group were not able to perform either any of the two tasks or only the ToM part because of their age or the high linguistic demand required to pass these tasks.

Following the approach of Sasson and Touchstone (2014), we defined three indexed as dependent variables: (1) Prioritization, time to first fixation to faces to assess attentional orientation, (2) Preference, the proportion of total fixation duration on each kind of stimuli to compare the distribution of visual attention between faces regarding the competing object, and (3) Duration, mean time per visit to faces to describe the gaze maintenance (sustained attention) in facial stimuli. Statistical tests were performed using the Statistical Package for the Social Sciences, 25 (SPSS, 25, IBM, Armonk, NY, United States of America).

## 5.2.5 Data analysis

To analyze visual attention pattern to faces in both groups, we conducted separate repeated measures ANOVAs on each dependent variable (Prioritization, Preference, and Duration), with Group (ASD, control) as the between-group factor and Type of Object (CIO, non-CIO) as the within-group factor. Likewise, to examine visual attention pattern to objects in each group, we again performed separate repeated measures ANOVAs on each dependent variable (Prioritization, Preference, and Duration), with Group (ASD, control) as the between-group factor and Type of Object (CIO, non-CIO) as the within-group factor and Type of Object (CIO, non-CIO) as the within-group factor and Type of Object (CIO, non-CIO) as the within-group factor. As groups significantly differed in receptive vocabulary (see Table 5.1), PPVT-III standard scores were tentatively included as covariates, but preliminary

analyses yielded no effect, therefore this measure was dropped from the final analyses. Effect sizes were also calculated for repeated measures ANOVAs (partial eta-squared,  $\eta_p^2$ ) considering small (< .01), medium (< .06), or large (< .14) effects. Finally, correlation analyses were carried out to check potential relationships between visual attention pattern and PPVT-III or NEPSY-II scores in any group. Thus, Pearson correlation coefficient was used for PPVT-III scores, while Spearman correlation coefficient was used for NEPSY-II scores as this measure did not fulfil requirements for performing parametric tests.

			ASD	TD		
			(n = 19) $(n = 19)$			
			M (SD)	M (SD)	<b>F</b> (1,	Р
					37)	
	PPV	Г-III Raw Score	43.05	50.21 (18.73)	1.33	.257
			(19.54)			
	<b>PPVT-III Standard Score</b>		94.00	107.47	4.33	.045*
			(25.85)	(11.38)		
			n	n	$\chi^2$	Р
NEPS	NEPSY-II Total Raw Score Affect Recognition		7	19	4.03	.045*
	NEPSY-II Verbal ToM NEPSY-II Contextual ToM		6	19	7.55 1.15	.006* .283
			6	19		
	NEPSY-II	Total Raw Score ToM	6	19	8.27	.004*
			ASD	(n = 19)	TD (1	n = 19)
			М	(SD)	M	(SD)
		TFF_Hapvs.CIO	1198 (385)		1077 (421)	
(SI		TFF_Hapvs.non-CIO	957	7 (464)	878 (403)	
n) no	S	TFF_Angvs.CIO	1287 (420)		969 (395)	
Prioritization (ms)	Faces	TFF_Angvs.non-CIO	956 (392)		814 (415)	
		TFF_Neuvs.CIO	123	1 (544)	1076 (551)	
. –		TFF_Neuvs.non-CIO		950 (452)		

 Table 5.1. Descriptive characteristics of variables of the study

TFF_Facvs.non-CIO954 (290)827 (262)TFF_CIOvs.Hap933 (551)937 (365)TFF_non-CIOvs.Hap735 (351)961 (344)TFF_CIOvs.Ang752 (427)758 (312)TFF_non-CIOvs.Ang830 (370)1079 (392)TFF_CIOvs.Neu779 (425)754 (342)TFF_TotalCIO820 (369)816 (215)TFF_Totalnon-CIO753 (207)1107 (236)PTFD_Hapvs.CIO14 (11)177 (4)PTFD_Hapvs.CIO14 (7)177 (6)PTFD_Neuvs.CIO16 (9)19 (6)PTFD_Neuvs.CIO10 (5)16 (3)PTFD_Facvs.CIO38 (13)49 (10)PTFD_Facvs.CIO38 (13)49 (10)PTFD_Facvs.CIO38 (13)49 (10)PTFD_Facvs.CIO38 (13)49 (10)PTFD_Facvs.CIO38 (13)49 (10)PTFD_Facvs.CIO38 (13)49 (10)PTFD_CIOvs.Hap17 (7)13 (5)PTFD_CIOvs.Ang19 (7)15 (4)PTFD_CIOvs.Neu21 (7)14 (6)PTFD_TotalCIO62 (13)51 (10)PTFD_TotalCIO53 (15)41 (11)TFV_Hapvs.CIO638 (214)992 (317)TOTTTV_Hapvs.CIO651 (315)980 (405)			TEE Form CIO	1220 (221)	1041 (262)	
TFF_CIOvs.Hap         933 (551)         937 (365)           TFF_non-CIOvs.Hap         735 (351)         961 (344)           TFF_CIOvs.Ang         752 (427)         758 (312)           TFF_non-CIOvs.Ang         830 (370)         1079 (392)           TFF_CIOvs.Neu         779 (425)         754 (342)           TFF_TotalCIO         820 (369)         816 (215)           TFF_TotalCIO         820 (369)         816 (215)           TFF_Totalnon-CIO         753 (207)         1107 (236)           PTFD_Hapvs.CIO         14 (11)         17 (4)           PTFD_Hapvs.CIO         14 (11)         17 (6)           PTFD_Neuvs.non-CIO         16 (9)         19 (6)           PTFD_Neuvs.CIO         10 (5)         16 (3)           PTFD_Neuvs.CIO         14 (4)         19 (6)           PTFD_Neuvs.CIO         10 (5)         16 (3)           PTFD_Neuvs.non-CIO         14 (4)         19 (6)           PTFD_Facvs.CIO         38 (13)         49 (10)           PTFD_Facvs.CIO         16 (5)         16 (3)           PTFD_CIOvs.Hap         22 (6)         18 (4)           PTFD_CIOvs.Ang         19 (7)         13 (5)           PTFD_CIOvs.Neu         21 (7)         18 (5)			TFF_Facvs.CIO	1239 (321)	1041 (362)	
Image: constraint of the system of the s			TFF_Facvs.non-CIO	954 (290)	827 (262)	
Market State     TFF_CIOvs.Ang     752 (427)     758 (312)       TFF_non-CIOvs.Ang     830 (370)     1079 (322)       TFF_CIOvs.Neu     779 (425)     754 (342)       TFF_non-CIOvs.Neu     696 (395)     1281 (388)       TFF_TotalCIO     820 (369)     816 (215)       TFF_Totalnon-CIO     753 (207)     1107 (236)       PTFD_Hapvs.CIO     14 (11)     17 (4)       PTFD_Hapvs.CIO     14 (11)     17 (6)       PTFD_Angvs.non-CIO     16 (9)     19 (6)       PTFD_Angvs.CIO     14 (7)     17 (6)       PTFD_Angvs.CIO     10 (5)     16 (3)       PTFD_Neuvs.CIO     10 (5)     16 (3)       PTFD_Neuvs.CIO     14 (4)     19 (6)       PTFD_Facvs.CIO     38 (13)     49 (10)       PTFD_Facvs.CIO     38 (13)     49 (10)       PTFD_Facvs.CIO     38 (13)     49 (10)       PTFD_CIOvs.Hap     22 (6)     18 (4)       PTFD_CIOvs.Ang     19 (7)     15 (4)       PTFD_non-CIOvs.Ang     15 (6)     14 (6)       PTFD_non-CIOvs.Neu     21 (7)     18 (5)       PTFD_TotalCIO     62 (13)     51 (10)       PTFD_TotalCIO     53 (15)     41 (11)       PTFD_TotalCIO     53 (15)     41 (11)			TFF_CIOvs.Hap	933 (551)	937 (365)	
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Vertical         PTFD_Neuvs.CIO         10 (5)         16 (3)           PTFD_Neuvs.non-CIO         14 (4)         19 (6)           PTFD_Facvs.CIO         38 (13)         49 (10)           PTFD_Facvs.CIO         38 (13)         49 (10)           PTFD_Facvs.non-CIO         47 (15)         59 (11)           PTFD_CIOvs.Hap         22 (6)         18 (4)           PTFD_non-CIOvs.Hap         17 (7)         13 (5)           PTFD_CIOvs.Ang         19 (7)         15 (4)           PTFD_CIOvs.Neu         21 (7)         18 (5)           PTFD_TotalCIO         62 (13)         51 (10)           PTFD_Totalnon-CIO         53 (15)         41 (11)           TPV_Hapvs.CIO         638 (214)         992 (317)			PTFD_Angvs.CIO	14 (7)	17 (6)	
PTFD_Neuvs.non-CIO         14 (4)         19 (6)           PTFD_Facvs.CIO         38 (13)         49 (10)           PTFD_Facvs.non-CIO         47 (15)         59 (11)           PTFD_CIOvs.Hap         22 (6)         18 (4)           PTFD_non-CIOvs.Hap         17 (7)         13 (5)           PTFD_CIOvs.Ang         19 (7)         15 (4)           PTFD_CIOvs.Neu         21 (7)         18 (5)           PTFD_non-CIOvs.Neu         21 (7)         14 (6)           PTFD_TotalCIO         62 (13)         51 (10)           PTFD_Totalnon-CIO         53 (15)         41 (11)		s	PTFD_Angvs.non-CIO	17 (9)	22 (5)	
Vỹ         PTFD_Facvs.CIO         38 (13)         49 (10)           PTFD_Facvs.non-CIO         47 (15)         59 (11)           PTFD_CIOvs.Hap         22 (6)         18 (4)           PTFD_non-CIOvs.Hap         17 (7)         13 (5)           PTFD_CIOvs.Ang         19 (7)         15 (4)           PTFD_non-CIOvs.Ang         15 (6)         14 (6)           PTFD_CIOvs.Neu         21 (7)         18 (5)           PTFD_TotalCIO         62 (13)         51 (10)           PTFD_Totalnon-CIO         53 (15)         41 (11)		Face	PTFD_Neuvs.CIO	10 (5)	16 (3)	
PTFD_Facvs.non-CIO         47 (15)         59 (11)           PTFD_CIOvs.Hap         22 (6)         18 (4)           PTFD_non-CIOvs.Hap         17 (7)         13 (5)           PTFD_CIOvs.Ang         19 (7)         15 (4)           PTFD_non-CIOvs.Ang         15 (6)         14 (6)           PTFD_CIOvs.Neu         21 (7)         18 (5)           PTFD_TotalCIO         62 (13)         51 (10)           PTFD_Totalnon-CIO         53 (15)         41 (11)           TPV_Hapvs.CIO         638 (214)         992 (317)			PTFD_Neuvs.non-CIO	14 (4)	19 (6)	
E         PTFD_non-CIOvs.Hap         17 (7)         13 (5)           PTFD_CIOvs.Ang         19 (7)         15 (4)           900         PTFD_non-CIOvs.Ang         15 (6)         14 (6)           PTFD_CIOvs.Neu         21 (7)         18 (5)           PTFD_non-CIOvs.Neu         21 (7)         14 (6)           PTFD_TotalCIO         62 (13)         51 (10)           PTFD_Totalnon-CIO         53 (15)         41 (11)           TPV_Hapvs.CIO         638 (214)         992 (317)			PTFD_Facvs.CIO	38 (13)	49 (10)	
E         PTFD_non-CIOvs.Hap         17 (7)         13 (5)           PTFD_CIOvs.Ang         19 (7)         15 (4)           PTFD_non-CIOvs.Ang         15 (6)         14 (6)           PTFD_CIOvs.Neu         21 (7)         18 (5)           PTFD_non-CIOvs.Neu         21 (7)         14 (6)           PTFD_TotalCIO         62 (13)         51 (10)           PTFD_Totalnon-CIO         53 (15)         41 (11)           TPV_Hapvs.CIO         638 (214)         992 (317)	ce (%		PTFD_Facvs.non-CIO	47 (15)	59 (11)	
E         PTFD_non-CIOvs.Hap         17 (7)         13 (5)           PTFD_CIOvs.Ang         19 (7)         15 (4)           900         PTFD_non-CIOvs.Ang         15 (6)         14 (6)           PTFD_CIOvs.Neu         21 (7)         18 (5)           PTFD_non-CIOvs.Neu         21 (7)         14 (6)           PTFD_TotalCIO         62 (13)         51 (10)           PTFD_Totalnon-CIO         53 (15)         41 (11)           TPV_Hapvs.CIO         638 (214)         992 (317)	leren		PTFD_CIOvs.Hap	22 (6)	18 (4)	
PTFD_non-CIOvs.Ang         15 (6)         14 (6)           PTFD_CIOvs.Neu         21 (7)         18 (5)           PTFD_non-CIOvs.Neu         21 (7)         14 (6)           PTFD_TotalCIO         62 (13)         51 (10)           PTFD_Totalnon-CIO         53 (15)         41 (11)           TPV_Hapvs.CIO         638 (214)         992 (317)			PTFD_non-CIOvs.Hap	17 (7)	13 (5)	
PTFD_CIOvs.Neu         21 (7)         18 (5)           PTFD_non-CIOvs.Neu         21 (7)         14 (6)           PTFD_TotalCIO         62 (13)         51 (10)           PTFD_Totalnon-CIO         53 (15)         41 (11)           TPV_Hapvs.CIO         638 (214)         992 (317)			PTFD_CIOvs.Ang	19 (7)	15 (4)	
PTFD_non-CIOvs.Neu       21 (7)       14 (6)         PTFD_TotalCIO       62 (13)       51 (10)         PTFD_Totalnon-CIO       53 (15)       41 (11)         TPV_Hapvs.CIO       638 (214)       992 (317)		cts	PTFD_non-CIOvs.Ang	15 (6)	14 (6)	
PTFD_TotalCIO         62 (13)         51 (10)           PTFD_Totalnon-CIO         53 (15)         41 (11)           TPV_Hapvs.CIO         638 (214)         992 (317)		Objec	PTFD_CIOvs.Neu	21 (7)	18 (5)	
PTFD_Totalnon-CIO         53 (15)         41 (11)           TPV_Hapvs.CIO         638 (214)         992 (317)			PTFD_non-CIOvs.Neu	21 (7)	14 (6)	
<b>TPV_Hapvs.CIO</b> 638 (214) 992 (317)			PTFD_TotalCIO	62 (13)	51 (10)	
			PTFD_Totalnon-CIO	53 (15)	41 (11)	
TPV_Hapvs.non-CIO         651 (315)         980 (405)           TPV_Angvs.CIO         646 (323)         986 (384)			TPV_Hapvs.CIO	638 (214)	992 (317)	
<b>TPV_Angvs.CIO</b> 646 (323) 986 (384)	ion	ICES	TPV_Hapvs.non-CIO	651 (315)	980 (405)	
	Durat	Fa	TPV_Angvs.CIO	646 (323)	986 (384)	

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	TPV_Angvs.non-CIO	728 (292)	1106 (322)		
	TPV_Neuvs.CIO	545 (208)	802 (197)		
	TPV_Neuvs.non-CIO	669 (453)	958 (340)		
	TPV_Facvs.CIO	615 (171)	912 (235)		
	TPV_Facvs.non-CIO	655 (201)	1016 (274)		
	TPV_CIOvs.Hap	1203 (768)	965 (336)		
	TPV_non-CIOvs.Hap	1222 (1504)	697 (313)		
	TPV_CIOvs.Ang	1480 (2779)	745 (250)		
cts	TPV_non-CIOvs.Ang	1578 (3382)	792 (344)		
Objects	TPV_CIOvs.Neu	1335 (1233)	927 (283)		
	TPV_non-CIOvs.Neu	1144 (1145)	816 (295)		
	TPV_TotalCIO	1157 (896)	858 (211)		
	TPV_Totalnon-CIO	1128 (1426)	761 (225)		

PPVT-III Raw Score and Standard Score variables were tested using ANOVA. NEPSY-II scores were analyzed using H-Kruskal-Wallis

TFF, Time to First Fixation; PTFD, Proportion of Total Fixation Duration; TPV, Time per Visit

## 5.3 Results

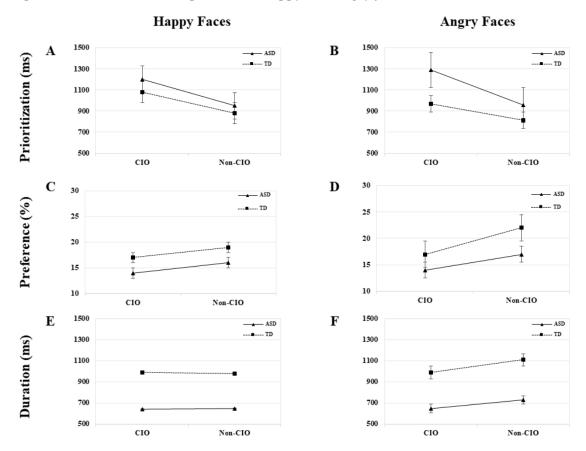
## 5.3.1 Descriptive Characteristics of Variables of the Study

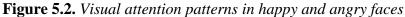
All participants assessed were included in the statistical analysis as the eye tracker apparatus collected more than 50% of their fixations, which is a standard cut-point applied in many studies (e.g., Franchini, Glaser, Gentaz et al., 2017; Franchini, Glaser, Wood de Wilde et al., 2017; Pierce et al., 2011; Pierce et al., 2016; Shi et al., 2015). Thus, all the 38 children were suitable for the analysis. Sample characteristics are summarized in Table 5.1. No significant differences were found between groups in age or gender (p > .05). However, ASD and TD groups differed in terms of vocabulary comprehension (PPVT-III standard scores), emotion recognition abilities (NEPSY-II Total Score Affect Recognition) and ToM (NEPSY-II Verbal ToM and Total Score ToM).

#### 5.3.2 Social Attention

#### 5.3.2.1 Prioritization

No effect for group × type of object interaction was found in any emotion. A main effect for group was only found in angry faces ( $F_{(1, 37)} = 4.54$ , p < .04,  $\eta_p^2 = .11$ ), indicating that children with ASD looked at angry faces significantly later than TD children (see Figure 5.2.B). A main effect for type of object was significant in all emotions independently: happiness ( $F_{(1, 37)} = 5.17$ , p < .03,  $\eta_p^2 = .13$ ), anger ( $F_{(1, 37)} = 10.45$ , p < .00,  $\eta_p^2 = .23$ ), neutral ( $F_{(1, 37)} = 6.47$ , p < .02,  $\eta_p^2 = .15$ ), and in total faces ( $F_{(1, 37)} = 22.38$ , p < .00,  $\eta_p^2 = .38$ ), implying that all participants looked at faces paired with non-CIOs earlier than those paired with CIOs (see Figures 5.2.A, 5.2.B, and 5.3.A).





#### 5.3.2.2 Preference

No group × type of object interaction effect was yielded in any particular emotion or total faces, but a main effect for group was found in angry ( $F_{(1, 37)} = 7.40, p < .01, \eta_p^2$ 

= .17), neutral ( $F_{(1, 37)} = 14.78$ , p < .00,  $\eta_p^2 = .29$ ), and total faces ( $F_{(1, 37)} = 10.29$ , p < .00,  $\eta_p^2 = .22$ ), indicating that children with ASD attended to angry and neutral faces substantially less than their TD peers (see Figures 5.1.D, 5.1.F, 5.2.D, and 5.3.C). A main effect for type of object also emerged in angry ( $F_{(1, 37)} = 5.97$ , p < .02,  $\eta_p^2 = .14$ ), neutral ( $F_{(1, 37)} = 16.83$ , p < .00,  $\eta_p^2 = .32$ ), and total faces ( $F_{(1, 37)} = 31.57$ , p < .00,  $\eta_p^2 = .47$ ), implying that both groups paid significantly more attention to angry and neutral faces paired with non-CIOs than those paired with CIOs. Any effect was found in happy faces, which suggests that both groups looked at these faces the same amount of time regardless of the type of object (see Figures 5.1.C, 5.1.E, and 5.2.C).

#### 5.3.2.3 Duration

No group × type of object interaction effect was highlighted in any specific emotion or total faces, but a main effect for group was found in all emotions: happy ( $F_{(1, 37)} = 13.66$ , p < .00,  $\eta_p^2 = .28$ ), angry ( $F_{(1, 37)} = 14.41$ , p < .00,  $\eta_p^2 = .29$ ), neutral ( $F_{(1, 37)} = 11.80$ , p < .00,  $\eta_p^2 = .25$ ), and total faces ( $F_{(1, 37)} = 24.08$ , p < .00,  $\eta_p^2 = .40$ ), indicating that children with ASD made significantly shorter visits to all type of faces than their TD peers. A main effect for type of object was also found in neutral ( $F_{(1, 37)} = 4.62$ , p < .04,  $\eta_p^2 = .11$ ), and total faces ( $F_{(1, 37)} = 6.59$ , p < .02,  $\eta_p^2 = .16$ ). This implies that the lack of facial emotion drove both groups to pay more attention to faces paired with non-CIOs than those paired with CIOs; otherwise, when faces displayed any positive or negative emotion, the type of object had no impact in their sustained attention (see Figures 5.2.E, 5.2.F, and 5.3.E).

#### 5.3.3 Object Attention

#### 5.3.3.1 Prioritization

Significant effects were found for group × type of object interaction ( $F_{(1, 37)} = 12.85, p < .00, \eta_p^2 = .26$ ), group ( $F_{(1, 37)} = 6.26, p < .02, \eta_p^2 = .15$ ), and type of object ( $F_{(1, 37)} = 5.08, p < .03, \eta_p^2 = .12$ ), indicating that, relative to TD, children with ASD were significantly faster at focusing on non-CIOs (see Figure 5.3.B). These effects were replicated only when objects were paired with neutral faces, but not when these were paired with happy or angry faces, implying that both groups showed similar orientation to objects when these competed with emotional faces.

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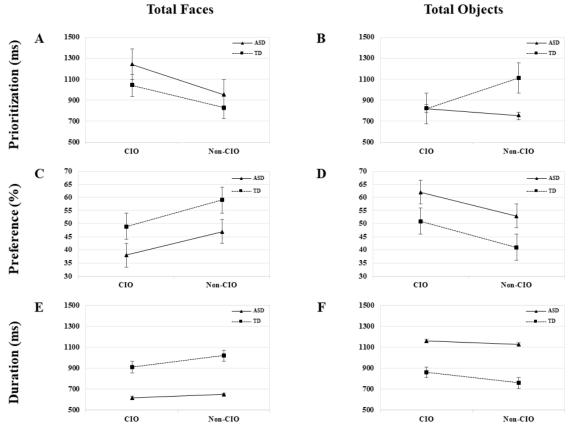


Figure 5.3. Visual attention patterns in total faces and objects Total Faces

#### 5.3.3.2 Preference

No group × type of object interaction was found in this index, but a main effect emerged for group ( $F_{(1, 37)} = 10.29$ , p < .00,  $\eta_p^2 = .22$ ), indicating that children with ASD looked at objects substantially longer than their counterparts (see Figure 5.3.D). An effect for type of object was also found ( $F_{(1, 37)} = 31.57$ , p < .00,  $\eta_p^2 = .47$ ), implying that both groups spent more time viewing CIOs than non-CIOs. When considering emotions, these effects were replicated only for happy and neutral faces, but these were marginal for angry faces, which suggests that children with ASD showed typical preference for objects when they competed with angry faces.

#### 5.3.3.3 Duration

No significant effect was found at any level in this index, indicating that children with ASD as well as TD made similar visits to objects, regardless of its typology (see Figure 5.3.F).

#### 5.3.4 Correlation analysis

To explore whether the visual attention pattern found in each group was related to their performance in the PPVT-III and the NEPSY-II, we conducted correlation analyses. No significant correlation between PPVT-III and any dependent variable was found in any group. Only Spearman correlation coefficient yielded significant values (see Table 5.2) for Affect Recognition Total Raw Score and prioritization of happy faces ( $r_{s(7)} =$ .775, p = .04) in the ASD group. For the TD group, a significant correlation was found in Affect Recognition Total Raw Score and TFF to CIO ( $r_{s(19)} = .588$ , p = .01). This data indicates that: (1) The higher Affect Recognition scores children with ASD have, the later they look at happy faces; (2) The higher Affect Recognition scores TD children have, the later they focus on CIO.

		$\mathbf{ASD}\;(\mathbf{n}=7)$			<b>TD</b> ( <b>n</b> = <b>19</b> )				
		TRSAF	VT	СТ	TRST	TRSAF	VT	СТ	TRST
	TFF Happy Faces	.775*	.309	.463	.486	251	350	211	376
tion	TFF Angry Faces	.054	.093	802	200	027	030	129	070
Prioritization	TFF Neutral Faces	.355	.313	204	.232	.171	131	129	154
Prio	TFF CIO	.487	.278	.617	.486	.588**	.329	.303	.341
	TFF non-CIO	.414	.278	.309	.371	.218	.124	.190	.371
	PTFD Happy Faces	545	309	.000	314	145	.100	.351	.226
e,	PTFD Angry Faces	255	517	125	493	.023	.086	.056	.054
Preference	PTFD Neutral Faces	093	751	.031	638	.238	049	.424	.086
Pre	PTFD CIO	.055	.494	.185	.486	092	151	295	191
	PTFD non-CIO	.577	.525	.309	.600	.000	053	267	153
	<b>TPV Happy Faces</b>	.036	278	.278	143	.044	.264	.200	.344
Duration	<b>TPV Angry Faces</b>	.270	.062	.031	.086	.287	.345	.054	.315
	<b>TPV Neutral Faces</b>	288	617	463	714	.379	.124	.207	.146
	TPV CIO	.306	.525	.000	.486	.105	115	.176	011
	TPV non-CIO	.360	.741	.309	.771	.178	.085	035	.080

Table 5.2. Spearman correlation between dependent variables and NEPSY-II indexes

TRSAF, Total Raw Score Affect Recognition; VT, Verbal ToM; CT, Contextual ToM; TRST, Total Raw Score ToM; TFF, Time to first fixation; PTFD, Proportion of total fixation duration; TPV, Time per visit Significance levels: \*\*\*p < 0.001; \*\*p < 0.01; and \*p < 0.05

## 5.4 Discussion

This study aimed at comparing SA in young children with ASD and TD peers analyzing the effect of competing facial emotions and type of objects in their visual attention pattern to faces and objects. Taken together, our results suggest that children with ASD display a visual attention pattern typical in terms of direction (early orientation, longer looking time, and more detailed exploration of faces paired with non-CIOs than those paired with CIOs, and visual preference for CIOs over non-CIOs), but quantitatively atypical. Thus, relative to TD children, children with ASD showed a lack of attention to faces and an excessive visual preference for objects in general.

Our results revealed that children with ASD looked at faces later, during less time and they made shorter visits than their TD peers. In light of these results, our first hypothesis was confirmed as children with ASD differed from TD children in reduced attention to faces. This statement aligns with many studies reporting reduced attention to social images in favor of non-social ones (Chevallier et al., 2015; Chita-Tegmark, 2016a, 2016b; Falck-Ytter et al., 2013; Guillon et al., 2014; Guillon et al., 2016; Nakano et al., 2010; Zantinge et al., 2017) and, particularly, decreased attention to faces (Franchini, Glaser, Gentaz et al., 2017; Vivanti et al., 2011; von Hofsten et al., 2009; Zantinge et al., 2017).

We also predicted different effects of competing facial emotions in children's visual behavior (H2). These differences were found in prioritization and preference indexes during the analysis of the visual attention pattern to faces, confirming emotional sensitivity in children with ASD. Thus, our ASD group looked at angry faces later than the control one, indicating late orientation to angry faces compared to controls. Moreover, happy faces were the only ones in which children with ASD displayed a typical visual preference behavior, which implies that happiness was relatively unimpaired in our ASD sample. Likewise, the effect of the type of object in sustained attention to faces, which suggests that the absence of emotion increases atypical visual behavior in this population. This emotional sensitivity was again replicated in the analysis of visual attention pattern to objects, as children with ASD only showed atypical visual orientation to objects when they competed with neutral faces, otherwise there were no differences between groups, type of object, or interaction. These assumptions are in line with those studies reporting

emotional sensitivity (Nuske et al., 2014; Vivanti et al., 2011) and highlighting that negative emotions are compromised in this population, while positive emotions are relatively intact (Evers et al., 2015; Farran et al., 2011; Li et al., 2011; Lozier et al., 2014; Uljarevic & Hamilton, 2013). This finding is worthy to be considered when designing and developing interventions. Hence, clinicians may avoid negative emotions (which drive to attention disengagement) and foster positive ones (which are attention-getters); this may help involve the child within the activity, increasing the effectiveness and potential benefits of interventions. Emotional sensitivity was not reported in Sasson and Touchstone (2014), this difference between studies could be due to the fact that we included more essays per condition, which may have contributed to uncover atypicalities in visual behavior to different emotional faces in the ASD group.

Regarding our H3, we expected to find a differential role of the type of object in young children with ASD, which would drive them to display less attention to faces competing with CIO, but typical attention when they competed with non-CIO. This hypothesis was refused in this study as children with ASD paid less attention to faces in both conditions. Moreover, TD children seemed to identify quickly the type of object they were looking at, as they focused on CIOs significantly earlier than on non-CIOs; however, children with ASD did not make this distinction, instead they fixated on both type of objects equally fast. Therefore, the type of object was not what made the greatest impact on SA in children with ASD, but the competing facial emotion was. This disagreement with conclusions from Sasson and Touchstone (2014) may rely on the age of participants in both studies (being ours older) or the fact that our participants had been receiving early intervention for a deemed mean time of 30 months (SD = 8 months), which implies a previous work on their potential social-emotional deficits. In this sense, future studies should address the effect of early diagnosis and intervention in SA in children with ASD.

We also found a powerful effect of CIOs over non-CIOs in both groups, this effect was found in the three indexes when analyzing visual attention pattern to faces, but only in prioritization and preference when considering the visual attention pattern to objects. The lack of differences in duration index during the analysis of visual attention pattern to objects could be partially explained by the fact that the ASD group was more heterogeneous in terms of duration and number of visits to both type of objects. This shows the wide heterogeneity of CI among children with ASD due to their idiosyncratic Chapter 5. Visual Preference for Social Vs. Non-Social Images in Young Children with Autism Spectrum Disorders. An Eye Tracking Study

feature. Due to the close relationship between the duration index and the visual processing style, we may say that children with ASD made less detailed processing of faces compared to their TD peers, but not a different visual processing of objects. Still, these data confirm our H4 suggesting that CIOs disrupt social attention in children with ASD as well as TD. This result is in line with a large body of literature on the catching role of CIOs in children with ASD and TD (Cho et al., 2017; DeLoache et al., 2007; Sasson et al., 2008; Sasson et al., 2011; Sasson et al., 2012; Sasson & Touchstone, 2014; South et al., 2005). Nevertheless, given the fact that children with ASD have difficulties engaging with faces, we think that clinicians should be cautious including CIOs in interventions as they can help engage the child in an activity, but they can also distract him/her from its real purpose.

On the other hand, no significant correlation between vocabulary comprehension or ToM and visual attention pattern was highlighted in any group. Only affect recognition was significantly related to prioritization of happy faces and CIO in the ASD group and TD group, respectively. This indicates that the more proficiency in affect recognition, the later children with ASD look at happy faces and TD children look at CIO. This outcome is reasonable in the case of TD children as affect recognition is closely related to social attention (Frith & Frith, 2006; Klin, 2008; Parish-Morris et al., 2013). Thus, more proficiency in affect recognition could imply diminishing attention to non-social stimuli in favor of social ones. However, the pattern found in ASD is surprising and cannot be explained based on existing literature; hence, it could stem from the small sample of children with ASD to whom NEPSY-II could be applied.

Finally, we consider some limitations of this study. First of all, the sample size was relatively small, although it is in line with that of related studies (Guillon et al., 2016; Jones et al., 2008; Liberati et al., 2017; Sasson & Touchstone, 2014; Vivanti et al., 2011; von Hofsten et al., 2009). Studies with larger samples may help to stress differences between groups and remark patterns within-group. Secondly, we controlled for receptive language, affect recognition, and ToM abilities, but not for expressive language, which would have been interesting given its key role in this population (Stagg et al., 2014). Moreover, the small number of children with ASD who could not be assessed with the social-emotions subscale of the NEPSY-II may have distorted results on correlations between affect

recognition and SA (Frith & Frith, 2006; Klin, 2008; Parish-Morris et al., 2013) as well as facial emotion recognition and ToM (Davidson et al., 2019; Trevisan & Birmingham, 2016) in this population. Alongside these limitations, it is important to be cautious when interpreting results of SA in children with ASD as some variables may impact on them. Among these variables, research has highlighted the following: (1) The stimulus type, being interactive and naturalistic stimuli those which better find differences between clinical and non-clinical groups (Chevallier et al., 2015; Klin et al., 2002b); (2) The social content of scenes, that is, the more people involved in the scene or the more scene complexity, the less SA deployed by the population with ASD (Chita-Tegmark, 2016b; Guillon et al., 2014; Shi et al., 2015); (3) The presence of speech during social interactions, which has been related to slower latencies and less social orientation to faces in children with ASD compared to TD (Magrelli et al., 2013); and (4) The language proficiency, with typical SA in children with ASD and normal language but reduced SA in those with language delayed (Stagg et al., 2014). For these reasons, futures studies must include larger sample sizes, a more thorough assessment of language profile of the clinical sample, as well as affect recognition and ToM measures, and more comparison groups to contribute to differential diagnosis.

## **5.5 Conclusions**

Children with ASD have demonstrated SA atypicalities which may impair their relationship with the social environment and, consequently, their social expertise from early ages. Current eye tracking research on SA has been aimed at identifying these atypicalities in young children, which has substantial repercussion in clinical practice. In this sense, the finding of specific visual attentional markers such as the less detailed processing of faces and the bias to non-social objects in the population with ASD are valuable from a diagnosis point of view, but also the emotional sensitivity found in these children is significant from an intervention perspective, as it may help practitioners to guide their interventions emphasizing the role of emotions, taking advantage of those more engaging for children with ASD, and avoiding the most repelling ones. The worthy applicability of this kind of studies alongside the widely reported advantages of early intervention for children future outcomes and development substantiate the need of continuing this line of research enriching it with larger samples, deeper cognitive assessments, and more comparison groups.

## Acknowledgements

We gratefully acknowledge to all families who agreed that their children took part in this study, as well as the Early Intervention Centers and the school which also accepted our request of participation very kindly.

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# Chapter 6. Social attention and autism in early childhood: Evidence on psychophysiological markers based on visual scanning of emotional faces with eye tracking methodology<sup>3</sup>

#### Abstract

Background: Children with autism spectrum disorders (ASD) show atypical scanning of faces, which has been suggested as an obstacle to social expertise, hindering their lifelong social relationships. Method: This study aimed at comparing social attention in young children with and without ASD (N = 34). We designed two eye tracking paired preference tasks to assess effects of emotional expression and poser's gender (Experiment 1) and poser's age (Experiment 2) on visual scanning pattern of faces in both groups. Results: Data analysis yielded three psychophysiological markers in the ASD group: (1) Late orientation to angry faces, (2) Late orientation to child faces, and (3) Superficial facial processing (shorter visits). Conclusions: Clinical and research implications are discussed regarding potential biomakers that could contribute to early diagnosis and intervention.

**Keywords:** Autism Spectrum Disorders, social attention, psychophysiological markers, emotions, eye tracking, early intervention.

#### 6.1 Introduction

Autism Spectrum Disorders (ASD) are defined as a heterogeneous set of neurodevelopmental and pervasive conditions with serious social and communication deficits as hallmarks (American Psychiatric Association [APA], 2013; Autism Europe [AE], 2015; Charman & Baird, 2002; Kjelgaard & Helen Tager-Flusberg, 2001; World Health Organization [WOS], 1992). The concept of spectrum involves a wide heterogeneity of the three core features among people with ASD: we can find individuals with ASD who develop functional language and good social competence, while other individuals in the spectrum present a more severe profile (e.g., they do not acquire functional language and their social competence is substantially poor). This heterogeneity

<sup>&</sup>lt;sup>3</sup> This chapter corresponds to a scientific article which is currently under review in a high impact scientific journal.

hinders early diagnosis and, consequently, early intervention, which are closely related to better prognosis as they allow the display of resources and appropriate support for a child at early ages, when the brain is still in development (AE, 2015; Bennet et al., 2014; Corsello, 2005; Mandell et al., 2005; Nicholas et al., 2009; Özerk, 2016; Zablotsky et al., 2017). Therefore, research as well as clinical practice require more data to detect the presence of the disorder as early as possible. Here, the analysis of social attention (SA, understood as the allocation of visual attention to social stimuli or socially relevant content of scenes, instead of non-social elements) has become a source of interest by recent research as it contributes to unveil specific psychophysiological markers of ASD. This has been possible partially due to the development and spread of new enabling approaches and methodologies such as eye tracking.

Eye tracking is a non-invasive methodology which let analyze individuals' looking behavior while they view a scene. It relies upon corneal reflection technology, which allows to track and monitor eye movements and, consequently, yields reliable psychophysiological markers (Aslin & McMurray, 2004; Di Stasi et al., 2013; Gredebäck et al., 2009; Hepach et al., 2015). It has been widely used in research on ASD as it has proved to be accurate in detecting subtle biases in visual emotion processing, as well as in unveiling the main mechanisms behind the decoding of facial emotional expressions, which is a significant area of interest for researchers in the field (Bölte et al., 2016; Falck-Ytter et al., 2013; Harms et al., 2010; Klin et al., 2002; Vacas et al., 2021a).

Most eye tracking studies on SA in children with ASD have reported an atypical visual scanning pattern (VSP) of social stimuli in this population. The VSP (also known as gaze or looking pattern) is defined as the sequence of looking shifts during images visualization. Its study provides relevant insight into the way children with ASD observe social images. The VSP of these children has been characterized by reduced attention to social stimuli or the social content of a scene, increased attention to non-social elements compared to their typically developing (TD) peers (Chevallier et al., 2015; Chita-Tegmark, 2016a, 2016b; Falck-Ytter et al., 2013; Frazier et al., 2017; Guillon et al., 2014; Guillon et al., 2016; Nakano et al., 2010; Vacas et al., 2021b; Zantinge et al., 2017), and scattered eye movements across the scene when facing social situations (Liberati et al., 2017). This particular VSP has been found in population with ASD since their first year

Similarly, attention to faces become crucial when analyzing SA, as these are important sources of information of others' inner states, enabling us to develop abilities such as mentalizing, emotion recognition and comprehension, and empathy among others (Frith & Frith, 2006; Klin, 2008). Furthermore, attention to faces has been found to emerge early in development as a natural mechanism which foster neural specialization and improve social functioning (Klin, 2008; Johnson, 2000; Nelson, 2001; Shultz, 2005). Children with ASD have demonstrated to pay less attention to faces than their TD peers (Franchini, Glaser, Gentaz et al., 2017; Vacas et al., 2021b; Vivanti et al., 2011; von Hofsten et al., 2009; Zantinge et al., 2017) and less orientation to faces have been found across the lifespan in people with ASD (Fletcher-Watson et al., 2009; Freeth et al., 2010; Harrop et al., 2018; Sasson et al., 2007; Sasson & Touchstone, 2014; Wieckowski & White, 2020). However, literature on specific facial features is inconclusive, with some studies highlighting reduced attention to the eyes and a strong bias toward the mouth in children with ASD, while other studies fail to find support for these theses (see Black et al., 2017; Harms et al., 2010; Papagiannopoulou et al., 2014; Vacas et al., 2021a for review). Recent reviews have concluded that this population presents atypical face processing, that is, individuals with ASD scan faces in a different way compared to TD population (Black et al., 2017; Harms et al., 2010; Sasson, 2006). This atypical VSP toward faces has been found in children with ASD from the age of two (Jones et al., 2008). This finding has led some researchers to propose that the lack of social input that children with ASD experience since their early infancy hinders their neural specialization which, subsequently, jeopardizes their social functioning later on (Jones et al., 2008; Klin, 2008; Sasson, 2006; Sasson et al., 2008; Sasson et al., 2011; Sasson & Touchstone, 2014; Shultz, 2005).

# 6.1.1 The Role of Emotion, Gender, and Age in Social Attention in Children with Autism Spectrum Disorders

To date, the role of emotion, poser's gender, and age on SA in children with ASD has been scarcely considered. Few studies have highlighted a bias toward emotionality versus neutrality in faces (Nuske et al., 2014, 2016; Vacas et al., 2021b; Vivanti et al., 2011) and some extent of sensitivity to positive and negative emotions (de Wit et al.,

2008) in this population. Thus, similar to their TD counterparts, children with ASD have shown sensitivity to emotion and familiarity when looking at faces. Emotional sensitivity implies that faces expressing emotions seem to catch their attention more than neutral ones (Nuske et al., 2014, 2016; Vacas et al., 2021b; Vivanti et al., 2011). Familiar faces also seem to activate their pupil reactivity and drive their attention to faces to a greater extent than unfamiliar ones (Nuske et al., 2014). Likewise, de Wit et al. (2008) found that children with ASD as their TD peers were able to distinguish between positive and negative facial expressions in terms of modifying their VSP according to the valence of the emotion. This means that both groups increased the time spent on the eye region when scanning faces with negative expressions. Despite this similarity, children with ASD still deploy less attention to the core areas of the face than their typical counterparts. This finding implies that even though there is a trend toward typicality in children with ASD regarding emotional sensitivity, there are still differences from their TD peers. Impairments in recognition of negative emotions have also been yielded in children with ASD (Harms et al., 2010; Li et al., 2011; Uljarevic & Hamilton, 2013), which could be related to their reduced VSP to faces according to these authors. However, Black et al. (2017) concluded that the effect of emotion on SA as well as its relationship with emotion recognition in children with ASD were still difficult to explain given current data. These inconsistencies, alongside some studies reporting similar VSP to faces (regardless of the emotional expression) between children with ASD and TD (Leung et al., 2013; Van der Geest et al., 2002), do not allow for drawing firm conclusions about the role of specific emotions on SA in children with ASD.

On the other hand, to our knowledge no study has so far addressed the effect of the poser's gender on SA in the population with ASD, while one study has tackled the own-age bias (potential advantage for processing peer faces, possibly due to a major exposure to them) in adolescents with ASD (Hauschild et al., 2020). In this study, the own-age bias was tested in terms of accuracy in facial emotion recognition, yielding a sound age sensitivity in adolescents with ASD as well as their TD peers. This means that both groups showed a major advantage in recognizing emotions in peer than in adult faces. However, participants' visual processing during the task performance was not tracked, thus no assumption could be made about the relationship between participants' emotion recognition performance and their visual behavior. Conversely, children with ASD are characterized by severe social difficulties, particularly with their peers (APA, effect of the poser's gender and age on SA in children with ASD relative to their peers and the possible contribution of these effects to the early identification of this clinical condition.

Previous studies have focused on analyzing the attention to specific facial features in children with ASD considering, sometimes, the role of different emotions; however, the study of visual processing of emotional faces deserves more detailed analysis. Given the potential of studies on this field and the lack of research on some aspects around it in children with ASD (e.g., visual orientation, preference, or engagement), the aim of the present paper was to compare SA in young children with and without ASD regarding the role of emotional expressions, the poser's gender, and the poser's age. To this end, we focused on VSP, which was categorized in terms of attentional orientation, visual preference, and sustained attention. Thus, we designed two eye tracking experiments following a paired preference approach to measure the effect of the emotional expression and poser's gender (Experiment 1) and the poser's age (Experiment 2) on VSP in both children with ASD and TD. The eye tracking paired preference approach consists of presenting participants with paired stimuli to assess different visual parameters and compare their visual behavior toward each one; it has been applied in several previous studies in the field (Harrop et al., 2018; Sasson & Touchstone, 2014; Unruh et al., 2016; Vacas et al., 2021b).

## 6.2 Experiment 1. Emotional Expression and Poser Gender

In this experiment, we paired happy, angry, and neutral female faces with the same emotions in male faces to determine whether the VSP differed between groups depending on the emotional expression (happy, angry, and neutral) or the poser's gender (female and male faces). Following the emotional sensitivity hypothesis in ASD (Nuske et al., 2014, 2016; Vacas et al., 2021b; Vivanti et al., 2011), both ASD and TD groups were expected to discriminate between emotional and neutral faces, orienting faster and deploying more SA to emotional than neutral ones (H1). We also aimed at finding differences in VSP to positive versus negative facial expressions; however, studies on emotion discrimination in children with ASD did not follow the paired preference approach but an emotion recognition or response approximation (Harms et al., 2010; Li et al., 2011; Uljarevic &

Hamilton, 2013). Thus, we could suppose that both groups would attend faster and for a longer time to happy faces due to their reported higher accuracy in recognizing positive emotions and the close relationship between visual processing and emotion recognition. Nevertheless, as there are very few studies addressing that issue and those that have applied a different approach, we did not make any prediction at this respect. Finally, we also expected that visual behavior would show the same pattern in both groups, but it would differ quantitatively (H2) as suggested by results of de Wit et al. (2008). Based on previous literature, this would imply that children with ASD would show less orientation to and a reduced scanning of faces compared to controls (Franchini, Glaser, Gentaz et al., 2017; Vacas et al., 2021b; Vivanti et al., 2011; von Hofsten et al., 2009; Zantinge et al., 2017). Regarding the potential effect of the poser's gender, we had no predictions as we had no knowledge of previous studies addressing that issue; meaning that any significant effect of this variable would be a breakthrough.

#### 6.2.1 Method

#### 6.2.1.1 Participants

Forty participants (20 children with ASD and 20 TD) took part in this experiment as they met basic conditions for completing the assessment process (e.g., minimum level of instructions comprehension and following, and ability to sustain attention during the whole eye tracking task). However, the eye tracker apparatus did not capture a minimum of 50% of fixations of three participants with ASD; thus, they were excluded from the study. To balance the sample, we also excluded three participants from the TD group considering the age and gender equivalence in the process. Hence, 34 preschoolers: 17 with ASD (16 boys, 1 girl;  $M_{age} = 56.59$  months,  $SD_{age} = 9.34$ , range = 44-72 months) and 17 TD (16 boys, 1 girl;  $M_{age} = 53.18$  months,  $SD_{age} = 8.10$ , range = 42-71 months) completed the assessment successfully and were included in this experiment. Consistent with a sensitivity analysis carried out in GPower 3.1.9.7 (Faul et al., 2007, 2009), this sample size could yield between-groups differences with 95% of power and an effect size of .30. The ASD group was recruited from centers of early childhood intervention in the province of Córdoba. Inclusion criteria for this group involved: (1) The regular attendance at a center of early childhood intervention, (2) An ASD diagnosis provided by a licensed experienced clinician according to the DSM-5/ICD-10 criteria, following the protocol of Chapter 6. Social attention and autism in early childhood: Evidence on psychophysiological markers based on visual scanning of emotional faces with eye tracking methodology

an Infant Mental Health program at a community mental health service, and (3) The absence of any mental or medical condition. Moreover, we applied a pretest for potential candidates to check whether they were able to follow instructions and to attend to visual stimuli for the necessary amount of time; thus, the selected children were mid-level in terms of functioning. The TD group was recruited from a public school in Córdoba. In this case, inclusion criteria comprised: (1) The absence of any history of developmental disorder either now or in the past, and (2) The gender and chronological age matching with the clinical group. We matched groups on chronological instead of on developmental age as the passive-observational tasks applied in this study did not require high-level cognitive abilities. Groups were also matched on gender to control for a potential gender bias in SA between groups. Results from ANOVA and Chi-square analyses showed that there were no significant differences between groups either in age or gender. However, to further describe our sample, we also assessed receptive vocabulary with the Peabody Picture Vocabulary Test-Third Edition (PPTV-III; Dunn & Dunn, 1997) and emotion recognition and Theory of Mind (ToM) with the social perception domain of the Developmental Neuropsychological Assessment (NEPSY-II; Korkman et al., 2007). Regarding these cognitive abilities, groups differed in terms of basic attention during the eye tracking task, receptive vocabulary, verbal ToM, and total ToM (see Table 6.1 left).

		Experiment	1	Experiment 2				
	ASD	TD			ASD	TD		
	(n = 17)	(n = 17)			(n = 17)	(n = 17)		
	M (SD)	M (SD)	<b>F</b> <sub>(1</sub> , 32)	Р	M (SD)	M (SD)	<b>F</b> <sub>(1</sub> , 32)	Р
Age (months)	56.59 (9.34)	53.18 (8.10)	1.30	.26	54.18 (8.54)	55.53 (8.29)	.22	.64
<b>Basic attention</b>	70 % (10)	80 % (13)	7.44	.01*	72 % (10)	83 % (8)	10.72	.00*
PPVT-III Standard Score	97.94 (12.62)	107.82 (11.43)	5.73	.02*	97.75 (12.99)	108.06 (11.51)	5.84	.02*
	n	n	$\chi^2$	Р	n	n	$\chi^2$	Р
Gender (boy/girl)	16/1	16/1	1	.76	16/1	16/1	1	.76
	ASD	TD			ASD	TD		
	(n = 6)	(n = 17)			(n = 6)	(n = 17)		
	Median (Range)	Median (Range)	U	Р	Median (Range)	Median (Range)	U	Р
NEPSY-II Total Raw Score Affect Recognition	10.50 (10)	13.00 (15)	25.00	.07	10.00 (10)	14.00 (13)	21.50	.02*
NEPSY-II Verbal ToM	2.00 (6)	9.00 (10)	11.00	.01*	2.00 (6)	9.00 (11)	8.50	.00*
NEPSY-II Contextual ToM	2.00 (3)	2.00 (5)	34.00	.21	2.00 (3)	3.00 (5)	31.50	.16
NEPSY-II Total Raw Score ToM	4.50 (9)	11.00 (10)	9.50	.00*	4.50 (9)	11.00 (13)	7.00	.00*

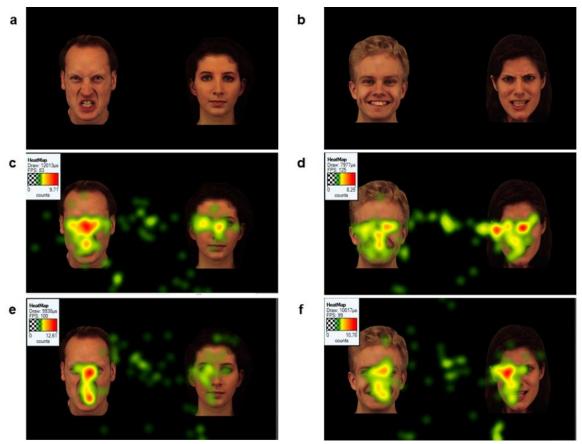
**Table 6.1.** Sample Characteristics in Experiment 1 (left) and Experiment 2 (right)

Age, Basic attention, and PPVT-III Standard Score variables were tested using ANOVA. Gender was analysed using Chi-Square. NEPSY-II scores were analysed using Mann-Whitney U.

#### 6.2.1.2 Stimuli

In this experiment, the designed paired preference task showed paired faces displaying different emotions in female and male posers for assessing visual preference over emotion versus neutrality, positive versus negative emotions, and female or male faces. Thus, we paired pictures of young adults displaying three emotions (happiness, anger, and neutral). We obtained six experimental conditions which were repeated six times including different stimuli (a total of 36 trials). Hence, 36 facial pictures from 12 female posers were paired with 36 facial pictures of 12 male ones (in both cases the same identities were used for representing the three emotions). The posers' gender and the location in the screen were counterbalanced to prevent their potential effect. This resulted in 36 slides with those combinations (see Figures 6.1.a and 6.1.b). Images were taken from Karolinska Directed Emotional Faces database (KDEF; Lundqvist et al. 1998). Slides were resized to  $15 \times 15$  cm approximately and original backgrounds were removed.

**Figure 6.1.** *Stimuli examples of the paired preference task applied in Experiment 1 and heatmaps in ASD and TD groups* 



(a) Example stimulus pairing an angry male (AM10ANS) and a neutral female (AF13NES). (b) Example stimulus pairing a happy male (AM05HAS) and an angry female (AF07ANS). (c and d). Heatmaps of the ASD group performance. (e and f) Heatmaps of the TD group performance.

## 6.2.1.3 Procedure

This study obtained the approval of the Research Ethics Committee of Córdoba. We informed the potential candidate centers of early childhood intervention about the project and professionals provided families with project information, collected signed written informed consents of those interested in participating, and arranged the date for the testing sessions at families and centers convenience. The testing sessions took place in a quiet room at the referential center of each child to ensure a natural environment, with the researchers being the ones who moved with the portable assessment equipment. Children were assessed individually, in some cases parents were present during the testing upon request. All the testing was conducted in one single session of nearly 45 minutes.

Eye movements were tracked and recorded by the system Tobii X2-30 (Tobii Technology AB, Stockholm, Sweden) which has a sampling rate of 30 Hz and a gaze accuracy of  $0.4^{\circ}$ . Children were seated at nearly 60 cm and 36° above a 15'screen of the laptop on which tasks were displayed. They were given no instruction except to look at the screen and observe the images. As we applied a pretest before including children in the experiment, all participants followed instructions appropriately. Before running the task, there was a nine-point calibration showing an animated cat as target stimulus. In this case, children were instructed to look at the cat and follow it with their gaze. Calibration was repeated for those children who failed the first attempt. All children succeeded in the calibration either the first or the second time. After calibration, children were presented with the sequence of 36 slides. It started with an animated fixation point which was displayed for 1 s. to lead children's attention to the center of the screen, then the stimuli appeared in the screen for 5 s.

After the eye tracking task, we applied the PPVT-III (Dunn & Dunn, 1997) and the two subtests included in the social perception domain of the NEPSY-II (Korkman et al., 2007) to all children in order to assess cognitive abilities such as receptive vocabulary, emotion recognition, and ToM to define our groups more comprehensively. These tests were selected for their extensive use by clinicians and researchers, and the fact that both include typification data with special groups (including ASD). However, 11 children from the ASD group could not complete the assessment with the NEPSY-II due to their young age and the excessive language demands of the affective recognition and ToM subscales (see Table 6.1 left). All participants received a gift as reward after completing the testing session.

## 6.2.1.4 Data Analysis

We designed three indexes derived from eye tracking parameters to describe the VSP in both groups as in Sasson and Touchstone (2014): (1) Prioritization, resulting from the time to first fixation parameter to assess attentional orientation, (2) Preference, the proportion of total fixation duration on each stimulus to compare the distribution of visual attention between emotional expressions and poser's gender, and (3) Duration, mean time per visit to faces to account for sustained attention. Some data were transformed to create the aforementioned indexes. Thus, the Preference index resulted from dividing the Total

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Fixation Duration to a kind of stimuli (e.g., "happy male faces") by the Total Fixation Time that a participant looked at the screen during the whole task. Similarly, the Duration index was created by dividing the Total Visit Duration to a kind of stimuli (e.g., "angry female faces") by the number of visits to this kind of stimuli. Finally, the Prioritization index was obtained from the Time to First Fixation parameter of the eye tracker software and did not receive any transformation. It is also important to note that fixations were defined as those looks of at least 100 ms, which implies that gazes under this value were considered missing data. Similarly, we established a cut-off of 50% of fixations collected by the eye tracker in line with previous research (Franchini, Glaser, Gentaz et al., 2017; Franchini, Glaser, Wood de Wilde et al., 2017; Pierce et al., 2011; Pierce et al., 2016; Shi et al., 2015). Under this value, participants or trials were excluded from further analysis. As mentioned in the Participants subsection, three participants of the ASD group were excluded but any trial had to be removed in this experiment.

Statistical tests were conducted in the Statistical Package for the Social Sciences, 25 (SPSS, 25, IBM, Armonk, NY, United States of America). To describe the VSP to different emotional expressions in both groups considering the poser's gender (renamed 'Gender' for data analysis), we performed separated repeated measures ANOVAs on Prioritization, Preference, and Duration indexes, with Group (ASD, control) as the between-group factor and Emotion (happiness, anger, neutral) and Gender (female, male) as the within-group factors. We also calculated effect sizes for repeated measures ANOVAs (partial eta-squared,  $\eta_p^2$ ) considering them small (< .01), medium (< .06), or large (< .14).

#### 6.2.2 Results

Table 6.2 shows means and standard deviations for all experimental conditions in Experiment 1 on the left side (see Table 6.2 left).

#### 6.2.2.1 Prioritization

The ANOVA performed on prioritization data revealed a significant interaction between Group and Emotion ( $F_{(2, 64)} = 4.08$ , MSE = .09, p = .02,  $\eta_p^2 = .11$ ). Planned contrasts revealed a main effect of Group with angry faces ( $F_{(1, 32)} = 5.87$ , MSE = .13, p =.02,  $\eta_p^2 = .16$ ), where children with ASD fixated on angry faces later (1023 ms) compared to control children (813 ms), but not for happy ( $F_{(1,32)} < 1$ ) or neutral faces ( $F_{(1,32)} < 1$ )

(Figure 6.2.a). Likewise, a significant main effect for Gender was found ( $F_{(1, 32)} = 6.81$ , MSE = .05, p = .01,  $\eta_p^2 = .18$ ), where both groups looked at female faces earlier (control group: 853 ms; ASD group: 920 ms) than male ones (control group: 960 ms; ASD group: 978 ms) (Figure 6.2.b).

#### 6.2.2.2 Preference

The ANOVA performed on preference data revealed a marginal interaction between Group and Emotion ( $F_{(2, 64)} = 2.82$ , MSE = .002, p = .07,  $\eta_p^2 = .08$ ). Planned contrasts revealed a main effect of Group with happy faces ( $F_{(1, 32)} = 4.69$ , MSE = .001, p = .04,  $\eta_p^2 = .13$ ), where children with ASD spent a smaller proportion of time looking at happy faces (30 %) compared to controls (33 %); a marginal effect of Group with neutral faces ( $F_{(1, 32)} = 3.31$ , MSE = .001, p = .08,  $\eta_p^2 = .09$ ), where children with ASD spent a larger proportion of time staring at neutral faces (31 %) compared to their typical peers (29 %); and no difference between groups regarding angry faces ( $F_{(1,32)} < 1$ ) (Figure 6.2.c). Furthermore, a main effect of Emotion was also found ( $F_{(2, 64)} = 20.49$ , MSE =.002, p = .000,  $\eta_p^2 = .39$ ), where both groups spent a larger proportion of time looking at angry than happy (control group: 37 % vs. 33 %; ASD group: 38 % vs. 30 %) or neutral faces (control group: 37 % vs. 29 %; ASD group: 38 % vs. 31 %) (see Figures. 6.1.c-f). No effect was found for Gender in this index (see Figure 6.2.d).

#### 6.2.2.3 Duration

The ANOVA performed on duration data revealed a significant main effect of Group ( $F_{(1, 32)} = 8.62$ , MSE = .21, p = .01,  $\eta_p^2 = .22$ ), where children with ASD made significantly shorter visits to faces (697 ms) compared to their peers (888 ms) (see Figures 6.2.e and 6.2.f). Furthermore, a main effect of Emotion was found ( $F_{(2, 64)} = 7.61$ , MSE = .05, p = .001,  $\eta_p^2 = .19$ ), where angry faces received significantly longer visits than happy and neutral ones (angry faces: 876 ms, happy faces: 752 ms, and neutral faces: 750 ms). In fact, control group made significantly longer visits to angry than neutral male faces, whereas the ASD group made significantly longer visits to angry than happy male faces (see Table 6.2 left). No effect was found for Gender in this index either (see Figure 6.2.f).

Елрег	riment 1 (left) and Ex EXPERIME	<u> </u>	EXPERIMENT 2				
DV	Experimental Condition	ASD (n = 17) <i>M</i> ( <i>SD</i> )	TD (n = 17) <i>M</i> ( <i>SD</i> )	Experimental Condition	ASD (n = 17) <i>M</i> ( <i>SD</i> )	TD (n = 17) <i>M</i> ( <i>SD</i> )	
Prioritization (ms)	TFF_Happy Male	934 (320)	982 (426)	TFF_Happy Adult	794 (266)	864 (250)	
	TFF_Happy Female	841 (275)	902 (359)	TFF_Happy Child	966 (232)	815 (325)	
	TFF_Angry Male	1079 (362)	845 (363)	TFF_Angry Adult	966 (320)	927 (447)	
	TFF_Angry Female	967 (298)	781 (322)	TFF_Angry Child	937 (343)	813 (219)	
	TFF_Neutral Male	920 (322)	1052 (340)	TFF_Neutral Adult	968 (268)	962 (357)	
	TFF_Neutral Female	949 (292)	877 (341)	TFF_Neutral Child	1001 (365)	701 (276)	
Preference (%)	PTFD_Happy Male	16 (4)	16 (5)	PTFD_Happy Adult	15 (4)	14 (3)	
	PTFD_Happy Female	14 (4)	17 (3)	PTFD_Happy Child	16 (4)	17 (6)	
	PTFD_Angry Male	21 (5)	18 (4)	PTFD_Angry Adult	18 (3)	17 (4)	
	PTFD_Angry Female	17 (5)	19 (4)	PTFD_Angry Child	17 (4)	19 (4)	
	PTFD_Neutral Male	15 (2)	15 (5)	PTFD_Neutral Adult	16 (4)	15 (5)	
	PTFD_Neutral Female	16 (4)	14 (5)	PTFD_Neutral Child	18 (6)	17 (5)	
Duration (ms)	TPV_Happy Male	636 (256)	893 (318)	TPV_Happy Adult	738 (154)	767 (267)	
	TPV_Happy Female	642 (186)	838 (294)	TPV_Happy Child	741 (226)	953 (353)	
	TPV_Angry Male	817 (306)	999 (303)	TPV_Angry Adult	792 (230)	822 (301)	
	TPV_Angry Female	717 (337)	970 (227)	TPV_Angry Child	769 (254)	809 (232)	
	TPV_Neutral Male	702 (229)	798 (216)	TPV_Neutral Adult	674 (192)	829 (371)	
	TPV_Neutral Female	672 (220)	828 (250)	TPV_Neutral Child	745 (180)	839 (306)	

**Table 6.2.** *Mean and standard deviation (in brackets) for each experimental condition in Experiment 1 (left) and Experiment 2 (right)* 

TFF, Time to first fixation; PTFD, Proportion of total fixation duration; TPV, Time per visit

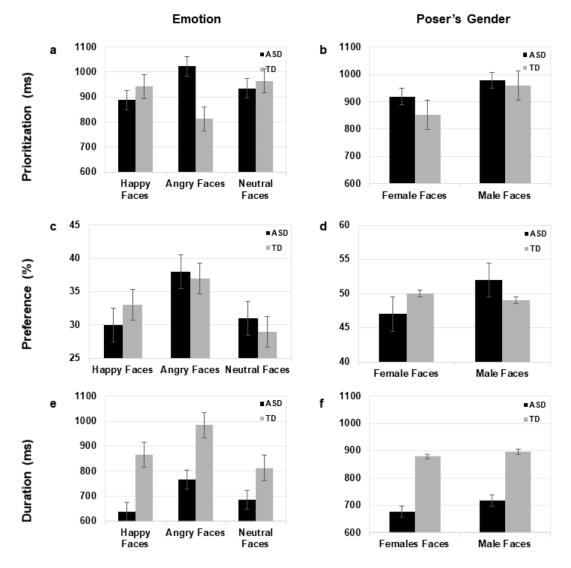


Figure 6.2. Visual attention patterns considering different emotions and poser's gender

(a-b) Prioritization index. (c-d) Preference index. (e-f) Duration index

#### 6.2.3 Discussion

The aim of this experiment was to compare SA in young children with and without ASD depending on the emotional expression (neutral, happy, or angry faces) and the poser's gender (female or male). To this end, we analyzed their VSP of faces assessing the effects of both variables. Therefore, our H1 was partially confirmed as both groups demonstrated emotional sensitivity by scanning faces differently depending on the emotion. When looking at specific emotions, we found that both ASD and TD groups looked longer and more thoroughly at angry faces than at happy or neutral ones, as indicated by the Preference and Duration indexes. Thus, we can state that children with

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ASD not only identified emotionality from neutrality, but they also distinguished between emotions as they performed different visual processing for positive and negative ones. The finding of emotional sensitivity in the population with ASD aligns with previous studies (Nuske et al., 2014, 2016; Vacas et al., 2021b; Vivanti et al., 2011); however, our results differ from research suggesting a bias toward positive emotions in this population (Harms et al., 2010; Li et al., 2011; Uljarevic & Hamilton, 2013). Differences in this sense may be due to the approach applied in prior literature. Some of these studies applied an emotion recognition approach which yielded greater accuracy for recognizing positive than negative emotions in participants with ASD (Harms et al., 2010; Uljarevic & Hamilton, 2013). Therefore, the attentional bias in these studies was considered in terms of better recognition rates and emotional response to positive emotions. Conversely, we did not account for these variables in our sample; instead, we applied an eye tracking paired preference approach to analyze VSP to neutral, positive, and negative facial expressions and categorized the attentional bias in terms of prioritization, visual preference, and visit duration. It could be plausible that participants in the ASD group would better recognize and respond to positive emotions, although angry faces captured their attention for a longer time and in more detail (perhaps because they have more phylogenetic relevance as was found in TD population in the study of Pérez-Dueñas et al., 2014; or because of the development of a compensatory mechanism). Nevertheless, our results are in line with conclusions from a recent systematic review in which a bias toward threatening faces was found in individuals with ASD of all ages (Fan et al., 2020). In this review, authors analyzed studies using different behavioral approaches (excluding the eye tracking one) and concluded that individuals with ASD show an attentional bias toward threatening faces compared to happy but not to neutral ones.

Despite these similarities among conditions, we found between-groups differences in terms of Prioritization, Preference, and Duration indexes:

1) Children with ASD fixated on angry faces significantly later than their typical counterparts, which also implies emotional sensitivity and emotion discrimination to some extent. It could be that these children had rapidly identified angry faces as aversive stimuli and they unconsciously oriented to them later, or it could reveal difficulties in attentional orientation toward negative emotions which, in line with the assumptions of

Pérez-Dueñas et al. (2014), could be related to adaptive difficulties in this population if they detected relevant emotional stimuli (e.g., negative emotions) with delay.

2) Children with ASD spent a smaller proportion of time looking at happy faces (not at neutral or angry ones) compared to controls, and they looked at angry male faces more thoroughly than happy male ones (but not neutral male ones), which also aligns with results of Fan et al. (2020). Likewise, it seems surprising that the control group made significantly longer visits to angry than to neutral male faces, whereas the ASD group made significantly longer visits to angry than happy male faces. This, alongside our finding that children with ASD attended to happy faces less (and to neutral ones more) than their TD peers, could entail a specific difficulty in processing happy faces in the population with ASD.

3) Children with ASD made shorter visits to all faces (regardless of the emotional expression or the poser's gender) compared to their TD peers, which implies that they made an atypical and more superficial scanning of these faces. This finding confirms our H2 and aligns with previous literature (Franchini, Glaser, Gentaz et al., 2017; Jones et al., 2008; Vacas et al., 2021b; Vivanti et al., 2011; von Hofsten et al., 2009; Zantinge et al., 2017). Visit duration is an index developed to assess sustained attention, which is intimately related to functions such as visual working memory (Chun, 2011) and covert orienting (Xie & Richards, 2017). It can be considered an indicator of how accurately we process stimuli due to its impact on our perception of the world (Colombo, 2001; Pérez-Edgar et al., 2010; Richards, 2010; Xie & Richards, 2017). Hence, a more superficial facial processing in children with ASD could be impairing their comprehension of the world and their social relationships according to Pérez-Edgar et al. (2010). This theory is in line with research suggesting that children with ASD have less social input and experience since their early infancy, which affects their social expertise and hinders their neural specialization; this, consequently, compromises their social functioning later on (Jones et al., 2008; Klin, 2008; Sasson, 2006; Sasson et al., 2008; Sasson et al., 2011; Sasson & Touchstone, 2014; Shultz, 2005). On the other hand, if a sound body of evidence suggests that children with ASD attend to faces less than their TD peers but they show an attentional bias toward threatening faces, it could be that this reduced SA was contingent upon the nature, valence, and salience of facial emotions. The attentional bias away from angry faces has been related to social communication impairments (GarcíaThe highlighted prioritization and duration atypicalities in children with ASD were also found in a recent study of this research group pairing social and non-social stimuli (Vacas et al., 2021b), which reinforces the idea of anger-orienting difficulties and reduced VSP to faces as differential psychophysiological markers of the ASD condition that could help detect ASD early in development.

Finally, we had made no prediction about VSP to faces regarding gender as no previous literature had addressed this issue to our knowledge. We did not find betweengroups differences, but a faster orientation to female faces was highlighted in both groups by the Prioritization index. This finding may be also explained by a phylogenetic reasoning: if children were more acquainted with female caretakers, they may have developed this fast orientation to them.

# 6.3 Experiment 2. Poser Age

In this case, we paired happy, angry, and neutral child faces with the same emotions shown in adult faces to determine differences between conditions in VSP depending on the poser's age. Based on the clinical definition of the ASD, we predicted that these children would display less attention to child faces regardless of the emotion compared to the TD group (H3), as their social difficulties with peers have been widely reported (APA, 2013; WOS, 1992). However, given the results of Hauschild et al. (2020), the null hypothesis could also be possible. Finally, consistent with Experiment 1 and previous literature (Franchini, Glaser, Gentaz et al., 2017; Vacas et al., 2021b; Vivanti et al., 2011; von Hofsten et al., 2009; Zantinge et al., 2017), we also expected to find between-groups differences in terms of the kind of scanning for faces, being more reduced in the ASD group (H4).

## 6.3.1 Method

# 6.3.1.1 Participants

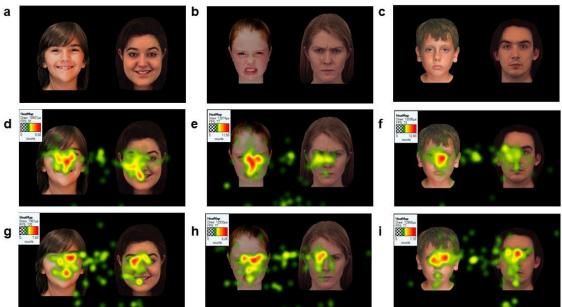
We included 40 participants (20 children with ASD and 20 TD) in this experiment as potential candidates for completing the assessment according to the same criteria as in Experiment 1 (e.g., minimum level of instructions comprehension and following, and ability to sustain attention during the eye tracking task). Eye tracking data of three participants with ASD did not reach the threshold to be included in the analysis (at least 50% of fixations in the task); thus, they were excluded from further analyses. We again balanced the sample excluding three participants from the TD group according to an age and gender equivalence criterium. Therefore, 34 preschoolers: 17 with ASD (16 boys, 1 girl;  $M_{age} = 54.18$  months,  $SD_{age} = 8.54$ , range = 44 - 72 months) and 17 TD (16 boys, 1 girl;  $M_{age} = 55.53$  months,  $SD_{age} = 8.29$ , range = 44 - 72 months) were included in this experiment. Results from ANOVA and Chi-square analyses yielded no significant differences between groups either in age or gender, but again groups differed in basic attention during the eye tracking task, receptive vocabulary, emotion recognition, verbal ToM, and total ToM (see Table 6.1 right). The sensitivity analysis and recruitment procedure were identical to those of Experiment 1.

## 6.3.1.2 Stimuli

In this experiment, the designed paired preference task displayed the same emotion in child and adult faces to determine visual preference regarding the poser's age. Thus, we paired pictures of children and young adults displaying three emotions (happiness, anger, and neutral). We again obtained 36 trials (six experimental conditions per six repetitions each). Hence, 36 facial pictures from 13 child posers were paired with 36 facial pictures of 21 adult ones (as in Experiment 1, we tried to ensure that the same identities depicted the three emotions, but in this case, it was not possible due to difficulties in finding databases with a substantial number of child posers and pairing these posers with adult ones, meaning that the number of identities varied across ages). Given these difficulties, we had to take child and adult pictures from different databases. Therefore, child pictures were taken from the Developmental Emotional Faces Stimulus Set (DEFSS; Meuwissen et al., 2016), while adult pictures were taken again from the KDEF (Lundqvist et al., 1998) as in the Experiment 1. Age of children posers in the

Chapter 6. Social attention and autism in early childhood: Evidence on psychophysiological markers based on visual scanning of emotional faces with eye tracking methodology DEFSS (Meuwissen et al., 2016) ranged from eight to 13 years, whilst adults posers in the KDEF (Lundqvist et al., 1998) were aged between 20 and 30 years. The fact that images came from different databases forced us to modify pictures shine to make them more similar to each other, which increased differences between stimuli (see Figures 6.3.a – 6.3.c).

**Figure 6.3.** *Stimuli examples of the paired preference task applied in Experiment 2 and heatmaps in ASD and TD groups* 



(a) Example stimulus pairing happy child (2\_F8\_Happy) and adult (AF03HAS) faces. (b) Example stimulus pairing angry child (16\_F9Angry) and adult (AF22ANS) faces. (c) Example stimulus pairing neutral child (4\_M8\_Neutral) and adult (AM35NES) faces. (d-f) Heatmaps of the ASD group performance. (g-i) Heatmaps of the TD group performance

## 6.3.1.3 Procedure and Data Analysis

Both procedure and data analysis were almost identical in Experiments 1 and 2 (with the same locations used, same instructions given to participants, same cognitive assessment applied, same data treatment and processing developed, etc.). Again, three participants were excluded from the original sample, as they did not meet the minimum criterium of 50% of fixations to be selected for further analysis, but any trial had to be omitted.

In this case to describe the VSP to different emotional expressions in both groups considering the poser's age (renamed 'Age' for data analysis). We conducted separated

repeated measures ANOVAs on Prioritization, Preference, and Duration indexes, with Group (ASD, control) as the between-group factor and Emotion (happiness, anger, neutral) and Age (child, adult) as the within-group factors.

#### 6.3.2 Results

Table 6.2 shows means and standard deviations for all experimental conditions in Experiment 2 on the right side (see Table 6.2 right).

#### 6.3.2.1 Prioritization

The ANOVA performed on prioritization data revealed a significant interaction between Group and Age ( $F_{(1, 32)} = 8.53$ , MSE = .06, p = .01,  $\eta_p^2 = .21$ ). Planned contrasts revealed a main effect of Group with child faces ( $F_{(1, 32)} = 9.25$ , MSE = .01, p < .01), where the ASD group fixated on child faces later (967 ms) than their control peers (776 ms), but there was no difference between groups for adult faces ( $F_{(1, 32)} < 1$ ) (Figure 6.4.a).

#### 6.3.2.2 Preference

The ANOVA performed on preference data revealed a significant main effect for Age ( $F_{(1, 32)} = 5.20$ , MSE = .002, p = .03,  $\eta_p^2 = .14$ ), where both groups spent a smaller proportion of time looking at adult faces (control group: 46 %; ASD group: 49 %) compared to child ones (control group: 53 %; ASD group: 51 %) (Figures 6.3.d – 6.3.i and 6.4.b).

#### 6.3.2.3 Duration

The ANOVA performed on duration data revealed a significant interaction between Group, Emotion, and Age ( $F_{(2, 64)} = 3.44$ , MSE = .02, p = .04,  $\eta_p^2 = 10$ ). To examine this interaction further, separated ANOVAs were conducted for each Emotion, with Group (ASD, control) as the between-group factor and Age (adult, child) as the within-group factor. The ANOVA for happy faces revealed a main effect of Age ( $F_{(1, 32)}$ = 5.23, MSE = .03, p = .03,  $\eta_p^2 = .14$ ), where happy child faces received longer visits (847 ms) than happy adult faces (753 ms); and a significant interaction Group x Age ( $F_{(1, 32)} =$ 4.97, MSE = .03, p < .05), where the Age effect was significant for the control group ( $F_{(1, 32)} <$ 32) = 10.19, MSE = .03, p < .01) but not significant for the ASD group ( $F_{(1, 32)} <$ 1). This

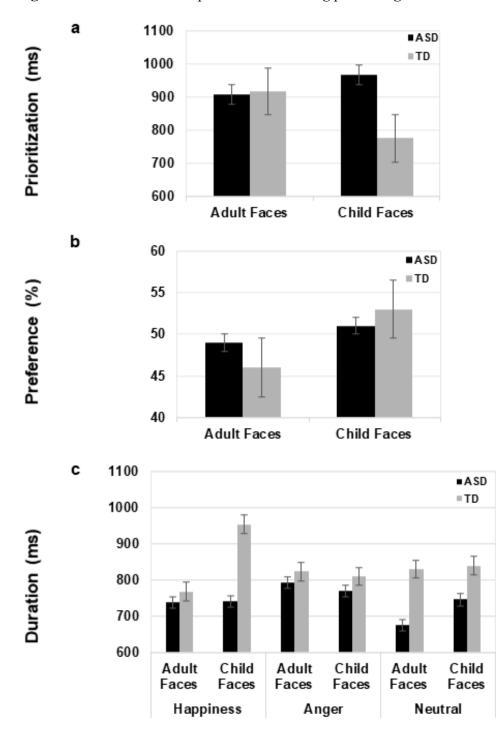


Figure 6.4. Visual attention patterns considering poser's age

<sup>(</sup>a) Prioritization index. (b) Preference index. (c) Duration index. Triple interaction

# 6.3.3 Discussion

In this experiment, we aimed at finding potential differences between ASD and TD groups in SA depending on the poser's age. To this purpose, we analyzed participants' VSP of faces assessing the effects of that variable. We expected that children with ASD would display less attention to child faces compared to the TD group due to their peer difficulties (H3); however, we found that the ASD group showed the same visual preference for child faces as their counterparts, thus refuting our hypothesis. This behavior implied reduced attention to adult faces, which was consistent in both groups. Despite the fact that this result may conflict with clinical characterization of the ASD condition, it aligns with the age sensitivity and own-age bias reported in adolescents with ASD in a recent study (Hauschild et al., 2020). It could be that this population showed typical visual preference and cognitive strategies in dealing with their peers, but they failed in pragmatics and the development of consistent behaviors, which would inevitably impact on their social interactions as manuals report. On the other hand, the Prioritization index yielded between-groups differences in attentional orientation to child faces in the ASD group. More research is needed to disentangle the nature of this behavior and its connection to peer difficulties in individuals with ASD.

Finally, data in this experiment partially confirmed our H4, which expected to replicate the reduced scanning of faces found in the ASD group. In this case, visit duration to both child and adult happy faces differed in the ASD and TD groups; thus, TD children processed child happy faces in more detail than adult ones, while children with ASD did not make this differentiation. This scattered difference provides further insight into how children with ASD process social-emotional information according to specific stimuli characteristics. It may indicate that these children do not show a generalized difficulty for processing all emotions in all people, but some difficulties may be associated with particular conditions. On the other hand, we presume that the nature of the stimuli (the fact that they were taken from different databases) or the effect of intervention (it is noteworthy that our ASD sample had been receiving early intervention for approximately 30 months -SD = 8 months-, which could be influencing their facial processing) could be responsible for the finding of scattered differences between groups in the Duration index. Therefore, future studies should account for these variables to evaluate their effect.

#### 6.4 General Discussion

The main aim of the present paper was to compare SA in young children with and without ASD regarding the role of emotional expressions, the poser's gender, and the poser's age. To this end, we designed two eye tracking paired preference tasks assessing VSP to different faces in terms of attentional orientation, visual preference, and sustained attention in the ASD and control groups. Despite some typical visual behaviors found in the ASD sample (e.g., emotional sensitivity and discrimination or visual preference for child faces), we consider remarkable the detection of three specific psychophysiological markers in the ASD group which could contribute to early diagnosis: (1) Late orientation to angry faces, (2) Late orientation to child faces, and (3) Superficial processing of faces (shorter visits). In this sense, we may say that Prioritization and Duration indexes have proved to be efficient in distinguishing groups, replicating results from previous studies (Vacas et al., 2021b); thus, these psychophysiological markers become relevant from a clinical point of view as they may be applied as diagnostic criteria at early ages. These distinctive markers are also worth considering in regard to intervention as they may impact on children's adaptive behavior, not only from a biological but also from a social perspective, given that both are considered essential tools for effective social and environmental adaptation. Thus, increasing the acquaintance with negative facial expressions and peer faces could potentially improve adaptive social behavior in children with ASD. This hypothesis should be tested in future studies. The nature of difficulties in processing happy faces in children with ASD are also worth investigating in more detail in future studies. Furthermore, we observed that some effects of emotion found in Experiment 1 disappeared in Experiment 2 (mainly those regarding anger). In this sense, we consider that Experiment 2 highlighted particular stimuli characteristics (e.g., poser' s age), which may contribute toward narrowing the nature of social-emotional processing in children with ASD. Hence, replicating Experiment 1 including only child faces could help to disentangle the real influence of poser's age on emotion visual processing. Likewise, we consider that the control and the longitudinal study on the effects of intervention on VSP in children with ASD and the comparative research on differential diagnosis assessing these psychophysiological markers could improve knowledge on early intervention in ASD and consequently, significantly enhance clinical practice.

Finally, it is relevant to highlight that the use of different databases for child and adult faces in Experiment 2 could have biased participants attention due to differences in

image settings. This limitation calls for a future replication involving different stimuli and alternative assessments to account for potential correlations. Furthermore, although our sample size was similar to that of some related studies (Guillon et al., 2016; Jones et al., 2008; Liberati et al., 2017; Sasson & Touchstone, 2014; Vacas et al., 2021b; Vivanti et al., 2011; von Hofsten et al., 2009), larger samples are also welcomed to confirm patterns or disentangle subtle atypicalities.

## Acknowledgements.

We gratefully acknowledge to all families and children who participated in this study. We extend our sincere acknowledgement to those Early Intervention Centers of the province of Córdoba and the public school Al Andalus (Córdoba, Spain), which kindly accepted our request of participation and facilitated the labor of the researchers.

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# Chapter 7. Visual Behavior as Differential Marker for Early Diagnosis in Young Children with Autism Spectrum Disorders and Developmental Language Disorder. An Eye Tracking Comparative Study<sup>4</sup>

#### Abstract

Children with Autism Spectrum Disorders (ASD) and Developmental Language Disorder (DLD) may present similar cognitive and linguistic profiles at early ages, which has been highlighted as a major obstacle for early diagnosis. Given the importance of early diagnosis for children prognosis, this comparative study attempted to find differences in terms of the visual scanning pattern (VSP) to faces and objects between a group of young children with ASD, another one with DLD, and a control group (N = 33; age range 32-72 months). We designed a paired preference task matching happy, angry, and neutral faces with objects related to autism circumscribed interests (CIOs) or not related to them (non-CIOs). We used eye tracking methodology to analyse the VSP of participants. This VSP was operationalized as prioritization, visual preference, and visit duration to social and non-social stimuli. Results showed three main insights: (1) A superficial face processing in both clinical groups compared to controls (revealed by the Duration index), (2) An attentional bias toward angry faces in all groups (highlighted by the Preference index), and (3) A sound effect of CIOs on all groups (delaying prioritization, reducing preference, and diminishing visit duration to faces). Findings from this comparative study encourage the inclusion of children with DLD in the assessment of social attention through comparative studies as a way to find specific psychophysiological markers that may contribute to differential diagnosis of these conditions.

**Keywords:** Autism Spectrum Disorders, Developmental Language Disorder, Psychophysiological Markers, Eye-tracking, Early intervention, Differential diagnosis.

<sup>&</sup>lt;sup>4</sup> This chapter follows the structure of a scientific article as it is in process of being adapted for submission to a high impact scientific journal.

# 7.1 Introduction

Autism Spectrum Disorders (ASD) comprises a set of neurodevelopmental and pervasive conditions in which communication deficits are among their core symptoms (American Psychiatric Association [APA], 2013; Kjelgaard & Tager-Flusberg, 2001;World Health Organization [WOS], 1992). On the other hand, Developmental Language Disorder (DLD; also known as specific language impairment or language disorder) is a developmental condition characterized by severe and persistent deficits in the acquisition or the use of language with great impact on individuals' daily functioning (APA, 2013; Bishop et al., 2016, 2017; Leonard, 1998; WOS, 1992).

Traditionally, ASD and DLD have been considered as different conditions given that language difficulties were apparently well-differentiated in each disorder. While individuals with ASD usually show deficits in pragmatics, the impairment in structural language is a hallmark of population with DLD. However, the wide heterogeneity of both profiles, not only in terms of language impairments but also in observed autistic traits and social behavior (Bishop, 2000; Bishop & Norbury, 2002; Conti-Ramsden et al., 2006; Leyfer et al., 2008; Taylor & Whitehouse, 2016), has raised the issue of the unclear boundaries between these disorders (Bishop, 2003, 2010; Georgiou & Spanoudis, 2021; Taylor et al., 2015; Taylor & Whitehouse, 2016). On this issue, some researchers argue that similarities between disorders are superficial, and they cannot account for an overlap among conditions (Schaeffer, 2018; Taylor et al., 2014; Taylor et al., 2012; Williams et al., 2008). On the other hand, other researchers posit that a significant percentage of individuals are far from being 'textbook cases' as they show a mixed clinical picture. This implies that both conditions may belong to different points of the same continuum of impairment at different levels of severity, instead of being two distinct categories (Bishop, 2000, 2003, 2010; Bishop & Norbury, 2002; Conti-Ramsden et al., 2006; Georgiou & Spanoudis, 2021; Leyfer et al., 2008; Taylor & Whitehouse, 2016; Tomblin, 2011). These mixed cases are the most challenging in terms of assessment, diagnosis, and intervention (Bishop, 2000). This fact along with the different trajectories that both disorders may follow (Bishop & Norbury, 2002; Charman, 2010; Conti-Ramsden et al., 2006; Williams et al., 2008) have led the research community to claim for more comparative studies, thus highlighting the relevance of differential diagnosis (Bishop et al., 2016; Georgiou & Spanoudis, 2021; Rice, 2016; Vacas et al., 2021b; Weismer, 2013).

#### Developmental Language Disorder. Studies with Eye Tracking

Given the difficulties in distinguishing these disorders according to their language profiles and the marked social impairment associated to both conditions, the study of socio-cognitive domains may shed light into the issue of differential diagnosis. At this respect, many researchers have shown interest in the analysis of social attention (SA, the distribution of visual attention to social stimuli or socially relevant elements rather than non-social ones), at least in population with ASD. Moreover, the emergence of new approaches and methodologies (e.g., eye tracking) in the last decades has contributed to the developmental understanding of that sociocognitive ability. The eye tracking methodology traces and monitors individuals' eye movements during the observation of visual content, promoting the study of their looking behavior. It leans on corneal reflection technology to unveil reliable attentional markers in a non-invasive way (Aslin & McMurray, 2004; Gredebäck et al., 2009; Hepach et al., 2015). This pros along with its high sensitivity to uncover slight attentional atypicalities in visual emotion processing and unravel the core mechanisms of facial emotional decoding have laid the ground for its use in research on ASD (Bolte et al., 2016; Falck-Ytter, 2013; Harms et al., 2010; Klin et al., 2002a).

Many eye tracking studies on SA in children with ASD have observed diminished attention to social stimuli and a clear attentional bias to non-social elements in this population (Chevallier et al., 2015; Chita-Tegmark, 2016a, 2016b; Falck-Ytter et al., 2013; Guillon et al., 2014; Guillon et al., 2016; Nakano et al., 2010; Vacas et al., 2021c; Zantinge et al., 2017). Some studies have found this unusual attentional pattern to social events alongside difficulties in attention disengagement in infants with ASD from their first year (Falck-Ytter et al., 2013; Sasson et al., 2008). Other study reported lack of arousal modulation towards others' emotions as a concurrent symptom accompanying the non-social attentional pattern in their sample of children with ASD (Zantinge et al., 2017). In this line, the visual scanning pattern (VSP, also known as gaze or looking pattern, which is the sequencing of gaze shifts during the processing of any visual content) has also taken a place in the research on children with ASD. The assessment of VSP in ASD samples has revealed a highly dispersed pattern of observation of social content in this

population, which means that these children showed scattered eye movements across the images when viewing social situations (Liberati et al., 2017).

On this matter, faces are the main social entryway that provides key information to perceive, recognize, and comprehend others' inner emotional states (Frith & Frith, 2006; Klin, 2008). Hence, these have been widely studied inside the analysis of SA in different populations. Some studies have concluded that attention to faces is a natural mechanism which emerges early in development in typical individuals, allowing them to achieve brain specialization and increasing their social expertise (Klin, 2008; Johnson, 2000; Nelson, 2001; Shultz, 2005). Instead, children with ASD have demonstrated diminished attention to faces (Franchini et al., 2017; Vacas et al., 2021c; Vivanti et al., 2011; von Hofsten et al., 2009; Zantinge et al., 2017). Nevertheless, this population has also demonstrated that their visual behavior is susceptible to emotional faces as these captured their attention and provoke pupillary reactions to a higher extent than neutral ones (Nuske et al., 2014, 2016; Vacas et al., 2021c; Vivanti et al., 2011). It is noteworthy that their reactions still differed from those of their typically developing (TD) peers, but still this finding implies that children with ASD, similar to their counterparts, show emotional sensitivity. Apart from emotionality, another potential trigger for pupillary reactivity is familiarity, as it has been proved that it fostered the peak magnitude of pupils to fear faces in children with ASD (Nuske et al., 2014). Concerning the attention to critical facial features (eyes, nose, and mouth), results regarding the eyes and the mouth regions are inconsistent if we consider individual studies; nevertheless, recent reviews have concluded that children with ASD process faces atypically (Black et al., 2017; Harms et al., 2010; Sasson, 2006). Jones et al. (2008) even found this atypical processing style in two years old toddlers with ASD. This feature of the autism spectrum has led several academics to postulate that these children may receive less social learning input from their early infancy, which may be, at least, partially responsible for their future social development as it would be impairing their brain specialization (Jones et al., 2008; Klin, 2008; Sasson, 2006; Sasson et al., 2008; Sasson et al., 2011; Sasson & Touchstone, 2014; Shultz, 2005).

On the other hand, as we highlighted in a previous work (Vacas et al., 2021a) no study has addressed SA specifically in children with DLD with eye tracking. Instead, most eye tracking studies with this population have focused on their narrative abilities

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(either expressive or receptive) after observing prompted images (Andreu et al., 2011, 2013; Palmović & Jušić, 2011; Pons et al., 2013; Serra et al., 2017) or their capacity for word recognition and prediction (Andreu et al., 2012; van Alphen et al., 2021). However, two studies have tackled SA from a cross-syndrome perspective (Hanley et al., 2014; Hosozawa et al., 2012). In this case, both studies found similar VSP in children with DLD and their TD peers using dynamic and ecological stimuli. This VSP was described as social oriented and focused. Conversely, gaze behavior in both ASD groups was atypical in terms of non-social orientation and reduced attention to people's faces, more specifically to the eye region (Hanley et al., 2014; Hosozawa et al., 2012). Likewise, accurate attention shifts were observed in DLD and TD samples when discourse was introduced, that is, both groups looked alternatively at the appropriate speaker, while children with ASD failed to follow the speakers with their gaze (Hosozawa et al., 2012). Despite these similarities between studies, findings of both investigations diverge in their reports on the attentional bias to the mouth region in children with DLD. Hence, Hosozawa et al. (2012) found evidence of that behavior in their DLD sample, while the effect was marginal in the study of Hanley et al. (2014) suggesting a slight trend (close to significancy) to a language compensatory mechanism in children with DLD.

## 7.1.2 The Role of Competing Objects in Social Attention

The influence of the saliency of competing non-social objects on the visual behavior of children with ASD is also worthy to consider when assessing SA in this population (Sasson et al., 2008; Sasson et al., 2011; Sasson & Touchstone, 2014). Some researchers have found a restricted and repetitive attentional bias to non-social stimuli in children with ASD. This behavior may be related to the same cognitive mechanisms involved in the repetitive behaviors associated to this disorder, mainly the circumscribed interests (CI, Pierce & Courchesne, 2001; Sasson et al., 2011; Sasson & Touchstone, 2014). Notwithstanding the relevance of this point, only a few papers have addressed the influence of non-social objects competing with social stimuli in SA in children with ASD. In this sense, Sasson et al. (2008) found a restricted looking behavior to CI-related objects (CIOs) in school-aged children with ASD. This atypical behavior was not found in the same sample regarding non-CI-related objects (non-CIOs) or social elements. Likewise, this pattern of visual behavior was replicated with younger children with ASD in Sasson

et al. (2011). In both cases, the VSP of these children was described as circumscribed, perseverative, and detail oriented.

In the same line, Sasson and Touchstone (2014) examined SA in young children with ASD using a paired preference task which matched faces displaying several emotions (happiness, sadness, fear, anger, and neutrality) with CIOs and non-CIOs. This authors found that children with ASD exhibited typical visual preference for faces when these were paired with non-CIOs; therefore, only CIOs could overcome preference for faces in the ASD group. Following the same paradigm, a previous study of this research team partially replicated this finding (Vacas et al., 2021c). We found similar VSP between the ASD and control groups in terms of prioritization, preference, and longer visit duration to faces paired with non-CIOs rather than to those paired with CIOs, but the clinical group showed in general significantly reduced attention to faces in all conditions. Another relevant finding in our previous research was the identification of a bias away from angry faces exclusively in children with ASD. This is an indicator of emotional sensitivity, but it could also be related to adaptive difficulties in this population as negative emotions serve a purpose of survivance (Pérez-Dueñas et al., 2014).

Given that the presence of CI is a hallmark of ASD, and that this restricted looking behavior seems to be also specific of the autistic phenotype, the effect of the saliency of competing non-social objects on SA may yield differences between children with ASD and those with DLD.

## 7.1.3 Aims and Hypotheses

Considering the aforementioned information, previous literature has proved that eye tracking studies on SA in both populations may yield differential psychophysiological markers that increase knowledge of both disorders. Likewise, the particular role of CIOs in attention to faces in children with ASD may also contribute to differential diagnosis even in those mixed cases in which the autistic symptoms cooccur with language impairments. Finally, this kind of studies could also unveil key commonalities and differences between disorders which may contribute to the definition of the target disorders and, consequently, shed some light into the current debate on their potential shared etiology. Chapter 7. Visual Behavior as Differential Marker for Early Diagnosis in Young Children with Autism Spectrum Disorders and Developmental Language Disorder. An Eye Tracking Comparative Study

Following this reasoning, the main aim of this work was to develop a comparative study to assess young children with ASD, DLD, and controls in terms of attention to faces and objects (a key component which allows the study of a more complex ability -SA-) considering the role of emotional expressions and competing objects in their VSP. To do so, we applied an eye tracking paired preference task combining social stimuli (faces showing happiness, anger, or neutral expressions) with non-social ones (CIOs or non-CIOs) to compete for participants' attention. The eye tracking methodology allowed us to collect and analyze participants' eye movements during the task performance; while the application of a paired preference paradigm (consisting of the presentation of paired stimuli) enabled us to compare the saliency of each kind of stimuli as well as the interference effect that stimuli may exert on each other.

On the basis of previous literature, we expected that young children from the ASD group would display significantly less attention to faces than their counterparts of the control group, while we did not have sufficient grounds to predict the visual behavior of children from the DLD group at this respect (H1). Moreover, we varied the emotional expression in facial stimuli to assess the effect of emotionality in SA in all groups. Hence, we hypothesized that emotional faces (happy and angry ones) would lead to a particular visual processing different from that of neutral ones in all groups (H2). This hypothesis is in line with studies reporting emotional sensitivity in children with ASD (Nuske et al., 2014, 2016; Vacas et al., 2021c; Vivanti et al., 2011). We had no knowledge of differences between conditions in attention to faces depending on the emotion, so any finding at this respect would be novel. Finally, in line with results from Sasson & Touchstone (2014), we theorized that the type of object would have a major impact on children in the ASD group, with CIOs significantly reducing their attention to faces, while non-CIOs would lead to typical attention to faces (H3). As CIs are a hallmark in ASD, we expected that children from the DLD and TD groups were less distracted by CIOs than those of the ASD group, which would yield differences between conditions.

## 7.2 Method

## 7.2.1 Participants

Thirty-three children were assessed with the eye tracking paired preference task: 11 with ASD (10 boys, 1 girl;  $M_{age} = 56$  months,  $SD_{age} = 8.78$ , range = 44-71 months), 11

with DLD (9 boys, 2 girls;  $M_{age} = 56.45$ ,  $SD_{age} = 13.67$ , range = 32-72 months), and 11 TD (10 boys, 1 girl;  $M_{age} = 54.55$ ,  $SD_{age} = 10.49$ , range = 39-72 months). It is particularly noteworthy that we established a threshold of 50% of fixations collected by the eye tracker to consider the inclusion of a participant or a trial, in line with previous research (Franchini, Glaser, Gentaz et al., 2017; Franchini, Glaser, Wood de Wilde et al., 2017; Pierce et al., 2011; Pierce et al., 2016; Vacas et al., 2021c). According to this criterion, any participant was excluded from further analysis.

The clinical groups were recruited from centers of early intervention in the city of Cordoba (Spain). For children with ASD, inclusion criteria involved a positive M-CHAT test. For both clinical conditions, other inclusion criteria comprised: (1) A thorough assessment and the subsequent provision of a diagnosis of ASD or DLD conducted by a licensed experienced team of clinicians belonging to the mental health public system; (2) The regular attendance to a center of early intervention; (3) Some level of attention maintenance, comprehension, and following instructions; and (4) The absence of any mental or medical condition. Children of the TD group were recruited from the preschool classes at a public mainstream school in Cordoba (Spain). The only inclusion criterion for this group was not having history of developmental disorder either now or in the past. This group was matched on gender and chronological age with the clinical ones. As in previous studies, we matched groups on chronological age instead of on mental age due to the low demand of the task applied, as passive-observational tasks do not require highlevel cognitive abilities (Sasson and Touchstone, 2014; Vacas et al., 2021c). We also matched groups on gender to control its potential effect on CI (Cho et al., 2017; DeLoache et al., 2007; Sasson et al., 2012; Sasson & Touchstone, 2014).

Despite matching all the three groups on gender and chronological age, children in the clinical groups differed from controls in terms of receptive vocabulary (assessed with the Peabody Picture Vocabulary Test Third Edition -PPVT-III-; Dunn & Dunn, 1997) and basic attention (understood as the percentage of overall attention that participants deployed to the screen during the visualization of the task as collected by the eye tracker) (see Table 7.1). Post hoc analyses indicated that there were no significant differences between clinical groups in basic attention, but the DLD group significantly differed from the control one (p = .007) and the ASD group marginally differed from their TD peers (p = .057). Regarding PPVT-III Raw Score, differences were marginal between Chapter 7. Visual Behavior as Differential Marker for Early Diagnosis in Young Children with Autism Spectrum Disorders and Developmental Language Disorder. An Eye Tracking Comparative Study the DLD and control groups (p = .062). Finally, with respect to PPVT-III Standard Score significant differences were found between the DLD group and the control one (p = .000) as well as between the ASD and control groups (p = .014), but not between clinical groups.

	ASD	DLD	TD		
	(n = 11)	(n = 11)	(n = 11)		
	M (SD)	M (SD)	M (SD)	<b>F</b> <sub>(2, 32)</sub>	Р
Age	56.00	56.45	54.55	.088	.916
	(8.78)	(13.67)	(10.49)		
Basic attention	76.36%	72.91%	86.73%	5.91	.007
	(8.63)	(10.54)	(10.16)		
PPVT-III Raw Score	42.10 (16.53)	38.10	55.73	3.35	.05
		(16.76)	(16.12)		
PPVT-III Standard Score	94.90 (13.45)	86.80	110.91	11.22	.000
		(15.18)	(5.19)		
	n	n	n	$\chi^2$	Р
Gender (boy/girl)	10/1	9/2	10/1	.569	.752

 Table 7.1. Sample Characteristics

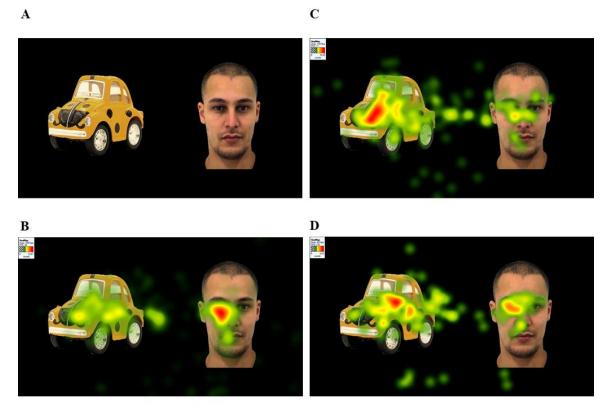
Age, Basic attention, and PPVT-III Raw Score and Standard Score variables were tested using ANOVA. Gender was analysed using Chi-Square.

#### 7.2.2 Stimuli

As mentioned before, in this study we applied a commonly used paired preference task that paired social and non-social images to evaluate participants' VSP toward faces and objects and to weight the impact of the competing stimuli over attention to faces in each group (Sasson and Touchstone, 2014; Vacas et al., 2021c). Thus, the task included faces showing three different emotions (happiness, anger, and neutral) and two types of objects (CIO and non-CIO). After pairing emotional faces and objects, we obtained six experimental conditions, which were repeated six times varying facial identities. This process resulted in a task composed of 36 trials. The posers' gender was counterbalanced to ensure the same representation of male and female faces; likewise, the laterality of all stimuli was also counterbalanced to control for the potential effect of the stimuli location in the screen. Therefore, 36 pictures from 12 different posers (the same posers represented the three emotions, but any of them was repeated for the same emotion) were taken from the Amsterdam Dynamic Facial Expression Set (ADFES; Van der Schalk et al., 2011). These facial pictures were matched with 36 illustrations of objects (18 CIOs and 18 non-

CIOs, see Figure 7.1.A) taken from the free copyright database Pixabay under the Creative Commons CC0 license, or of our own creation by making photographs of common toys and puzzles used in the early intervention center at the University of Cordoba. Objects were included in the CIO category on the basis of previous literature on CI-related topics and objects in ASD (Sasson et al., 2008, 2011; South et al., 2005). Thus, the CIO category comprised toys, puzzles, means of transport, animals, and blocks, while the non-CIO category included plants, furniture, musical instruments, tools, school material, and clothes.

## Figure 7.1 . Examples of stimuli and heatmaps in ASD, DLD, and TD groups



(A) Example stimulus pairing a neutral face (M07-Neutral) and a CIO. (B) Heatmaps of the TD group performance. (C) Heatmaps of the ASD group performance. (D) Heatmaps of the DLD group performance.

We used PowerPoint to design the pairs. Thus, we readjusted the pictures size (to  $15.3 \times 11.3$  cm approximately), controlled their location in the screen, and removed the background. The background removal required a pictures readjustment to ensure that all stimuli looked similar in size; therefore, all images do not have the same size. Resulting compositions were rescaled to  $1500 \times 843$  pixels to fit with the resolution of the laptop used to run the task and the presentation order was counterbalanced.

## 7.2.3 Procedure

This study counted on the approval of the Research Ethics Committee of Cordoba. We tracked the potential candidate centers of early attention, then we contacted them and gave the project information to distribute it among families. Professionals interested in the project informed families of potential candidates about the procedure and collected the signed written informed consents of those parents or tutors receptive to their child participation. The appointments for evaluation sessions were scheduled at families' and centers' convenience, as they were developed in a quiet room free of distractors at the referral centers to facilitate participation. Most children took the assessment alone with the researcher; only were some parents present in the room in isolation, if the child or themselves required it. All evaluations were undertaken in one single session of nearly 45 minutes. Firstly, children visualized the paired preference task, which lasted 3.5 minutes approximately. Eye tracking data were collected using Tobii X2-30 (Tobii Technology AB, Stockholm, Sweden) which tracks eye movements at a sampling rate of 30 Hz with a spatial accuracy of 0.4°. Children were seated at 60 cm and 36° above a 15' laptop screen. They were just instructed to look at the screen all the time. Before starting the task, a nine-points calibration process was running with an animated stimulus as target. In some cases, the calibration was repeated, if the child failed to follow the points. After calibration, the task consisted of visualizing the 36 slides pairing one face with one object. Each slide was presented for 5 seconds and was preceded by an animated fixation point (a minion) displayed for 1s to converge their attention to the center of the screen.

After the eye tracking task, children receptive vocabulary, affect recognition, and Theory of Mind (ToM) abilities were assessed using the previously mentioned PPTV-III (Dunn & Dunn, 1997) and the two subtests comprised in the social perception domain of the Developmental Neuropsychological Assessment (NEPSY-II: Korkman et al., 2007) to describe groups more thoroughly. These scales were chosen due to their wide usability for clinical and research purposes, and the fact that both yield typification data with special groups (including ASD and DLD). At the end of the experimental session, all children received a craft medal with a happy emoticon on it as reward for participating.

## 7.2.4 Data Analysis

Following a well credited approach, we developed three indexes to describe VSP of participants in each group: (1) Prioritization, latency of the first fixation to compare attentional orientation to both faces and objects, (2) Preference, proportion of the total fixation duration to each stimuli to analyze the distribution of visual attention between social and non-social images, and (3) Duration, mean time per visit to faces and objects to account for the depth of visual processing of each kind of stimuli (Sasson & Touchstone, 2014; Vacas et al., 2021c). These indexes resulted from the processing and transformation of some eye tracker parameters. Thus, the Prioritization index came from the raw data of Time to First Fixation; the Preference index resulted from the division of Total Fixation Duration towards a specific area of interest (AOI; e.g., happy faces paired with CIOs) by the Total Fixation Duration towards all AOIs; and the Duration index derived from the division of the Total Visit Duration to an AOI (e.g., angry faces paired with non-CIOs) by the number of visits to the same AOI. Data analysis was carried out using the Statistical Package for the Social Sciences, 25 (SPSS, 25, IBM, Armonk, NY, United States of America).

We performed normality and homoscedasticity tests to decide the most appropriate data analysis. Kolmogorov-Smirnov and Levene Tests yielded *p* values over .05; thus, we eventually determined to perform parametric tests. We performed two different sets of analyses: the former to describe the VSP to faces and the second one to define the VSP to objects in each group. Thus, to analyse attention to faces we conducted separate repeated measures ANOVAs on each index, with Group (ASD, DLD, control) as the between-group factor and the Type of object (CIO, non-CIO) and Emotion (happy, angry, neutral) as the within-group factors. On the other hand, to analyse attention to objects we conducted separate repeated measures ANOVAs on each index, with Group (ASD, DLD, control) as the between-group factor and the Type of object (CIO, non-CIO) and Emotion (happy, angry, neutral) as the within-group factors. On the other hand, to analyse attention to objects we conducted separate repeated measures ANOVAs on each index, with Group (ASD, DLD, control) as the between-group factor and the Type of object (CIO, non-CIO) as the within-group factor. Effects sizes were also included for all repeated measures ANOVAs (partial eta-squared,  $\eta_p^2$ ), its values are considered small effects when  $\eta_p^2 < .01$ ; medium effects when  $\eta_p^2 < .06$ ; and large effects when  $\eta_p^2 < .14$ .

## 7.3 Results

## 7.3.1 Faces Attention

## 7.3.1.1 Prioritization

No effect for any interaction was found. A main effect for Type of object was significant ( $F_{(1, 30)} = 12.40$ , MSE = .19, p = .01,  $\eta_p^2 = .29$ ), suggesting that participants of three groups attended to faces paired with non-CIOs earlier (880 ms) than to those paired with CIOs (1110 ms). To further examine between-group differences regarding this main effect, we performed individual one-way ANOVAs on faces paired with CIOs and non-CIOs separately. There was a marginal effect for Group with CIOs ( $F_{(2, 30)} = 2.71$ , MSE = .29, p = .08,  $\eta_p^2 = .15$ ), where the ASD group looked at faces significantly later compared to the control and DLD groups (see Table 7.2 left and Figure 7.2.A).

**Table 7.2.** Mean and standard deviation (in brackets) for each experimental condition in the Faces Attention (left) and Object Attention (right) analyses

FACES ATTENTION				OBJECT ATTENTION				
DV	Experimental Condition	ASD (n = 11) <i>M</i> ( <i>SD</i> )	DLD (n = 11) <i>M</i> ( <i>SD</i> )	TD (n = 11) <i>M</i> ( <i>SD</i> )	Experimental Condition	ASD (n = 11) <i>M</i> (SD)	DLD (n = 11) <i>M</i> ( <i>SD</i> )	TD (n = 11) <i>M</i> ( <i>SD</i> )
	Happy Face vs. CIO	1.18 (0.43)	0.92 (0.35)	1.02 (0.38)	CIO vs. Happy Face	1.08 (0.61)	0.80 (0.30)	1.05 (0.40)
Prioritization (TFF in ms)	Happy Face vs. non-CIO	0.90 (0.35)	1.00 (0.38)	0.77 (0.29)	Non-CIO vs. Happy Face	0.83 (0.39)	0.91 (0.34)	0.78 (0.31)
	Angry Face vs. CIO	1.36 (0.47)	1.05 (0.39)	1.11 (0.58)	CIO vs. Angry Face	0.84 (0.45)	0.96 (0.49)	0.69 (0.25)
	Angry Face vs. non-CIO	0.92 (0.46)	0.85 (0.46)	0.95 (0.44)	Non-CIO vs. Angry Face	0.84 (0.41)	0.94 (0.40)	0.91 (0.48)
	Neutral Face vs. CIO	1.34 (0.54)	0.99 (0.31)	1.02 (0.54)	CIO vs. Neutral Face	0.86 (0.46)	1.17 (0.48)	0.80 (0.37)
	Neutral Face vs. non-CIO	0.97 (0.41)	0.81 (0.46)	0.83 (0.39)	Non-CIO vs. Neutral Face	0.77 (0.36)	0.83 (0.44)	1.14 (0.40)
	Total Faces vs. CIO	1.29 (0.37)	0.99 (0.17)	1.05 (0.39)	Total CIO	0.92 (0.36)	0.98 (0.27)	0.85 (0.23)
	Total Faces vs. non-CIO	0.93 (0.24)	0.86 (0.36)	0.85 (0.22)	Total non- CIO	0.81 (0.21)	0.84 (0.30)	0.95 (0.31)
Preference (PTFD in	Happy Face vs. CIO	0.11 (0.03)	0.12 (0.04)	0.17 (0.04)	CIO vs. Happy Face	0.22 (0.07)	0.24 (0.07)	0.16 (0.06)
	Happy Face vs. non-CIO	0.18 (0.08)	0.18 (0.11)	0.17 (0.07)	Non-CIO vs. Happy Face	0.16 (0.07)	0.16 (0.07)	0.15 (0.07)

	Angry Face	0.16	0.17	0.18	CIO vs.	0.20	0.19	0.15
	vs. CIO	(0.07)	(0.08)	(0.08)	Angry Face	(0.05)	(0.07)	(0.05)
	Angry Face	0.15	0.15	0.20	Non-CIO vs.	0.15	0.18	0.13
	vs. non-CIO	(0.05)	(0.09)	(0.06)	Angry Face	(0.05)	(0.09)	(0.07)
	Neutral Face	0.11	0.09	0.16	CIO vs.	0.19	0.20	0.19
	vs. CIO	(0.06)	(0.09)	(0.06)	Neutral Face	(0.06)	(0.08)	(0.05)
		. ,	. ,				. ,	. ,
	Neutral Face	0.15	0.15	0.20	Non-CIO vs.	0.20	0.18	0.15
	vs. non-CIO	(0.05)	(0.07)	(0.05)	Neutral Face	(0.08)	(0.09)	(0.06)
	Total Faces	0.39	0.38	0.50	Total CIO	0.61	0.62	0.50
	vs. CIO	(0.11)	(0.13)	(0.13)		(0.11)	(0.13)	(0.13)
	Total Faces	0.48	0.48	0.57	Total non-	0.52	0.52	0.43
	vs. non-CIO	(0.15)	(0.21)	(0.13)	CIO	(0.15)	(0.21)	(0.13)
	Happy Face	0.60	0.73	0.91	CIO vs.	0.96	0.82	0.91
	vs. CIO	(0.25)	(0.34)	(0.20)	Happy Face	(0.31)	(0.24)	(0.45)
		. ,	. ,				. ,	. ,
	Happy Face	0.69	0.73	0.95	Non-CIO vs.	0.83	0.65	0.89
	vs. non-CIO	(0.39)	(0.27)	(0.40)	Happy Face	(0.56)	(0.38)	(0.31)
	Angry Face	0.66	0.73	0.94	CIO vs.	0.74	0.84	0.71
(SI	vs. CIO	(0.36)	(0.32)	(0.30)	Angry Face	(0.35)	(0.30)	(0.22)
Duration (TPV in ms)	Angry Face	0.71	0.65	1.10	Non-CIO vs.	0.63	0.89	0.80
V i	vs. non-CIO	(0.30)	(0.30)	(0.32)	Angry Face	(0.31)	(0.58)	(0.37)
EL)	Neutral Face	0.56	0.49	0.83	CIO vs.	0.88	0.98	0.96
on	vs. CIO	(0.23)	(0.25)	(0.85)	Neutral Face	(0.52)	(0.34)	(0.19)
ati	vs. CIO	(0.23)	(0.23)	(0.23)	Neutral Pace	(0.52)	(0.54)	(0.19)
m	Neutral Face	0.56	0.63	1.14	Non-CIO vs.	0.78	0.83	0.94
H	vs. non-CIO	(0.18)	(0.25)	(0.36)	Neutral Face	(0.38)	(0.34)	(0.28)
	Total Faces	0.60	0.66	0.90	Total CIO	0.85	0.87	0.84
	vs. CIO	(0.20)	(0.22)	(0.20)		(0.33)	(0.22)	(0.20)
	Total Faces	0.64	0.66	1.06	Total non-	0.72	0.74	0.88
	vs. non-CIO	(0.22)	(0.18)	(0.25)	CIO	(0.29)	(0.27)	(0.24)
	, j. non CiO	(0.22)	(0.10)	(0.23)		(0.2))	(0.27)	(0.24)

TFF, Time to first fixation; PTFD, Proportion of total fixation duration; TPV, Time per visit

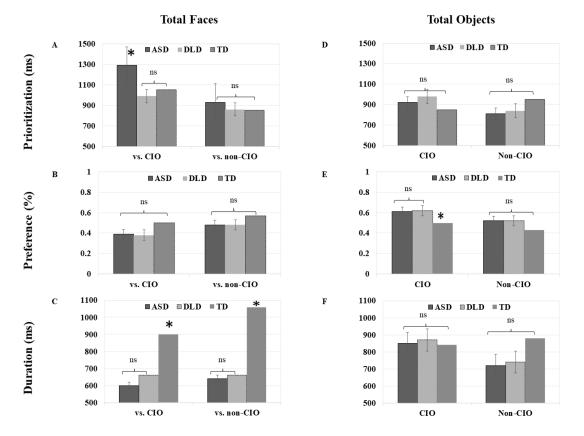


Figure 7.2. Visual attention pattern of total faces and objects

## 7.3.1.2 Preference

Repeated measures ANOVA did not yield any significant effect for Group × Type of object, Group × Emotion interaction or main effect for Group (see Figure 7.2.B), but a significant main effect for Type of object was revealed ( $F_{(1, 30)} = 12.40$ , MSE = .04, p = .00,  $\eta_p^2 = .39$ ), indicating that participants looked significantly less time at faces when they were paired with CIOs (0.42) compared to when they were with non-CIOs (0.51). A main effect for Emotion was also revealed ( $F_{(2, 29)} = 3.71$ , MSE = .01, p = .03,  $\eta_p^2 = .20$ ), specifically regarding anger and neutral faces ( $F_{(1, 30)} = 7.59$ , MSE = .04, p = .01,  $\eta_p^2 = .20$ ), where participants looked significantly more time at angry faces (0.17) compared to neutral ones (0.14). There was a marginal interaction between Emotion and Type of object ( $F_{(2, 29)} = 3.71$ , MSE = .01, p = .08,  $\eta_p^2 = .16$ ). Planned contrasts revealed that participants looked significantly less time to happy and neutral faces when they were accompanied with CIOs (happy:  $F_{(1, 30)} = 9.43$ , MSE = .03, p = .00,  $\eta_p^2 = .24$ ; neutral:  $F_{(1, 30)} = 15.70$ , MSE = .03, p = .00,  $\eta_p^2 = .34$ ) compared to when they were with non-CIOs (see Figure 7.3).

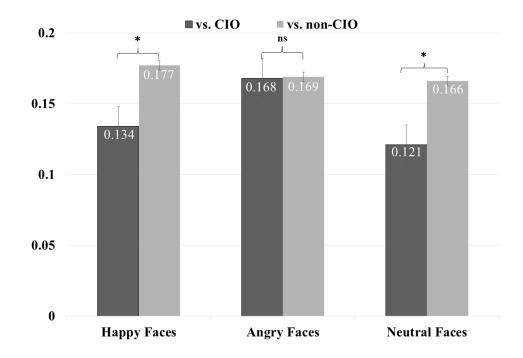


Figure 7.3. Visual preference for emotions regarding the competing object

#### 7.3.1.3 Duration

Again, the repeated measures ANOVAs yielded no effect for any interaction. However, a main effect for Group was highlighted ( $F_{(2, 30)} = 9.48$ , MSE = .26, p = .00,  $\eta_p^2 = .39$ ), indicating that the visit duration towards faces was significantly shorter in ASD (620 ms) and DLD (660 ms) groups with respect to that of the controls (970 ms; see Figure 7.2.C). Pairwise comparisons yielded p = .000 for control-ASD and p = .002 for control-DLD. An effect for Type of object was also revealed ( $F_{(1, 30)} = 5.40$ , MSE = .05, p = .02,  $\eta_p^2 = .15$ ), where participants made significantly shorter visits to faces when they were paired with CIOs (720 ms) compared to when they were with non-CIOs (790 ms; see Table 7.2 left).

# 7.3.2 Object Attention

# 7.3.2.1 Prioritization

Any effect for interaction or main effect was yielded, which implies that participants of all groups looked at both kind of objects at a similar speed (see Table 7.2 right and Figure 7.2.D).

# 7.3.2.2 Preference

No significant effect was yielded for the interaction or Group, but it was revealed a main effect for Type of object ( $F_{(1, 30)} = 18.70$ , MSE = .01, p = .00,  $\eta_p^2 = .38$ ), indicating that all participants spent more time looking at CIOs (0.58) than non-CIOs (0.49; see Table 7.2 right). To further examine between-group differences regarding this main effect, we again performed individual one-way ANOVAs on CIOs and non-CIOs separately. The ANOVA performed on CIOs highlighted a main effect for Group ( $F_{(2, 30)}$ = 3.66, MSE = .06, p = .04). Planned contrasts revealed significant differences between the ASD and DLD groups compared with the control one ( $F_{(1, 30)} = 2.70$ , MSE = .09, p =.01), which means that children from the clinical groups attended to CIOs significantly more than controls (see Figure 7.2.E).

## 7.3.2.3 Duration

Again, any significant effect was found for the interaction or Group, but a main effect for Type of object was highlighted ( $F_{(1, 30)} = 5.04$ , MSE = .02, p = .03,  $\eta_p^2 = .14$ ), implying that, in general, all participants made longer visits to CIOs than to non-CIOs (see Table 7.2 right and Figure 7.2.F).

## 7.4 Discussion

The present study had the purpose of comparing young children with ASD, DLD, and TD in terms of attention to faces and to objects (a key component within the complex construct of SA) considering the role of emotional expressions and competing objects in their VSP. As a whole, our results indicate that children in the clinical groups exhibited substantial differences with respect to their TD peers, whose implications will be discussed in the following paragraphs. Moreover, we found significant differences

between clinical groups where children with ASD looked at faces significantly later than their counterparts when these faces were paired with CIOs.

Thus, children from both clinical groups differed from controls in terms of significantly shorter visits to faces (showed in the Duration index of the faces attention analysis) and an excessive bias towards CIOs (highlighted by the Preference index of the object attention analysis). Therefore, we may say that, compared to TD children, those with ASD and DLD processed faces less thoroughly, and CIOs captured their attention disproportionately. The former assumption confirms our H1 as we had speculated that young children with ASD would display significantly less attention to faces than their TD counterparts. In this sense, our results are in line with those studies highlighting reduced SA, particularly attention to faces, in children with ASD (Franchini et al., 2017; Hanley et al., 2014; Hosozawa et al., 2012; Vacas et al., 2021c; Vivanti et al., 2011; von Hofsten et al., 2009; Zantinge et al., 2017). In this particular study, the reduced attention to faces showed by the ASD group was yielded by the Duration index, which accounts for the level of detail or the kind of processing that an individual displays when viewing stimuli and it is also related to the ability to maintain attention to a target stimulus (similar to sustained attention). In this sense, a more superficial visual processing of faces may drive these children to obtain an incomplete or flawed input of people in daily live situations, hindering their brain specialization, and consequently, impairing their social interactions (Jones et al., 2008; Klin, 2008; Sasson, 2006; Sasson et al., 2008; Sasson et al., 2011; Sasson & Touchstone, 2014; Shultz, 2005).

On the other hand, as we had not made any prediction about children with DLD, the fact that they performed similarly to those with ASD is a breakthrough. Previous literature applying different paradigms had pointed to a social-oriented VSP of faces in children with DLD according to the proportion of time they spent on looking at people (and their faces) during observational and interactive tasks (Hanley et al., 2014; Hosozawa et al., 2012). However, our results indicate that preschoolers with ASD and those with DLD showed a similar superficial visual processing of faces. Methodological differences may be behind these discrepancies between studies; for example, Hanley et al. (2014) and Hosozawa et al. (2012) applied different paradigms (not a paired preference one), they also presented their participants with different stimuli (dynamic and interactive ones) and included samples with different characteristics than ours in terms of age and

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gender. Nevertheless, the trend shown in our study must be explored more in depth to clarify the implications of the superficial visual processing of faces in children with DLD. In this sense, as it has been proposed for children with ASD, this kind of processing may be hampering their brain specialization, which could be the root of their problems with social relationships and would partially explain the generalized social-emotional impairment reported in children with DLD that impacts on the quality of their social interactions (St Clair et al., 2011; Vissers & Koolen, 2016).

Regarding our H2, we had predicted that emotional faces would drive to a different visual processing than neutral ones in all groups. This has been confirmed by the Preference index of the faces attention analysis. This index highlighted a bias toward angry faces in all participants regardless of the group. Not only did all children look at these faces significantly more proportion of time, but also angry faces could override the effect of the type of object on the attention to faces. This means that, despite the fact that CIOs reduced attention to faces in all children, this effect was absent for angry faces. This result supports the hypothesis of emotional sensitivity in children with ASD (Nuske et al. 2014, 2016; Vacas et al., 2021c; Vivanti et al. 2011) and it is in line with Fan et al. (2020) who also reported through a systematic review an attentional bias towards negative emotions in children with ASD. The finding of emotional sensitivity in the clinical samples also evidences some conserved abilities related to social-emotional processing in children with ASD and those with DLD: these children distinguish between emotions, process them differently according to their saliency, and attend to them in a similar way than their TD peers. This may have implications for diagnosis as well as for interventions. Firstly, it would be worthy to assess SA using neutral faces rather than emotional ones during the diagnostic process, as emotional expressions could mask difficulties in visual processing of faces. Secondly, exaggerating emotional expressions during intervention could be a useful strategy to capture and maintain children attention.

Concerning our H3 which claimed that the type of object would have a major impact on children in the ASD group, with CIOs (but not non-CIOs) reducing their attention to faces, we refuted that premise as CIOs had a great impact on all groups, significantly delaying visual orientation to faces, reducing attention to them (except in angry faces), and diminishing their processing. This effect was yielded in all analyses performed (except in the repeated measures ANOVA on Prioritization index in the

analysis of object attention); thus, we may say that the saliency of CIOs is highly influential for all children to a similar extent.

On the other hand, we found a marginal difference between groups in the Prioritization index of the faces attention analysis. This difference concerned children with ASD who looked at faces paired with CIOs significantly later than their counterparts of the other two groups. As this index is related to attentional orientation, the highlighted difference may be trivial or may be an indicator of non-social orientation in children with ASD. Regarding children with DLD, if this assumption was confirmed, it may imply that children with DLD (contrary to those with ASD) would have prosocial attentional orientation. Hence, we consider that this hypothesis should be tested with wider samples as Sasson & Touchstone (2014) had previously highlighted a greater effect of CIOs on visual attention to faces specifically in children with ASD.

On the other hand, as mentioned before, the Preference index in the analysis of object attention yielded significant differences between both clinical groups with respect to the controls in the proportion of time spent on looking at CIOs. Although these results are tentative due to the small samples of the three groups, they highlight some trends which are worthy to explore in future studies in order to confirm or rule them out, as it would have relevant implications for better understanding the roots of these disorders as well as for improving differential diagnosis among DLD and ASD.

Furthermore, we would like to highlight other findings derived from this study. Firstly, we believe that Prioritization index could be useful in differentiating between clinical conditions, as it has yielded a trend to atypicality specifically in children with ASD (observed in planned contrasts). If this trend were confirmed in studies with wider samples, its implications for differential diagnosis would be highly relevant. The link of this index to unconscious attentional orientation could help disentangle the different etiologies of ASD and DLD, as children with DLD would be more socially oriented at first than their counterparts with ASD (although, regarding the conscious attentional processes, individuals with both disorders would avoid faces similarly).

Secondly, Preference and Duration indexes have proved to be valuable at identifying clinical samples like those involved in this study. As these indexes have highlighted clear differences in attention to faces (Duration) and objects (Preference) Chapter 7. Visual Behavior as Differential Marker for Early Diagnosis in Young Children with Autism Spectrum Disorders and Developmental Language Disorder. An Eye Tracking Comparative Study
between children with ASD and those with DLD compared to their TD peers, they may
be psychophysiological markers of the presence of both conditions. The Duration index
had already been stressed as an attentional psychophysiological marker of ASD in our
previous study (Vacas et al., 2021c), but these new findings extend our previous ones by
adding the Preference index in the object attention analysis to the potential attentional
psychophysiological marker in this condition.

Thirdly, between groups differences in this study were mostly circumscribed to the analysis of social stimuli (e.g., the Duration index yielded significant differences between clinical and control groups exclusively in the faces attention analysis, not in the object attention one). This suggests that atypicalities found in attention to faces in the clinical samples are not likely to be due to difficulties in general visual processing (as participants in both clinical groups viewed objects in a similar way than their TD counterparts), but they are probably unveiling specific difficulties with social content.

In summary, this work was designed as a comparative study to dive into the possibilities of using the eye tracking methodology and paired preference tasks as tools to improve differential diagnosis in young children with ASD or DLD. In this sense, the small sample size could explain the lack of between groups differences which, in some cases, disagree with previous literature. Despite that fact, we believe that our results open a pathway to the differential study of these populations with this methodology, highlighting the importance of considering the study of SA in children with DLD and unveiling subtle differences between clinical groups which deserve to be more investigated. Likewise, in this study we tried to assess receptive vocabulary, affect recognition, and ToM in our participants, but we failed to obtain data from a significant number of children from the clinical groups (specially during the application of the NEPSY-II) due to their young age and the high demands of both subscales. In this sense, it was noteworthy that the number of data lost in the assessment of affect recognition and ToM was related to the specific condition (10 children from the ASD group, 6 from the DLD group, and none from the control one). For that reason, we believe that future studies should include more scales to assess participants cognitive abilities and to perform correlation analyses in order to describe more thoroughly the clinical samples. Finally, if possible, upcoming research should divide the sample in different groups regarding laguage and cognitive abilities to put to test the hypothesis of a continuum of impairment

between ASD and DLD and the existence of a subtype of ASD which goes along with language impairment (Bishop, 2000, 2003, 2010; Bishop & Norbury, 2002; Conti-Ramsden et al., 2006; Georgiou, & Spanoudis, 2021; Leyfer et al., 2008; Taylor & Whitehouse, 2016; Tomblin, 2011).

## 7.5 Conclusions

The main aim of this comparative study was to find differences between children with ASD and those with DLD at early ages in visual processing of faces and objects, to contribute to early differential diagnosis and to the current state-of-the-art about the similar or distinct etiologies of both conditions. Our results did not yield several differencies between clinical groups, but they showed some trends which are convenient to explore with wider samples. Furthermore, this study has disentangled some atypicalities in attention to faces in children with DLD, which had never been explored. In this sense, our data highlighted similar difficulties in VSP of social stimuli in both clinical populations, which differentiate them from their TD peers. These difficulties have been extensively investigated in the population with ASD, but they had been disregarded in children with DLD, which is suprising due to the widely reported high rates of social difficulties, psychopathology (internalizing and externalizing problems), and bullying affecting the quality of their social relationships among children and adolescents with DLD (St Clair et al., 2011; Toseer et al., 2021; Vissers & Koolen, 2016; Wadman et al., 2011). In this sense, we believe that including DLD samples in the study of SA could contribute not only to better orientate their interventions, but also to facilitate differential diagnosis with similar disorders at early ages. This along with the fact that early diagnosis has proved to improve intervention and prognosis in children with several neurodevelopmental disorders support the continuity of this line of research.

## Acknowledgements.

We gratefully acknowledge to all families and children who participated in this study. We extend our sincere acknowledgement to those Early Intervention Centers of the province of (Córdoba) and the public-school Al Andalus, which kindly accepted our request of participation and facilitated the labor of the researchers.

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# Third Part. Discussion and Conclusions

#### **Chapter 8. Discussion and Conclusions**

#### 8.1. Discussion

At this point, it is noteworthy to remind the reader the reason why we started this project. Our main purpose was always to contribute to early differential diagnosis of certain NDDs whose common feature was the presence of language impairment. On the other hand, there was a sound body of knowledge supporting the relevance of early diagnosis and intervention for improving children prognosis. Thus, with this idea in mind we run our first study on cognitive profiles in school-aged children with SLD, SLI, AS, and two subtypes of ADHD (predominantly inattentive and combined). From this study (see Appendix 1), we concluded that cognitive profiles were not highly enlightening regarding the distinction of the selected populations and that we needed to work with younger samples in order to make a real contribution to field of ECI.

Hence, we sought into the literature new possibilities to address our goal and we found that DLD and ASD were two conditions with which professionals had many difficulties at early ages due to their many phenotypic similarities. Moreover, we also learnt that some abilities comprised in the EC (particularly the attention and visual processing of social stimuli) were remarkably informative in describing these conditions. That was how we set the focus and the final purpose of this work.

Therefore, the final aim of this doctoral dissertation was threefold: (1) To analyze similarities and differences between young children with DLD and those with ASD in terms of social-emotional abilities, (2) To make use of the potential of the eye tracking technology to fill the gaps of the behavioral tests regarding the description and differentiation of these disorders, (3) To contribute to early differential diagnosis by providing specific markers of each condition.

In this sense, the fulfilment of the previous mentioned aims would result in relevant implications for the field: (1) From a theoretical standpoint, we would advance in the knowledge of DLD and ASD; (2) From a methodological perspective, we could

consolidate an effective assessment methodology which may be included in clinical and research protocols; and (3) From a practical view, the detection of differential markers between conditions would improve clinical practice, having a real impact on all the stakeholders (children, families, professionals, and ultimately, the society).

After a comprehensive review of the state of the art (presented in chapters one to three), we realized that there were important shortcomings regarding the understanding of the social-emotional competence in children with these conditions. On the one hand, the vast and highly heterogeneous amount of literature on children with ASD had yielded very inconsistent results: most research pointed to deficits in FER, ToM, or emotion regulation (among other social-emotional abilities) in this population, but these results had few reliability due to the wide heterogeneous methodologies and tasks applied to assess these constructs. On the other hand, literature on children with DLD was so scarce that we could barely draw any conclusion about their profile. Thus, we decided to undertake a set of intertwined experimental studies to elucidate some questions derived from our aims.

Among all domains tackled in the literature, visual processing of faces drew our attention for three main reasons: (1) It reflects unconscious attentional mechanisms which set the basis for some more complex social-emotional abilities (e.g., FER or ToM); (2) Faces are one of the main gateways to perceive, recognize, and understand others' inner emotional states (Frith & Frith, 2006; Klin, 2008), thus they may be useful for the evaluation of populations with marked social difficulties; and (3) Attention to faces may be assessed objectively through the use of cutting-edge technologies such as eye-tracking.

Therefore, we designed three eye tracking studies comparing children with ASD and controls (studies 1 and 2), and later an additional sample of children with DLD (study 3) in terms of VSP of faces in different conditions. In this sense, we operationalized the VSP on the basis of three indexes: Prioritization (related to attentional orientation), Preference (reflecting visual preference), and Duration (linked to sustained attention and the kind of processing). As these indexes referred to a different variable, it was not mandatory that all indexes yielded significant differences between groups. In fact, the main strength of this way of operationalizing the VSP was that each index provided different information on a specific feature of individuals' visual processing, which allowed to get a comprehensive view of how children with ASD and those with DLD processed faces and which were their particular difficulties.

Furthermore, we applied a paired preference paradigm, which implied presenting participants with matched stimuli to weight the effect of different variables (type of object, emotional expression, poser's gender, or poser's age) on their VSP toward faces and objects. This paradigm alongside the eye tracking technology allowed us to assess different visual parameters and compare participants' visual behavior regarding each one of them. Although this paradigm and the eye tracking technology had been applied in several previous studies in the field (Harrop et al., 2018; Sasson & Touchstone, 2014; Unruh et al., 2016), we consider that in this project we have tested the whole methodology and demonstrated its potential in describing and comparing disorders. Thus, this could be stated as a powerful clinical and research tool in the future after more research confirming its validity and reliability for the assessment, identification, and differentiation of similar NDDs.

Hence, in order to achieve the general goals of this project, we applied three intertwined tasks with which we assessed children's VSP toward faces considering the effect of non-social competing object, the competing emotional expression, the poser's gender, and the poser's age in studies 1 and 2. On the other hand, we reproduced the experiment 1 adding a sample of children with DLD with the aim of unveiling differential psychophysiological markers between conditions and, subsequently, to contribute to differential diagnosis in study 3. Table 8.1 shows the main contribution of each study to the general aim of this project.

Thus, results from study 1 revealed three atypicalities in the VSP of children with ASD. First of all, they showed an attentional bias away from angry faces yielded by the Prioritization index, as they looked at these faces significantly later than their TD counterparts. Secondly, they displayed a significantly reduced scanning of faces as the Duration index revealed, implying difficulties in sustained attention or, at least, a superficial visual processing of faces (not of objects). And thirdly, despite the fact that CIOs impacted on all children similarly, non-CIOs had a major effect on those with ASD as highlighted by the Prioritization and Preference indexes of the object attention analysis. This reinforces the well-known excessive appealing for objects in children with ASD regardless of its typology.

Study	Outcomes				
Study 1. Social Preference	Three atypical visual behaviors (potential psychophysiological markers) in children with ASD as compared to their TD peers:				
	- Later orientation to angry faces (Prioritization Index).				
	- More superficial scanning of faces (Duration Index).				
	- Greater distracting effect of non-CIOs (Prioritization and Preference Indexes).				
	Three typical visual behaviors in children with ASD (conserved abilities):				
	- Emotional sensitivity (Prioritization and Preference Indexes).				
	- Social oriented VSP when faces competed with non-CIOs				
	(Prioritization, Preference, and Duration Indexes).				
	- Greater saliency of CIOs over non-CIOs (Prioritization, Preference, and Duration Indexes).				
Study 2. Emotional Preference	Three atypical visual behaviors (potential psychophysiologica markers) in children with ASD with respect to their TD counterparts: - Later orientation to angry faces (Prioritization Index).				
	- Later orientation to child faces (Prioritization Index).				
	- Later orientation to clind faces (Prioritzation index). - More superficial scanning of faces (Duration Index).				
	Two typical visual behaviors in children with ASD (conserved				
	abilities):				
	- Emotional sensitivity (Prioritization, Preference, and Duration Indexes).				
	- Typical visual preference for child faces (Preference Index).				
Study 3. Comparative Study	Two atypical visual behaviors (potential psychophysiological markers) in both clinical groups in contrast to the control one (similarities between disorders):				
	- Greater distractive effect of CIOs (Preference Index).				
	- More superficial scanning of faces (Duration Index).				
	Two typical behaviors in children with ASD and those with DLE (conserved abilities):				
	- Emotional sensitivity (Preference Index).				
	- Greater saliency of CIOs over non-CIOs (Prioritization, Preference, and Duration Indexes).				
	A marginal difference between disorders (potential differentia psychophysiological marker):				
	- Greater distractive effect of CIOs (Prioritization Index).				

Table 8.1. Main outcomes of the experimental eye tracking studies

With respect to the study 2, it replicated previous findings on an attentional bias away from angry faces and reduced scanning of faces in children with ASD compared to their TD peers as shown by the Prioritization and Duration indexes. Furthermore, this study extended previous results by yielding a new bias away from child faces in children with ASD, again revealed by the Prioritization index.

Finally, the study 3 again replicated previous results on the reduced scanning of faces in children with ASD through the Duration index. This atypical behavior was also found in the group of children with DLD. Moreover, this study extended previous results

by highlighting a major effect of CIOs on both clinical populations, which means that these type of objects specifically prevented children with ASD and those with DLD from looking at faces as shown by the Preference index. Any significant difference was yielded between clinical conditions, but children with ASD looked at faces paired with CIOs marginally later than their peers from the other two groups. This may be an indicator of a specific effect of CIOs on children with ASD (which may contribute to differential diagnosis), or it could be a due to the sample size in our study. Regardless of the explanation, we consider that a deeper examination of this hint with wider sample sizes would be worthy.

Despite the mentioned atypicalities in VSP of faces in children with ASD and, to a lesser extent in children with DLD, we did also find similarities between clinical and control groups, which suggests that some abilities are intact in both conditions (see Table 8.1). Thus, in line with previous studies we found evidence of emotional sensitivity in children with ASD and those with DLD (Nuske et al., 2014, 2016; Vacas et al., 2021c; Vivanti et al., 2011). Throughout out three studies, we found a specific scanning for emotional faces (specially for angry ones) different to that of neutral faces. Specially, children with ASD showed a bias away from angry faces as they orientated to them later than their TD peers. Conversely, the Preference and Duration indexes of experiment 1 in study 2 yielded a particular significant visual trend to angry faces in terms of more visual attention and longer visits to them compared to the other emotional expressions. This particular trend was found in the ASD and TD groups, and it was replicated in study 3 for all the three groups (in this case, only by the Preference index).

The mixed results found in children with ASD regarding angry faces (late attentional orientation but greater visual preference and longer visit duration) may indicate difficulties in this population to orient initially to threatening faces, which could relate to adaptive difficulties given the phylogenetic relevance of this kind of stimuli (Pérez-Dueñas et al., 2014). However, the typical visual preference for angry faces in the population with ASD is in line with conclusions from Fan et al. (2020). These authors demonstrated an attentional bias towards threatening faces in individuals with ASD after reviewing several studies applying behavioral paradigms other than eye tracking. With respect to this visual behavior towards angry faces found in children with DLD, as we did not have made any previous assumption about this population in terms of emotional

sensitivity, we considered that this finding is interesting for the definition and better understanding of this condition.

Regarding emotional sensitivity, it is noteworthy to comment on a discrepancy between studies 1 and 2 concerning visual preference in children with ASD. Results from study 1 yielded typical preference for happy faces in children with ASD (while preference for angry and neutral faces was significantly less than in controls); on the other hand, study 2 revealed less preference for happy faces compared to the TD group and typical preference for angry faces. This apparent contradiction could be explained by the distinct nature of each study, which determined the use of different strategies for pairing stimuli and analyzing data. Thus, in study 1 we mainly aimed at weight the effect of the type of object (and to a lesser extent the impact of emotion) on participants' VSP of faces; while in study 2 we specifically analyzed the influence of emotional expressions (along with the poser's gender and age) on the VSP of faces. These differences in terms of aims conditioned the task design and the data analysis. Hence, the number of trials per condition was different in both studies; this alongside the fact that happy faces were competing with objects in study 1 but they competed with other emotional faces in study 2 may account for the mentioned discrepancy. In this sense, we consider that results derived from study 2 are more robust to define the sample of children with ASD, as we isolated the variable of emotional expression to test specifically the effect of emotionality.

On the other hand, we also found a similar pattern in all groups regarding the impact of the type of objects on VSP of faces. Thus, children of the three groups oriented earlier, demonstrated more visual preference, and made longer visits to faces paired with non-CIOs than to those paired with CIOs (see Table 8.1). This pattern was consistent among all indexes and was replicated in studies 1 and 3.

Finally, we found a specific VSP for child faces in children with ASD and their TD peers. As occurred with angry faces, we again found mixed results in children with ASD regarding child faces. These children oriented their gaze to them later that their TD peers; however, once detected, they preferred them over adult ones as much as their typical counterparts. This may imply an initial difficulty in children with ASD to engage with peers that eventually disappear in favor of a typical preference for them. This hypothesis must be tested in future studies with wider samples and alongside a more thorough assessment of the level of social impairment of the clinical sample in order to

dive into potential correlations. Furthermore, we are in process of examining this behavior in children with DLD, as it would be interesting to weight the effects of emotionality, poser's gender, and poser's age on VSP of faces in these children too.

Taken together, the mentioned results suggest that children with ASD show difficulties with attentional orientation towards relevant social stimuli (e.g., threatening faces and peer faces) and a reduced scanning of faces. This pattern of result seems consistent as it was replicated across studies. Thus, we may say that Prioritization and Duration indexes proved to be valuable in distinguishing the ASD condition from typical development. The Preference index also yielded interesting results, but they were less consistent and mostly pointed at similarities between groups. This atypical visual processing of faces (which are crucial gateways to others' inner states, thus determining social interactions) may be at the root of the social difficulties of children with ASD, which have been widely reported (Jones et al., 2008; Klin, 2008; Sasson, 2006; Sasson et al., 2011; Sasson & Touchstone, 2014; Shultz, 2005).

Results on the sample of children with DLD are tentative but not less important. The Preference and Duration indexes here seemed to be more revealing to differentiate clinical groups from controls; however, the Prioritization index revealed a marginal trend that, in case of being confirmed, it could help distinguish between clinical conditions.

On the other hand, it is also important to remark that processing difficulties found in both disorders were circumscribed to social stimuli. This is relevant insofar as it does not imply a deficit in global processing (objects were processed as in depth as their TD peers), but a specific impairment in processing of faces.

At this stage, after discussing the main contributions derived from this doctoral dissertation, we may comment on the general purposes of the project. Regarding the former (to analyze similarities and differences between young children with DLD and those with ASD in terms of social-emotional abilities), we may say that we have unveiled some psychophysiological markers in children with ASD (e.g., late orientation to angry and child faces, reduced scanning of faces, and major influence of objects -mainly CIOs, but also non-CIOs- on their VSP toward faces) which help understand their widely reported difficulties in SA (Chevallier et al., 2015; Chita-Tegmark, 2016a, 2016b; Falck-Ytter et al., 2013; Frazier et al., 2017; Guillon et al., 2014; Guillon et al., 2016; Liberati et al., 2017; Nakano et al., 2010; Sasson et al., 2008; Zantinge et al., 2017).

Evidence on the profile of children with DLD is more diffused due to the reduced opportunity to work with them. However, our results suggest that their visual behavior at early ages could be very similar to that of children with ASD which, in case of being confirmed, would imply a common impairment in the processing of faces. Conversely, if the marginal differential trend found between conditions was confirmed, it would mean that each disorder had a distinctive characterization. In any case, we consider that this doctoral dissertation has highlighted the potential of comparative studies with eye tracking methodology to disentangle psychophysiological markers in both clinical populations.

This relates to our second and third aims (to make use of the potential of the eye tracking technology to fill the gaps of the behavioral tests regarding the description and differentiation of these disorders, and to contribute to early differential diagnosis by providing specific markers of each condition). Despite the fact that we did not have the opportunity to count on a significant sample size in the group of children with DLD, data of our third study revealed interesting hints which deserve to be replicated with wider samples due to their potential contributions for the understanding and differentiation of both clinical conditions.

To close this section, it is worth mentioning the main contributions of our preliminary studies to this dissertation. As previously mentioned, this project started with a study on cognitive profiles of school-aged children with different NDDs (SLD, SLI, AS, and ADHD in two subtypes -predominantly inattentive and combined-). This study revealed the potential of the pairwise discrepancy analysis of the indexes from the WISC-IV for the evaluation of the heterogeneity of cognitive profiles in the selected populations (see Appendix 1). Secondly, it yielded a particular profile in children with SLI which made them distinguishable from the other clinical samples. Thirdly, it set the bases to reorient our work by guiding us toward a younger sample with different clinical conditions and a different construct assessed with a specific methodology.

Finally, our second preliminary study on diagnostic migration (see Appendix 2) validated our change of route confirming that the early diagnosis of DLD and ASD was a real challenge for clinicians. Thus, in this study we realized that diagnostic migration was a current phenomenon which concerned professionals in ECI. Moreover, we learnt about the viewpoint of practitioners and we had some feedback from them. This fact was

important for us because, in our opinion, it connects the utility and applicability of this project with the reality of clinical practice as it arose from a real concern.

## 8.2. Limitations and Future Directions

Some limitations of this doctoral dissertation are noteworthy. On the one hand, the major challenge we had faced during the development of this project has been the difficulty to access to the clinical sample. When we first contacted with a wide range of Early Childhood Intervention Centers (ECICs), the project was welcomed; however, after several communications, we lost contact with some centers.

Moreover, we faced the reticence of some families to let their children take part in the project because of their young age or difficulties in coping with diagnosis. These difficulties converged with the current pandemic, which dramatically constrained the possibilities of access to the sample, and whose effects are still noticeable.

Another difficulty related to the sample recruitment was the fact that diagnoses of DLD as well as those of ASD are delayed in many cases because of the unclear profiles that young children usually present. This alongside the requirement of some level of cognitive functioning in terms of sustaining attention and ability to understand and follow instructions in our participants substantially impacted on the sample sizes of our studies (mainly the comparative one). That is the reason why we are still involved in a recruiting process to get a wider sample of children with DLD and those with ASD.

Therefore, our plan is to continue this project to increase the sample size of both groups and to go further in future replication studies in which we applied extra measures to control for cognitive variables and analyze potential correlations between cognitive and social-emotional aspects.

We also find interesting to include measures on emotion recognition and ToM to explore the potential relation between visual processing of faces and these abilities. The problems of some participants to pass the affect recognition and ToM subtests of the NEPSY-II is another limitation of this project, as it prevented us from testing the potential correlations. Thus, it is also our aim to incorporate new measures of these abilities in future studies.

In our view, replication studies with wider samples could be helpful in several ways: (1) To elucidate if the marginal indicator of later orientation to faces competing

with CIOs found in children with ASD is an actual psychophysiological marker of this condition and, if that was the case, if it would contribute to differentiate this disorder from DLD at early ages; (2) To clarify the rationale for the mixed visual behavior to angry and child faces in children with ASD (late prioritization but high preference); (3) To better describe and comprehend the underlined attentional mechanisms in children with DLD, as we have verified that little is known about this population in this regard.

In line with that, another limitation of this project has been the lack of replication of the tasks applied in study 2 with the sample of children with DLD in order to weight the effects of emotionality, poser's gender, and poser's age on VSP of faces in these children too. In this sense, as we mentioned before, we are still working on this line of research.

Finally, we also consider relevant for the wider comprehension of ASD and DLD, the development of longitudinal studies to follow up on the course of both disorders and the potential effect of ECI particularly on their visual processing of faces, but also more globally on other abilities of the social-emotional competence. In this sense, we must highlight that our clinical samples had been receiving intervention for several months as they were associated to an ECIC before being recruited for our studies. This fact may have impacted on our results (mainly regarding those abilities which were revealed intact in both samples). Thus, studies assessing newcomer children in the ECICs could allow to get a cleaner profile of these populations before receiving intervention. This is another line of research that we are currently addressing due to our strong link with the ECIC associated to the University of Cordoba.

## 8.3. Conclusions

After discussing the main results derived from this work and commenting on the main limitations and suggestions for future research, we consider relevant to remark and summarize the main conclusions to culminate this doctoral dissertation:

1) From the first part of this work, we highlight that current literature on social-emotional competence in children with ASD as well as in those with DLD shows great weaknesses. Regarding the former group of children, there is a large number of studies addressing different abilities of this competence in this population; however, they yielded inconsistent results which draw to ambiguous

conclusions. With respect to children with DLD, literature is insufficient which makes difficult to get an impression of their social-emotional profile. Similarly, concerning the comparison between disorders, this dimension has also been inexplicably disregarded, which hinders differential diagnosis at early ages as linguistic and cognitive profiles are very similar in both conditions specially during the infancy, toddlerhood, and childhood.

2) Given the beneficial effects of early diagnosis and intervention, it is mandatory to foster studies addressing the comparison between disorders. In this sense, it is equally important to explore different developmental areas that may help distinguishing conditions and make use of objective measures which may yield reliable data and, subsequently, lead to robust conclusions. In this sense, social-emotional competence has proved to be helpful in disentangling differences between disorders even at early ages. Moreover, the eye tracking methodology has also demonstrated to effectively adapt to the specificities of young children with ASD and those with DLD, and to be useful in yielding objective distinctive psychophysiological markers.

3) In line with the previous conclusion, in the second part of this work we noted that young children with ASD displayed an idiosyncratic VSP of faces. This is a strong conclusion given that it was replicated in all our three studies. In this regard, we unveiled difficulties in attentional orientation to angry and child faces in this population, an excessive effect of non-social objects on SA toward faces, and a reduced scanning of faces compared with their typical counterparts. This visual processing style of people' s faces may explain to some extent their problems to deal with social exchanges, especially with their peers. On the other hand, we also found intact visual behaviors which are also relevant from a clinical point of view (e.g., typical preference for angry and child faces once they engage with them, and emotional sensitivity).

4) Regarding the population with DLD, as we barely had previous knowledge on them to make predictions about their performance, the finding of an atypical VSP of faces similar to that of children with ASD was highly relevant. Although our results on young children with DLD are tentative and still require replication, we consider that these have significant implications for research as well as for clinical practice. On the one hand, they highlight a serious gap in research on this

population, which translates into an incomplete and distorted description of the disorder. On the other hand, if these children actually have difficulties in visual processing of faces, it is likely that practitioner are neglecting a critical aspect when intervening with them. In any case, more research at this respect is ensure in order to elucidate the VSP of faces in this population.

5) Finally, the third part of this work emphasizes the potential of these eye tracking studies to disentangle differences in social-emotional profiles of children with ASD and DLD, contributing not only to improve the understanding of each disorder but also to differential diagnosis of both conditions. Therefore, we provided some useful guidelines to continue research in this field overcoming the limitations of this work. Among them, the most relevant lines for future research are the replication of our three tasks with wider samples of both disorders and the inclusion of extra measures of other social-emotional, linguistic, and cognitive abilities to test for potential correlations between these domains and visual processing of faces in both populations. Moreover, longitudinal studies and comparative research are also welcomed in order to complete the knowledge and comprehension of both clinical conditions.

In conclusion, we consider that the main contributions of this doctoral dissertation can be summarized in four points: (1) It provides a thorough review of the state of the art of the social-emotional competence in children with ASD and those with DLD; (2) It tests and consolidates a specific methodology combining eye tracking technology with a paired preference paradigm as a way to assess, identify, describe, and compare children with ASD and those with DLD at early ages; (3) It yields robust psychophysiological markers in children with ASD and tentative hints of visual processing of faces in children with DLD; and (4) It provides suggestions for future directions in the research on the field. In this sense, we consider that this dissertation has served for a threefold purpose: (1) It has added some knowledge to the theoretical conceptualization of ASD and DLD; (2) It has consolidated a specific methodology for assessing young children with these disorders (and possibly with other similar NDDs); and (3) It has yielded relevant psychophysiological markers of each condition with the subsequent benefit for clinical practice in ECI.

## **Report on Papers Resulting from this Dissertation**

## **Published Papers**

Vacas, J., Antolí, A., Sánchez-Raya, A., Pérez-Dueñas, C., & Cuadrado, F. (2021). Visual preference for social vs. non-social images in young children with autism spectrum disorders. An eye tracking study. *PLoS ONE*, 16(6), e0252795. https://doi.org/10.1371/journal.pone.0252795

<u>Indexation</u>: AGRICOLA; Chemical Abstracts Service (CAS); EMBASE; FSTA (Food Science and Technology Abstracts); GeoRef; Google Scholar; Journal Citation Reports; MEDLINE; PsychInfo; PubMed; Science Citation Index Expanded (SCIE); Scimago Journal & Country Rank (SJR); Scopus; Zoological Record.

Impact Factor:

Journal Citation Report (JCR) 2020

Impact factor: 3.240 (Q2)

Position 26 out of 73 (Multidisciplinary Sicences)

Scimago Journal & Country Rank (SJR) 2020

Impact factor: 0.990 (Q1)

Position 13 out of 135 (Multidisciplinary)

<u>Indexation</u>: ANVUR; CNKI; Current Contents (Social & Behavioral Sciences); Dimensions; EBSCO; EMCare; Google Scholar; Institute of Scientific and Technical Information of China; Japanese Science and Technology Agency (JST); Journal Citation Reports (Social Sciences Edition); Naver; Norwegian Register for Scientific Journals and Series; OCLC WorldCat Discovery Service; ProQuest; PsycINFO; Scimago Journal & Country Rank (SJR); Scopus; Social Science Citation Index (SSCI); TD Net Discovery Service; UGC-CARE List (India).

Impact Factor:

Journal Citation Report (JCR) 2020 Impact factor: 2.600 (Q2) Position 35 out of 78 (Psychology, Developmental) Scimago Journal & Country Rank (SJR) 2020 Impact factor: 0.856 (Q2) Position 39 out of 84 (Behavioral Neuroscience)

Vacas, J., Antolí, A., Sánchez-Raya, A., & Pérez-Dueñas, C. (2021). Eye Tracking Methodology for Studying Emotional Competence in Children with Autism Spectrum Disorder (ASD) and Specific Language Impairment (SLI): a Comparative Research Review. *Review Journal of Autism and Developmental Disorders*. Advance online publication. https://doi.org/10.1007/s40489-021-00261-y

Position 187 out of 537 (Psychiatry and Mental Health)

 Vacas, J., Antolí, A., Sánchez-Raya, A., & Pérez-Dueñas, C. (2021). Emotional Competence in Children with Autism Spectrum Disorders and Specific Language Impairment: A Comparative Research Review. *Education and Training in Autism* and Developmental Disabilities, 56(3), 306–327. https://www.proquest.com/scholarly-journals/emotional-competence-childrenwith-autism/docview/2570258568/se-2?accountid=14520.

<u>Indexation</u>: Journal Citation Reports; Scimago Journal & Country Rank (SJR); Social Sciences Citation Index (SSCI).

Impact Factor:

Journal Citation Report (JCR) 2020

Impact factor: 1.578 (Q3)

Position 54 out of 74 (Rehabilitation) and position 32 out of 44 (Education, Special)

Scimago Journal & Country Rank (SJR) 2020

Impact factor: 0.649 (Q2)

Position 154 out of 339 (Developmental and Educational Psychology) Position 370 out of 1544 (Education)

Vacas, J., Antolí, A., Sánchez-Raya, A., & Cuadrado, F. (2020). Análisis de Perfiles Cognitivos en Población Clínica Infantil con Trastornos del Neurodesarrollo. *Revista Iberoamericana de Diagnóstico y Evaluación – e Avaliaçao Psicológica* (*RIDEP*), 54(1), 35-46. https://doi.org/10.21865/RIDEP54.1.03

<u>Indexation</u>: Cengage; Ebsco; Journal Citation Reports (Social Sciences Edition); Latindex; Psicodoc; PsycINFO; Redalyc; Scimago Journal & Country Rank (SJR); Scopus; Social Science Citation Index (SSCI).

Impact Factor:

Journal Citation Report (JCR) 2020 Impact factor: 1.000 (Q4) Position 115 out of 131 (Psychology, Clinical) Scimago Journal & Country Rank (SJR) 2020 Impact factor: 0.359 (Q3) Position 153 out of 263 (Psychology, Miscellaneous)

Vacas, J., Antolí, A., Sánchez-Raya, A., Pérez-Dueñas, C., & Cuadrado, F. (2021). Migración diagnóstica entre Trastorno Específico del Lenguaje y Trastorno del Espectro Autista: Estudio exploratorio del impacto en los Centros de Atención Infantil Temprana. *Revista de Investigación en Logopedia*, 11(Special Issue), 77-88. https://dx.doi.org/10.5209/rlog.70221

<u>Indexation</u>: Academic Journal Database; Dialnet; Directory of Open Access Journals (DOAJ); Emerging Sources Citation Index (ESCI); ERIH Plus; FECYT; Índices CSIC; Journal Citation Reports; Latindex; MIAR; Psicodoc; REDIB; ROAD; Scimago Journal & Country Rank (SJR); Scopus; Ulrich's Periodicals Directory.

Impact Factor:

Journal Citation Report (JCR) 2020 Journal Citation Indicator (JCI): 0.19 (Q4) Position 231 out of 259 (Linguistics) Scimago Journal & Country Rank (SJR) 2020 Impact factor: 0.154 (Q2) Position 438 out of 911 (Language and Linguistics)

## **Under Review Papers**

Vacas, J., Antolí, A., Sánchez-Raya, A., Pérez-Dueñas, C., & Cuadrado, F. Social attention and autism in early childhood: Evidence on psychophysiological markers based on visual scanning of emotional faces with eye tracking methodology.

## **In-Process Paper**

Vacas, J., Antolí, A., Sánchez-Raya, A., Pérez-Dueñas, C., & Cuadrado, F. Visual Behavior as Differential Marker for Early Diagnosis in Young Children with Autism Spectrum Disorders and Developmental Language Disorder. An Eye Tracking Comparative Study.

## PLOS ONE

#### RESEARCH ARTICLE

## Visual preference for social vs. non-social images in young children with autism spectrum disorders. An eye tracking study

# Julia Vacas <sup>1,2,3</sup> , Adoración Antolí<sup>1,2,3,4</sup>, Araceli Sánchez-Raya<sup>1,2,3,4</sup>, Carolina Pérez-Dueñas<sup>1,2,3</sup>, Fátima Cuadrado<sup>1,2,3</sup>

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#### OPEN ACCESS

Citation: Vacas J, Antoli A, Sánchez-Raya A, Pérez-Dueñas C, Cuadrado F (2021) Visual preference for social vs. non-social images in young children with autism spectrum disorders. An eye tracking study. PLoS ONE 16(6): e0252795. https://doi.org/10.1371/Journal.cone.0252795

Editor: Robert Didden, Radboud University, NETHERLANDS

Received: November 16, 2020

Accepted: May 24, 2021

Published: June 4, 2021

Peer Review History: PLOS recognizes the benefits of transparency in the peer review process; therefore, we enable the publication of all of the content of peer review and author responses alongside final, published articles. The editorial history of this article is available here: <u>https://doi.org/10.1371/journal.pone.0252795</u>

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Data Availability Statement: All data files are available from the OSF database (URL: <u>https://mfr.</u> osf.io/render?url=https%3A%2F%2Fosf.io% 2Fm5xhtb%2Fdown.bad). Abstract

Autism Spectrum Disorders (ASD) are associated to social attention (SA) impairments. A gaze bias to non-social objects over faces has been proposed as an early marker of ASD. This bias may be related to the concomitant circumscribed interests (CI), which question the role of competing objects in this atypical visual behavior. The aim of this study was to com pare visual attention patterns to social and non-social images in young children with ASD and matched typical controls (N = 36; age range 41-73 months) assessing the role of emotion in facial stimuli and the type of competing object. A paired preference task was designed pairing happy, angry, and neutral faces with two types of objects (related or not related to autism CI). Eye tracking data were collected, and three indexes were considered as dependent variables: prioritization (attentional orientation), preference, and duration (sustained attention). Results showed that both groups had similar visual pattern to faces (prioritization, more attention and longer visits to faces paired with objects non-related to their CI); however, the ASD group attended to faces significantly less than controls. Children with ASD showed an emotional bias (late orientation to angry faces and typical preference for happy faces). Finally, objects related to their CI captured attention in both groups, significantly reducing SA in children with ASD. Atypical SA is present in young children with ASD regardless the competing non-social object. Identifying strengths and difficulties in SA in this population may have substantial repercussion for early diagnosis, intervention, and ultimately prognosis.

#### Introduction

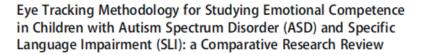
Autism Spectrum Disorders (ASD) comprise a set of neurodevelopmental and pervasive conditions in which social-emotional and communication impairments along with restricted and repetitive behaviors are core symptoms  $[\underline{1}-\underline{4}]$ . The wide heterogeneity of the autism spectrum hampers early diagnosis, which is considered an important predictor of future outcomes and

PLOS ONE https://doi.org/10.1371/journal.pone.0252795 June 4, 2021

1/16

Review Journal of Autism and Developmental Disorders https://doi.org/10.1007/s40489-021-00261-y

**REVIEW PAPER** 



Julia Vacas<sup>1</sup> · Adoración Antolí<sup>1,2</sup> · Araceli Sánchez-Raya<sup>1,2</sup> · Carolina Pérez-Dueñas<sup>1</sup>

Received: 6 March 2020 / Accepted: 22 April 2021

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#### Abstract

Children with autism spectrum disorder (ASD) and specific language impairment (SLI) exhibit serious social-emotional difficulties from early infancy which hamper the differential diagnosis of these two conditions. This review aims at analysing differences between ASD and SLI disorders in children from 0 to 12 years regarding emotional competence abilities using eye tracking methodology. The findings show specific markers in the ASD population (e.g. difficulties in facial emotion recognition, atypical visual scanning patterns, or non-social attentional orientation) and a notable scarcity of studies involving SLI children. Comparative research points at potential differences in visual scanning patterns. The conclusions support the use of this methodology for comparative studies between clinical populations.

Keywords Eye tracking · Emotional competence · Child population · Autism spectrum disorder · Specific language impairment

Neurodevelopmental disorders are defined as a set of conditions that typically occur in early development and which have a serious impact on individuals' personal, social, academic, or occupational functioning (American Psychiatric Association, 2013). Among them, autism spectrum disorder (ASD) and specific language impairment (SLI) have drawn the attention of researchers due to the social-emotional difficulties associated with these conditions.

According to the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association, 2013), ASD is a set of persistent traits that impair social communication and interaction, as well as restricted and repetitive patterns of behaviour and interests. The manual also highlights a significant deficit in social skills such as reciprocity or social interaction understanding, and a wide variability

Published online: 05 May 2021

within this population as core features of ASD. Likewise, the DSM-5 (American Psychiatry Association, 2013) reports an ASD prevalence of around 1% in both children and adults.

Conversely, SLI<sup>1</sup> falls under the scope of communication disorders and refers to a set of persistent difficulties in the acquisition and use of different language components and dimensions (DSM-5; American Psychiatry Association, 2013). Following the DSM-5 criteria, SLI entails early onset; language abilities significantly below average for the age when no auditory, sensory, or motor dysfunction, intellectual disability, or neurological condition is present; and important functional constraints in individuals' daily life.

Although the distinction between ASD and SLI is generally accepted, both disorders show strong similarities, particularly at early ages. These similarities have opened the discussion on a potential shared aetiology (see Taylor et al., 2012, for a review). On the one hand, compromised language abilities differ in each disorder. The pragmatic language function is more reduced in the ASD than in the SLI population, while structural components (e.g. phonology, morphology, or syntax) are more affected in SLI than in ASD. On the other hand,

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<sup>&</sup>lt;sup>2</sup> Early Childhood Care Centre, University of Cordoba, C/San Alberto Magno, s/n, 14071, Cordoba, Spain

<sup>&</sup>lt;sup>1</sup> It is important to note that although the DSM-5 removed the term 'specific language impairment' ('SLI') from the manual and uses the term 'anguage impairment' ('LI') instead, it is still widely used in the literature on the condition.

> Education and Training in Autism and Developmental Disabilities, 2021, 56(3), 306–327 © Division on Autism and Developmental Disabilities

## Emotional Competence in Children with Autism Spectrum Disorders and Specific Language Impairment: A Comparative Research Review

#### Julia Vacas, Adoración Antolí, Araceli Sánchez-Raya, and Carolina Pérez-Dueñas University of Cordoba

Abstract: Emotional competence (EC) refers to a set of skills to identify, understand, and respond to one's own emotions and those of others. It plays a fundamental role in socialization processes, where children with autism speatrum disorders (ASD) and specific language impairment (SLI) show marked deficits. However, due to the similarities between these two conditions, it is difficult to establish a differential diagnosis at early ages. A literature search of the WOS, SCOPUS, and Proquest databases was conducted. Year of publication (from 2000 to 2019), type of paper (reviews, meta-analyses, and experimental studies), language (English), and age of participants (children up to 13 years) were applied as inclusion criteria. Based upon these criteria, 34 papers were included in this review. Results revealed that children with ASD showed impairments in facial emotion recognition, whereas those with SLI showed difficulties in emotion recognition in auditory and audiovisual modalities. Results also highlighted that both children with ASD and SLI showed biases towards the recognition of positive emotions.

Autism spectrum disorders (ASD) and specific language impairment (SLI) have traditionally been regarded as distinct conditions given that the compromised language areas were often well-differentiated: Whereas pragmatic language function was impaired in ASD, structural language functions (e.g. phonology, morphology, or syntax) were compromised in SLI. However, given the significant heterogeneity of both populations, ample evidence has suggested a potential overlap between disorders, either because of the greater functional language disorders in ASD individuals that resemble SLI or the marked social difficulties in SLI individuals that resemble ASD traits (Taylor et al., 2014, 2015). Furthermore, according to the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association [APA], 2013) the high frequency with which ASD and SLI co-occur indicates a

Correspondence concerning this article should be addressed to Julia Vacas, Faculty of Education Sciences, University of Cordoba; C/San Alberto Magno, s/n, 14071, Cordoba, SPAIN. E-mail: 172varuj@uco.es high comorbidity between conditions, which makes it difficult to differentiate them.

There are two main lines to explain the cooccurrence of ASD and SLI. Some evidence supports a shared etiology, while a second theory holds that similarities between disorders are superficial and cannot support the unification of diagnoses (for a review, see Taylor et al., 2012). Given its impact on clinical practice, more research is needed to obtain substantive evidence of the relationship and differences between conditions (Taylor et al., 2012, 2014, 2015).

#### Emotional Competence

Traditionally, studies that involved these clinical populations have focused on their language ability and, to a lesser extent, their cognitive profile, while the social-emotional domain has received less attention. However, this trend seems to be changing considering the increasing number of papers addressing the study of emotion (Jeanneret et al., 2015). According to the bibliometric study of Jeanneret et al. (2015), for example, there has

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Revista Iberoamericana de Diagnóstico y Evaluación - e Avaliação Psicológica ISSN: 1135-3848 ISSN: 2183-6051 bgoncalves@psicologia.ulisboa.pt Associação Iberoamericana de Diagnóstico e Avaliação Psicológica Portugal

Vacas, Julia; Antolí, Adoración; Sánchez-Raya, Araceli; Cuadrado, Fátima Análisis de Perfiles Cognitivos en Población Clínica Infantil con Trastornos del Neurodesarrollo Revista Iberoamericana de Diagnóstico y Evaluación e Avaliação Psicológica, vol. 1, núm. 54, 2020, -Marzo Associação Iberoamericana de Diagnóstico e Avaliação Psicológica Portugal

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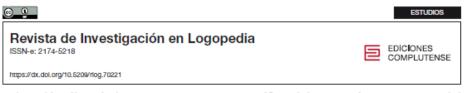


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Migración diagnóstica entre Trastorno Específico del Lenguaje y Trastorno del Espectro Autista: Estudio exploratorio del impacto en los Centros de Atención Infantil Temprana

Julia Vacas, Adoración Antolí, Araceli Sánchez-Raya, Carolina Pérez-Dueñas y Fátima Cuadrado1

Recibido 18 de junio de 2020. Primera revisión 9 de septiembre de 2020. Aceptado 11 de enero de 2021.

Resumen. Trastomo específico del lenguaje (TEL) y trastomo del espectro autista (TEA) son trastomos del neurodesarrollo muy similares a edades tempranas, lo que dificulta el diagnóstico diferencial temprano. El fenómeno 'migración diagnóstica' alude al cambio diagnóstico por evolución del trastorno o modificación de criterios diagnósticos, instrumentos o técnicas de evaluación. El objetivo de este estudio fue analizar la incidencia y el impacto del fenómeno en los centros de atención infantil temprana (CAITs) de la provincia de Córdoba. A través de un cuestionario en formato online habilitado desde marzo a mayo de 2020, los responsables de los CAITs proporcionaron datos cuantitativos sobre este fenómeno y su valoración del impacto en la intervención, el pronóstico y las familias de población infantil de 0 a 6 años. Diez CAITs con financiación pública respondieron al cuestionario. Los resultados arrojaron una alta incidencia del fenómeno migración diagnóstica, principalmente desde TEA hacia TEL. También se observó una diferencia significativa en el número de casos diagnosticados en ambos trastornos, siendo TEA el diagnóstico mayoritario. El impacto de este fenómeno sobre el pronóstico del niño y la familia se estimó positivo, ya que generalmente suponía una evolución hacia un trastorno más leve. Estos resultados ofrecen una aproximación hacia un tema de importancia y escasa investigación hasta la fecha. Continuar esta línea de investigación ampliando la muestra de centros, la información sociodemográfica de los mismos e incluyendo la perspectiva de las familias mejoraría el conocimiento de estos trastornos y favorecería la práctica clínica en el ámbito de la atención temprana. Palabras clave: Trastomo Específico del Lenguaje; Trastomo del Espectro Autista; migración diagnóstica; atención temprana

[en] Diagnostic migration between Specific Language Impairment and Autism Spectrum Disorder: An exploratory study on the impact on Early Intervention Centres

Abstract. Specific language impairment (SLI) and autism spectrum disorder (ASD) are very similar neurodevelopmental disorders mainly at early ages, which hampers early differential diagnosis. Diagnostic migration involves the shift of diagnosis either because of the development of the disorder or because of changes in diagnostic criteria or assessment tools. The aim of this study was to analyse the incidence and impact of this phenomenon in the centres of early intervention (CEIs) in Córdoba. Through an online survey available from March to May 2020, the CEIs managers provided quantitative data on the phenomenon as well as their appraisal of its impact on the intervention, prognosis, and family of children from 0 to 6 years. Ten public CEIs filled the survey. Results yielded a high incidence of diagnostic migration, this phenomenon occurred mainly from ASD to SLI. A significant disproportion of ASD and SLI diagnoses was also highlighted, being ASD the major one. The impact of diagnostic migration on child prognosis and family was deemed positive as it occurred mainly from a severe disorder to a more moderate one. Results from this study provide an initial approach to a relevant and unknown phenomenon. This line of research by increasing the sample deepening on socio-demographic information and including families perspective may improve our knowledge of SLI and ASD, as well as support clinical practice in early intervention. Keywords: Specific Language Impairment; Developmental Language Disorder; Autism Spectrum Disorder; diagnostic migration; early childhood intervention.

Sumario, Introducción, Método, Participantes, Instrumentos, Diseño y procedimiento, Resultados, Discusión, Conclusiones, Bibliografía

Cómo citar: Vacas, J., Antolí, A., Sánchez-Raya, A., Pérez-Dueñas, C., y Cuadrado, F. (2021). Migración diagnóstica entre Trastorno Específico del Lenguaje y Trastorno del Espectro Autista: Estudio exploratorio del impacto en los Centros de Atención Infantil Temprana. *Revista de Investigación en Logopedia, 11* (Núm. Especial), 77-88. https://dx.doi.org/10.5209/ rlog.70221

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Rev. Investig. logop. 11(Núm. Especial) 2021: 77-88

# Other Contributions Resulting from this Doctoral Dissertation

## **Congress Communications**

**Poster presented in XIV Encuentro SPPU (Servicios Psicológicos y Psicopedagógicos Universitarios)** (Córdoba, 8-9 de junio de 2017)

Vacas, J., Antolí, A., & Sánchez-Raya, A. Estudio del Perfil Cognitivo en Población Clínica con Trastornos del Desarrollo.

**Poster presented in 3rd International Congress of Clinical and Health Psychology on Children and Adolescents** (Sevilla, 16-18 de noviembre de 2017)

Vacas, J., Antolí, A., & Sánchez-Raya, A. Analysis of Cognitive Profiles in Clinical Population with Neurodevelopmental Disorders.

Single Oral Communication presented in VI Congreso Científico de Investigadores en Formación de la Universidad de Córdoba (Córdoba, 18-19 de enero de 2018)

Vacas, J., Antolí, A., & Sánchez-Raya, A. Análisis de Perfiles Cognitivos en Poblaciones Clínicas con Trastornos del Neurodesarrollo.

Single Oral Communication presented in 7th International Congress of Clinical and Health Psychology on Children and Adolescents (Online, 18-20 de noviembre de 2021) Vacas, J., Antolí, A., Sánchez-Raya, A., Pérez-Dueñas, C., & Cuadrado, F. Early Attentional Biomarkers in Autism Spectrum Disorders: An Eye-Tracking Study on Social Preference.

Single Oral Communication presented in 7th International Congress of Clinical and Health Psychology on Children and Adolescents (Online, 18-20 de noviembre de 2021) Cuadrado, F., Vacas, J., Antolí, A., Sánchez-Raya, A., & Pérez-Dueñas, C. Migración Diagnóstica entre TEL y TEA en los Centros de Atención Infantil Temprana.

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# Appendixes

# Appendix 1. Análisis de Perfiles Cognitivos en Población Clínica Infantil con Trastornos del Neurodesarrollo

Vacas, J., Antolí, A., Sánchez-Raya, A., & Cuadrado, F. (2020). Análisis de Perfiles Cognitivos en Población Clínica Infantil con Trastornos del Neurodesarrollo. *Revista Iberoamericana de Diagnóstico y Evaluación - e Avaliação Psicológica,* 54(1), 35-46. https://doi.org/10.21865/RIDEP54.1.03

#### Resumen

Dada su común afectación en el lenguaje, este estudio pretende explorar el perfil cognitivo asociado al Trastorno Específico del Aprendizaje, Trastorno Específico del Lenguaje, Síndrome de Asperger, Trastorno por Déficit de Atención e Hiperactividad subtipos Inatento y Combinado formulando criterios de exclusión que orienten el diagnóstico. Se contó con 156 casos clínicos de entre 6 y 15 años (M = 9.5, DT = 2.18) evaluados con WISC-IV, considerando como variables dependientes los índices comprensión verbal, razonamiento perceptivo, memoria de trabajo, velocidad de procesamiento y CI total. El análisis de datos se centró en detectar diferencias intergrupo e intragrupo. Los resultados muestran que el Trastorno Específico del Lenguaje presenta un perfil distintivo respecto a los demás. También se observa que generalmente memoria de trabajo es el índice más afectado, mientras que razonamiento perceptivo se mantiene intacto. Estas conclusiones repercuten en la práctica clínica permitiendo concretar distinciones entre trastornos e individualizar intervenciones.

Palabras clave: Perfil cognitivo, trastornos del neurodesarrollo, WISC-IV, análisis de discrepancias, diagnóstico.

# Abstract

Considering their common affection in certain language dimensions, this study attempts to explore cognitive profiles associated with Specific Learning Disorders, Specific Language Impairment, Asperger Syndrome and Attention-Deficit/Hyperactivity Disorder in two dimensions (Predominantly Inattentive and Combined). The sample consisted of a clinical population of 156 children between 6 and 15 years old (M = 9.5, SD = 2.18) who were applied WISC-IV. The four indexes from this scale (verbal comprehension, perceptual reasoning, working memory, and processing speed) and the full-scale intelligence quotient were considered as dependent variables. Data analysis was focused on the assessment of intergroup and intragroup differences. Results prove a highly distinctive profile for Specific Language Impairment compared to the other disorders and a general pattern of strengths and weaknesses among them. Conclusions from this study contribute to the improvement of clinical practice as our data highlight significant differences between neurodevelopmental disorders which may drive to more individualized interventions.

**Keywords:** Cognitive profile, neurodevelopmental disorders, WISC-IV, pairwise discrepancies analysis, diagnostic.

# 1.1Introducción

# 1.1.1 Trastornos del Neurodesarrollo

La quinta edición del Manual Diagnóstico y Estadístico de los Trastornos Mentales (DSM-5; American Psychiatric Association [APA], 2013) define los Trastornos del Neurodesarrollo (TND) como un conjunto de afecciones que aparecen precozmente en el curso del desarrollo infantil interfiriendo en el mismo y causando déficits en la manera en que el niño se desenvuelve en los ámbitos personal, social, académico u ocupacional. El propio manual insiste en que una de las características que mejor define los TND es la variabilidad tanto en las áreas afectadas como en el grado de afectación de las mismas, dando como resultado un amplio abanico de combinaciones y posibilidades que el profesional clínico debe manejar correctamente para ofrecer un diagnóstico acertado. Así, se puede afirmar que generalmente los niños y niñas no presentan un perfil clínico claro, es decir, no muestran todas las características asociadas a un trastorno concreto, sino que pueden manifestar rasgos de distintas patologías. Todo ello incrementa la dificultad de realizar un diagnóstico adecuado, por ello el propio DSM-5 recomienda revisarlo periódicamente para conocer su trayectoria por si pudiera haber evolucionado a un perfil clínico distinto. Dadas las dificultades mencionadas, resulta imprescindible tanto conocer las características propias de los diferentes trastornos como establecer comparaciones entre aquellos que presenten rasgos muy similares. Para evitar confusiones y diagnósticos erróneos, desde la investigación se deben proporcionar criterios diferenciales que guíen el diagnóstico y faciliten la toma de decisiones.

## 1.1.2 WISC-IV como Herramienta para la Evaluación de los TND

Este trabajo no está orientado exclusivamente al proceso diagnóstico, sino que también pretende generar reflexión sobre el papel que juegan los datos obtenidos en las pruebas psicométricas para la fundamentación de las intervenciones terapéuticas. Es bien sabido que en la intervención con personas con TND se deben tener en consideración tanto las fortalezas como las debilidades cognitivas del sujeto de manera que se favorezca el aprovechamiento de las primeras para reforzar y complementar las segundas. En este sentido, las nuevas técnicas de neuroimagen y los modelos computacionales han demostrado que las áreas del cerebro están en permanente conexión y evolución, de manera que los procesos de desarrollo y maduración cognitiva poseen un carácter dinámico e interactivo en el cual las diferentes áreas compiten y cooperan entre ellas (Wass, 2015).

Otro elemento fundamental de este estudio ha sido el empleo de la Escala de Inteligencia para Niños de Weschler versión 4 (WISC-IV; Weschler, 2003a). Esta batería cognitiva destaca como la principal herramienta clínica empleada para la detección y el diagnóstico de trastornos en la infancia según la opinión y la experiencia de los profesionales tanto a nivel europeo (Evers et al., 2012) como concretamente en España (Muñiz & Fernández-Hermida, 2010). Igualmente, es la escala más utilizada como instrumento de evaluación neuropsicológica en Estados Unidos y Canadá (Rabin et al., 2016). Según estos autores, el WISC-IV ocupa la quinta posición en la categoría general de "instrumentos de evaluación usados con mayor frecuencia", concretamente destaca en la evaluación específica de habilidades como atención, concentración, memoria de trabajo, inteligencia general, logro, habilidades visoespaciales, e incluso el estado mental y la cognición global. De este modo, se comprueba que a pesar de la actual controversia

sobre la adecuación del WISC-IV como herramienta de evaluación y diagnóstico, su utilización en la práctica clínica está ampliamente extendida.

#### 1.1.3 Estudios Previos sobre Perfiles Cognitivos en TND

En el presente estudio se han seleccionado un conjunto de trastornos que tienen en común alteraciones en determinadas áreas del lenguaje; si bien las habilidades lingüísticas alteradas son distintas en cada trastorno, en muchos casos la afectación puede manifestarse de formas similares, repercutiendo considerablemente en la concreción de su diagnóstico. Los trastornos seleccionados han sido: Trastorno Específico del Aprendizaje, concretamente Dificultades en el Aprendizaje de la Lectura o dislexia (DAL), Trastorno Específico del Lenguaje (TEL), Síndrome de Asperger (SA), Trastorno por Déficit de Atención e Hiperactividad predominantemente Inatento (TDAH-I) y su variante de tipo Combinado (TDAH-C). Otro motivo que justifica la selección de trastornos es la cantidad de estudios que resaltan una alta comorbilidad entre algunos de ellos, como es el caso de TEL-TDAH (Gómez et al., 2016; Miranda-Casas et al., 2011) y TEL-DAL (Schuchardt et al., 2013).

Hasta la fecha, múltiples trabajos han tratado de esclarecer los perfiles cognitivos asociados a las patologías mencionadas. Sin embargo, ninguno de ellos ha abarcado tal variedad de trastornos. No obstante, las conclusiones derivadas de estos estudios previos suponen el punto de partida de esta investigación. Así, se puede definir a la población con DAL por sus déficits en memoria de trabajo y velocidad de procesamiento (Berninger & O' Donell, 2005; Cornoldi et al., 2014; Giofrè & Cornoldi, 2015). Por otra parte, también se ha hallado una alta comorbilidad entre DAL y TEL matizando que ambas patologías presentan afectación en memoria de trabajo, aunque dentro de esta función cognitiva, son distintas las áreas que aparecen afectadas en uno y otro trastorno (Schuchardt et al., 2013). Por ello, el estudio en profundidad de este constructo podría ayudar a discernir entre una patología y otra.

Por otro lado, las dificultades en la memoria de trabajo son consideradas como un marcador fenotípico fiable para predecir la presencia de TEL (Conti-Ramsden, 2003; Lum et al., 2015; Schuchardt et al., 2013; Spanoudis & Natsopoulos, 2011). Sin embargo, algunos estudios ponen en duda esta afirmación al señalar que existe un porcentaje considerable de niños y niñas con TEL (entre un 20 y un 25%) que no presentan afectación en este índice (Alloway et al., 2009) o encontrar la memoria de trabajo intacta en una

Appendix 1. Análisis de Perfiles Cognitivos en Población Clínica Infantil con Trastornos del Neurodesarrollo

muestra de prescolares con TEL que presentaban alteración en la velocidad de procesamiento (Reichenbach et al., 2016). Por otro lado, respecto a comprensión verbal, numerosos trabajos evidencian que existen importantes déficits en este grupo en cuanto a habilidades lingüísticas fundamentales como la conciencia metalingüística y la comprensión de verbos mentales (Spanoudis & Natsopoulos, 2011) o la adquisición de estructuras gramaticales, morfológicas y sintácticas complejas (Castro-Rebolledo et al., 2004; van Weerdenburg et al., 2006). Todo ello permite hipotetizar que el constructo comprensión verbal estará notablemente afectado, aunque ningún estudio hasta el momento ha obtenido resultados claramente significativos empleando la escala WISC-IV.

Por su parte, el perfil cognitivo de la población con SA destaca por ser ampliamente heterogéneo, con picos que sobresalen la media y habilidades cognitivas significativamente por debajo de la norma (González-Gadea et al., 2014). En general, la investigación coincide en resaltar los déficits de estos sujetos en memoria de trabajo y velocidad de procesamiento (Oliveras-Rentas et al., 2012; Planche & Lemonnier, 2012), así como un patrón de comprensión verbal notablemente superior al razonamiento perceptivo que permite diferenciar a esta población de otros tipos de trastornos del espectro autista (Nedelcu & Buceta, 2012).

Finalmente, la población con TDAH muestra también una clara afectación en los índices memoria de trabajo y velocidad de procesamiento; sin embargo, no existe consenso acerca de su alcance. Algunos estudios defienden que dicha afectación está presente en ambos índices y subtipos: inatento y combinado (Bustillo & Servera, 2015) e incluso encuentran un patrón diferenciado entre ambos ya que el TDAH-I presentaría mayores déficits en memoria de trabajo que en velocidad de procesamiento, mientras que en TDAH-C la tendencia es a la inversa (Fenollar-Cortés et al., 2015); otros solo encuentran dificultades en memoria de trabajo sin distinción entre un subtipo y otro (García et al., 2013; Miranda-Casas et al., 2011); y un tercer grupo sugiere que solo el TDAH-I muestra bajo rendimiento en los índices mencionados (Capdevila-Brophy et al., 2006).

A la luz de la información expuesta, se comprueba que los estudios citados muestran resultados y conclusiones muy diferentes en cuanto al perfil cognitivo de estas poblaciones; de ahí que se mantenga la necesidad de seguir investigando en esta temática

con objeto de obtener datos más concluyentes. En este sentido, el objetivo general de este trabajo es ampliar y mejorar las posibilidades del WISC-IV como herramienta complementaria para orientar el diagnóstico y que sirva como punto de partida para al diseño de la intervención específica con niños y niñas con TND. Para lograrlo, se ha evaluado y comparado los perfiles cognitivos concretos de los TND seleccionados, estableciendo diferencias intergrupo que faciliten el diagnóstico diferencial. Paralelamente, con el objetivo de establecer diferencias intragrupo, se han analizado las discrepancias entre pares de índices. En otras palabras, los dos objetivos específicos del estudio se centran en el análisis intergrupo e intragrupo de los perfiles cognitivos de las poblaciones seleccionadas como medio para expandir la aplicabilidad del WISC-IV durante el proceso diagnóstico. La hipótesis de partida apunta a que, a nivel intergrupo, los índices memoria de trabajo y velocidad de procesamiento serán los más vulnerables en todos los trastornos evaluados, en el caso de la población con TEL podría encontrarse también afectación en comprensión verbal (H1). Igualmente, se espera encontrar diferencias entre trastornos según el grado de afectación de tales índices, siendo dicha afectación mayor en los sujetos con TEL, SA y TDAH y menor en aquellos con DAL (H2). A nivel intragrupo, la hipótesis principal sugiere que SA mostrará un perfil más heterogéneo, diferenciándose así del resto de trastornos (H3).

## 1.2 Método

## 1.2.1 Participantes

A partir del software GPower y estableciendo un tamaño del efecto de .06, un error  $\alpha$  de .05 y un error  $\beta$  de .95 se determinó que la muestra total debería incluir un mínimo de 130 participantes para poder realizar los análisis adecuados, de manera que se recogieron datos de 156 casos evaluados en la Unidad de Salud Mental Infantojuvenil (USMIJ) del Hospital Reina Sofía de Córdoba (Tabla A1.1). La principal variable definitoria del grupo muestral fue el diagnóstico, seguido por la edad. Como se puede observar en la Tabla A1.1, los sujetos examinados oscilan entre 6 y 15 años (M = 9.5, DT = 2.18, ET = .18). Por otro lado, respecto al sexo, la muestra concuerda con las conclusiones derivadas de la investigación epidemiológica en este tipo de trastornos que resalta una mayor afectación entre la población masculina, en torno a 2.8:8 a favor de las mujeres (Castro-Rebodello et al., 2004).

Variable		$\mathbf{DAL}$ $(n = 25)$		TEL (n = 25)		<b>SA</b> $(n = 35)$		<b>TDAH-I</b> $(n = 35)$		<b>TDAH-C</b> $(n = 36)$		<b>Total</b> $(N = 156)$	
		f	%	f	%	f	%	f	%	f	%	f	%
Sexo	Niño	19	12.2%	22	14.1%	33	21.2%	25	16%	27	17.3%	126	80.8%
	Niña	6	3.8%	3	1.9%	2	1.3%	10	6.4%	9	5.8%	30	19.2%
	Total	25	16%	25	16%	35	22.4%	35	22.4%	36	23.1%	156	100%
Edad	6-7	7	4.5%	10	6.4%	5	3.2%	5	3.2%	6	3.8%	33	21.2%
	8-9	10	6.4%	3	1.9%	18	11.5%	12	7.7%	11	7.7%	55	35.2%
	10-11	6	3.8%	9	5.8%	4	2.6%	8	5.1%	9	5.8%	36	23.1%
	12-13	2	1.3%	3	1.9%	5	3.2%	8	5.1%	6	3.9%	24	15.4%
	14-15	0	0%	0	0%	3	1.9%	2	1.3%	3	1.9%	8	5.1%
	Total	25	16%	25	16%	35	22.4%	35	22.4%	36	23.1%	156	100%

 Table A1.1. Características de la muestra

## 1.2.2 Instrumento

El WISC-IV (Wechsler, 2003a) es el resultado de la revisión y actualización de una larga tradición de pruebas de inteligencia diseñadas por David Wechsler que han tenido como objetivo superar el mero uso de las escalas como tests psicométricos (Flanagan & Kaufman, 2006). Recoge información sobre cuatro aspectos básicos de la inteligencia: comprensión verbal (CV), razonamiento perceptivo (RP), memoria de trabajo (MT) y velocidad de procesamiento (VP) a partir de quince tests (diez principales y cinco optativos). Está dirigido a población con edades comprendidas entre 6 y 16 años.

Existen numerosos estudios que han puesto a prueba las propiedades psicométricas de esta escala, entre ellas su tipificación, fiabilidad, saturación en g y validez estructural (Flanagan & Kaufman, 2006). Del mismo modo, esta batería de tests fue sometida a un importante procedimiento de tipificación que contó con 2200 sujetos en la versión estadounidense y 1590 en la española (Corral et al., 2005). En ambos casos se ofrecen resultados basados en grupos especiales que resultan de gran interés para obtener un marco de referencia con el que comparar puntuaciones.

La información obtenida en esta escala es muy variada, ya que ofrece datos que abarcan desde el CI total hasta medidas resultantes de la combinación de índices como el ICG (Índice de Capacidad General) derivado de la suma de las puntuaciones escalares de las subpruebas de los índices CV y RP y su correspondencia con los baremos del manual de interpretación de la escala, o el ICC (Índice de Competencia Cognitiva) obtenido a partir de la suma de las puntuaciones escalares de las subpruebas de los índices MT y VP y su correspondencia con los baremos del manual de interpretación de las puntuaciones escalares de las subpruebas de los índices MT y VP y su correspondencia con los baremos del manual de interpretación de la escala. Estos

índices han sido ampliamente utilizados en la investigación de perfiles clínicos, por lo que en el presente estudio se ha ampliado este análisis de discrepancias abarcando la comparación de pares de índices (véase en procedimiento y análisis estadístico).

### 1.2.3 Procedimiento y Análisis Estadístico

La selección de casos se realizó en función del diagnóstico entre la población clínica infantil atendida y evaluada por los profesionales especialistas acreditados en psicología clínica sanitaria de la USMIJ del Hospital Reina Sofía de Córdoba siguiendo el protocolo establecido en la unidad para tal efecto. Durante el proceso de recogida de datos, se garantizó que toda la información fuera tratada de forma anónima y confidencial de acuerdo con el Real Decreto-ley 5/2018, de 27 de julio, de medidas urgentes para la adaptación del Derecho español a la normativa de la Unión Europea en materia de protección de datos y siguiéndose de forma rigurosa los principios éticos promulgados en la declaración de Helsinki por la Asociación Médica Mundial. Igualmente, se obtuvo la aprobación y el consentimiento del Comité de Ética de la Investigación de Córdoba para el desarrollo del proyecto.

Tras recoger las puntuaciones de los sujetos seleccionados en las diferentes pruebas e índices de la batería cognitiva, estas fueron tratadas con el Statistical Package for the Social Sciences, 21 (SPSS, 21) con el que se llevó a cabo un análisis intergrupo. Así, se realizó un análisis multivariante de la varianza (MANOVA) introduciendo como factor fijo el diagnóstico y como variables dependientes los cuatro índices del WISC-IV (CV, RP, MT y VP) y el CIT para obtener un perfil cognitivo concreto y bien diferenciado de cada patología. Igualmente, para aislar cualquier posible efecto del sexo sobre los resultados, éste fue incorporado al análisis como covariable.

Por otro lado, también se llevó a cabo un análisis intragrupo de estos perfiles por medio de la mencionada comparación de discrepancias entre pares de índices. Dichas discrepancias entre los índices marcan la heterogeneidad del perfil y son una señal que indica la posibilidad de que exista un trastorno o el grado de afectación del mismo. Así, se pusieron en relación los distintos índices y se observaron las diferencias entre ellos a fin de comprobar si existe un fuerte contraste entre el rendimiento del sujeto en una habilidad cognitiva específica sobre las demás. Las parejas de índices contrastadas fueron: CV-RP, CV-MT, CV-VP, RP-MT, RP-VP, MT-VP.

Las comparaciones entre las puntuaciones individuales de un sujeto en cualquier prueba psicométrica y el contraste del perfil de dicho sujeto con las puntuaciones normativas son procedimientos fundamentales en la práctica neuropsicológica ya que ayudan a detectar y cuantificar la dimensión del déficit (Crawford et al., 2007). Existen diversos protocolos para el tratamiento de las comparaciones entre índices y el establecimiento de diferencias anormales. En este caso, se ha escogido el enfoque empírico del manual ya que la muestra de estandarización del WISC-IV es muy amplia y el propio proceso de estandarización ofrece una distribución normal multivariada (Crawford et al., 2010).

El procedimiento basado en el enfoque empírico consiste en hallar la diferencia entre las puntuaciones compuestas de las parejas de índices y comparar el valor resultante con los valores críticos aportados en los apéndices del manual (Tabla A1.2). En este proceso, se tiene en cuenta la edad de los sujetos, el valor positivo o negativo de la diferencia hallada (éste marca la dirección a favor de un índice u otro) y el nivel de confianza (85% o 95%) que se emplea para la detección de discrepancias significativas. Todo ello con objeto de dar un paso más en la concreción de los perfiles cognitivos afinando las diferencias intragrupo.

## **1.3 Resultados**

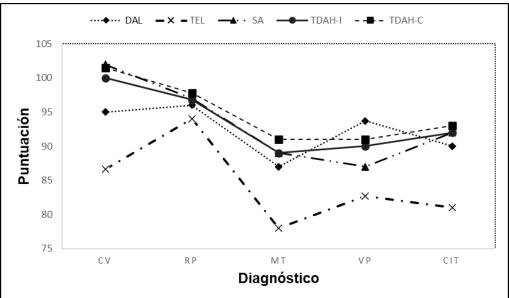
# 1.3.1Análisis Intergrupo

La Figura A1.1 permite apreciar de una manera gráfica los resultados obtenidos en cada perfil. Así, se observa que los sujetos con TEL muestran un perfil cognitivo significativamente más bajo que el resto de los trastornos estudiados cuyas puntuaciones medias son bastante similares en índices como RP, MT y CIT. Los índices CV y VP muestran más variabilidad entre trastornos, aunque manteniéndose dentro de los límites que definen a la población normativa (M = 100,  $DT = \pm 15$ ); sin embargo, en el caso de TEL, esta población se encuentra por debajo de estos límites en todos los índices medidos excepto en CV y RP.

Analizando más detenidamente la Figura A1.1, se puede ver que: en primer lugar, CV es el índice más destacado (por encima de 100) en SA y en ambos subtipos de TDAH. Por otra parte, RP es el punto fuerte en sujetos con TEL y DAL. Por el contrario, MT es el índice que muestra puntuaciones más bajas en todos los trastornos estudiados,

exceptuando SA; en el caso de los sujetos con TEL, este índice se configura claramente como un punto débil, ya que se aleja de la media en más de 20 puntos. Por otro lado, los resultados de la VP muestran que este es el índice con menor rendimiento entre la población con SA. Finalmente, el CIT de los sujetos con DAL, SA, TDAH-I y TDAH-C es bastante similar y se mueve dentro de los parámetros considerados normales, mientras que los sujetos con TEL presentan un CIT por debajo de la norma. De esta forma, la H1 se ve parcialmente confirmada ya que MT y VP son los índices que presentan puntuaciones más bajas en general, aunque solo en el caso de TEL las puntuaciones son significativamente bajas respecto a la población normativa; no obstante, el grupo con DAL mantiene intacto el índice VP, al igual que ocurre con el grupo con TEL en CV.

**Figure A1.1.** *Diferencias Intergrupo atendiendo al Perfil Cognitivo en puntuaciones en las subescalas del WISC-IV* 



En la Tabla A1.2 se muestran los resultados del MANOVA realizado sobre las puntuaciones de los índices del WISC-IV. La prueba de Levene demostró que las varianzas de los grupos en los diferentes índices eran similares arrojando los siguientes valores de significación: CV = .135, RP = .240, MT = .380, VP = .071 y CIT = .640. Por otra parte, el MANOVA reveló diferencias significativas en los índices según el diagnóstico ( $F_{(146,4)} = 2.22$ , p = .002,  $\eta^2 = .069$ ). Como se puede observar en la Tabla A1.2, estas diferencias se concretan en todos los índices excepto en RP. La prueba *post-hoc* de Bonferroni desveló que las diferencias en CV se dieron entre TEL y SA (p = .000), TEL y TDAH-I (p = .001) y TEL y TDAH-C (p = .000); en MT se observaron diferencias entre TEL y SA (p = .011), TEL y TDAH-I (p = .020) y TEL y TDAH-C (p = .003); en VP se

apreciaron diferencias entre TEL y DAL (p = .036); y en CIT se evidenciaron diferencias entre TEL y DAL (p = .050), TEL y SA (p = .001), TEL y TDAH-I (p = .006), TEL y TDAH-C (p = .001). Estos datos corroboran parcialmente la H2 ya que, aunque el MANOVA probó la existencia de diferencias entre trastornos con un tamaño del efecto grande, la prueba Bonferroni mostró que tales diferencias solo permitían distinguir el perfil TEL del resto de trastornos.

			Diagnóstic	0				
	DAL	TEL	SA	TDAH- I	TDAH-C			
	М	М	М	М	M	-		
Variable	(DT, ET)	(DT, ET)	(DT, ET)	(DT, ET)	(DT, ET)	$F_{(4,146)}$	р	$\eta^2$
	95.20	86.60	102.57	100.29	101.44 (13.55,	6.92	.000	.16
CV	(13.33,	(11.59,	(10.80,	(16.07,	2.22)			
	2.66)	2.66)	2.27)	2.26)				
	96.44	94.08	97.14	96.89	97.86	.32	.867	.01
RP	(15.40,	(15.03,	(12.22,	(10.50,	(12.55, 2.17)			
	2.61)	2.61)	2.23)	2.21)				
	87.32	78.08	89.06	89.37	91.06	4.12	.003	.10
MT	(15.42,	(13.54,	(12.96,	(10.96,	(12.08, 2.14)			
	2.57)	2.57)	2.20)	2.18)				
	93.76	82.72	87.09	89.97	91.08	2.44	.05	.06
VP	(13.00,	(11.68,	(14.75,	(9.80,	(12.12, 2.02)			
	2.42)	2.43)	2.07)	2.06)				
	90.32	81.20	92.37	91.77	93.19	5.26	.001	.12
CIT	(12.98,	(10.56,	(10.72,	(9.80,	(10.92, 1.83)			
	2.19)	2.19)	1.87)	1.86)				

Table A1.2. Análisis de Diferencias Intergrupo. MANOVA para Trastornos e Índices

*Nota:* La variable sexo fue tenida en cuenta como covariable

## 1.3.2 Análisis Intragrupo

Por su parte, en la Tabla A1.3 aparecen los resultados obtenidos a partir del proceso de comparación entre pares de índices. Así, se presenta la media de la diferencia obtenida por cada grupo diagnóstico en cada pareja de índices junto con su desviación típica, el error típico y la significatividad. De esta forma, se observa que en un nivel de confianza del 85% no existen diferencias significativas entre las parejas de índices en los sujetos con DAL. Por otro lado, en los sujetos con TEL sí aparece significatividad entre los pares de índices RP-MT y RP-VP a favor de RP en ambos casos. Del mismo modo, los sujetos con SA presentan discrepancias significativas entre los pares CV-MT y CV-VP, en todos los casos mostrando una diferencia positiva en favor del primer índice de la comparación. Por su parte, los sujetos con TDAH-I solo manifiestan diferencias significativas entre CV-MT en favor del primero, al igual que ocurre con el grupo con TDAH-C. Atendiendo al nivel de confianza del 95%, se observa que la significatividad

varía considerablemente, reduciéndose a los pares RP-MT en el caso de los sujetos con TEL, así como CV-MT y CV-VP en los sujetos con SA.

	Diagnosico									
Índices	DAL	TEL	SA	TDAH-I	TDAH-C	Total				
	M (DT, ET)	M (DT, ET)	M (DT, ET)	M (DT, ET)	M (DT, ET)	М (DT, ET)				
CV-RP	-1.24	-7.48	5.43	3.40	3.58	1.41				
	(15.13, 3.03)	(16.17, 3.23)	(12.94, 2.19)	(18.27, 3.08)	(15.88, 2.65)	(16.20, 1.30)				
CV-MT	7.88	8.52	13.51	10.91	10.39	10.51				
	(13.70, 2.74)	(13.58, 2.72)	(17.77, 3.00)***	(17.08, 2.89)*	(16.14, 2.69)*	(15.92, 1.28)				
CV-VP	1.44	3.88	15.49	10.31	10.36	9.03				
			(18.66, 3.15)***			(18.47, 1.48)				
RP-MT	9.12	16.00	8.09	7.51	6.81	9.10				
<b>KI</b> -1 <b>11</b>	(18.68, 3.74)					(16.69, 1.34)				
MT-VP	-6.44	-4.64	1.97	60	03	-1.47				
			(18.78, 3.18)			(16.16, 1.29)				
RP-VP	2.68 (20.65, 4.13)	11.36 (14.48, 2.90)*	10.06 (15.69, 2.65)	6.91 (11.67, 1.97)	6.78 (15.78, 2.63)	7.62 (15.70, 1.26)				

**Table A1.3.** Análisis de Discrepancias Intragrupo-Comparación entre Pares de Índices

Diagnóstico

*Nota:* Puntuaciones Medias de las Discrepancias entre Pares de Índices y sus Niveles de Significatividad en relación a los Valores Críticos establecidos en Wechsler (2003b) \*p < .15, \*\*\*p < .05

#### 1.4 Discusión y Conclusiones

El principal objetivo de este estudio era indagar las posibilidades del WISC-IV como herramienta diagnóstica y de diseño de intervenciones adaptadas a niños y niñas con distintos tipos de TND a partir del análisis de la variabilidad intergrupo e intragrupo de sus perfiles. Para ello, en primer lugar, se han evaluado los perfiles cognitivos concretos de los TND seleccionados, estableciendo diferencias intergrupo que permitieran afinar su diagnóstico. A este respecto, los resultados obtenidos con la variedad de trastornos que se han incluido en el estudio aportan claves que complementan el conocimiento que ya existe sobre esta temática. Así, se ha comprobado que existen importantes diferencias entre el perfil de niños y niñas con TEL y el resto de los trastornos estudiados (principalmente SA, TDAH-I y TDAH-C). Igualmente, se han hallado

diferencias significativas entre TEL y DAL en VP lo cual complementa las conclusiones de Schuchardt et al. (2013) añadiendo que el estudio de la VP puede ayudar a discernir entre ambos trastornos. Sin embargo, no se han encontrado grandes diferencias entre los demás diagnósticos, ya que las puntuaciones en los índices son bastante similares y se mantienen dentro de la norma. Igualmente, cabe señalar que los grupos con TDAH-I y TDAH-C de esta muestra han presentado perfiles idénticos con la misma tendencia en todos los índices, con lo cual en este estudio no se confirma la afectación de estos sujetos en ningún índice del WISC-IV (Bustillo & Servera, 2015; Fenollar-Cortés et al., 2015) ni la presencia de diferencias entre perfiles (Capdevila-Brophy et al., 2006; Fenollar-Cortés et al., 2015). Asimismo, resulta llamativa la falta de evidencia de déficits en el grupo con TEL en el índice CV, cuya existencia ha sido sobradamente probada en estudios previos (Spanoudis & Natsopoulos, 2011; Taylor et al., 2012; van Weerdenburg et al., 2006). En este estudio la media en este índice se encuentra en el límite de la normalidad, aunque muestra diferencias significativas con el resto de los trastornos, a excepción de DAL. Por ello, en línea con la propuesta de Miller & Gilbert (2008), se recomienda el uso de diferentes escalas para obtener un perfil más completo y contrastado. Concretamente en el caso de niños y niñas con TEL, se aconseja el empleo de escalas de lenguaje complementarias.

Por otro lado, este trabajo revela que el análisis de discrepancias entre pares de índices resulta una interesante fuente de datos que permite ahondar en las diferencias intragrupo y, de este modo, conocer no solo los puntos fuertes y débiles asociados a cada trastorno, sino también, qué índices muestran más disparidad, lo cual proporciona una valiosa referencia con vistas a la intervención. Las repercusiones de estos resultados en la práctica clínica parecen evidentes; si bien la literatura científica destacaba una afectación generalizada en los índices MT y VP entre los trastornos que han formado parte del estudio, a partir del análisis de discrepancias se ha comprobado que dicha afectación resulta aún más significativa en comparación al notable rendimiento en CV y RP en prácticamente todos los casos. De este modo, se ha observado que las discrepancias más significativas en un intervalo del 95% de confianza se han obtenido en la población con TEL y SA entre sus índices de mejor rendimiento (RP en el primer caso y CV en el segundo) y aquellos en los que presentan más dificultades (MT en el primer caso, y MT y VP en el segundo). Esta tendencia se mantiene si se atiende al intervalo de confianza del 85%, observando cómo se amplía el número de discrepancias significativas, aunque

siempre en concordancia con los puntos fuertes y débiles de cada trastorno. Igualmente, en línea con las conclusiones de Nedelcu & Buceta (2012) esta muestra de SA presenta un patrón de CV superior a RP, aunque el análisis de discrepancias no arroja significatividad estadística en esta diferencia.

Por su parte, el único trastorno que no muestra discrepancias significativas al comparar sus pares de índices es DAL, esto unido al hecho de que sus puntuaciones en cada uno de los índices analizados de manera aislada permanecen dentro de la media normativa, permite inferir que de todos los trastornos estudiados DAL es el que presenta un perfil más homogéneo y con menor afectación, lo cual coincide con las conclusiones de Berninger y O' Donell (2005). A continuación, se encuentran ambos subtipos de TDAH, que muestran disparidad entre algunos de sus índices en un intervalo de confianza del 85%; sin embargo, dicha disparidad desaparece al 95% de confianza y sus puntuaciones se mantienen dentro de la norma en todos los índices evaluados. Finalmente, los perfiles intragrupo más heterogéneos se observan entre las poblaciones con TEL y SA, dado que, tanto en la comparación entre pares de índices como en el análisis individual de los mismos, han obtenido puntuaciones más variables. Estos datos en conjunto permiten confirmar la H3 que sugería que la población con DAL tendría un perfil más homogéneo en tanto que la que presenta SA mostraría la mayor heterogeneidad; si bien no se esperaba encontrar tal heterogeneidad entre la población con TEL, estas afirmaciones concuerdan con las arrojadas por Alloway et al. (2009) y Lum et al. (2015) en el caso de TEL, así como González-Gadea et al. (2014) en el caso de SA.

Para concluir, es importante recalcar la necesidad de ser prudentes a la hora de interpretar y utilizar los resultados derivados de las escalas psicométricas ya que, si bien proporcionan una información muy valiosa tanto a nivel intersujeto como intrasujeto, debido a la multiplicidad de comparaciones realizadas se incrementan los niveles de error tipo 1. Además, es preciso tener en cuenta que existen diversos enfoques para comparar los resultados obtenidos con las tasas base y, de este modo, establecer la estimación de la anormalidad en base a la distribución de las diferencias en la muestra estandarizada (Crawford et al., 2007, 2009, 2010). Por todo ello, se recomienda utilizar este tipo de datos con cautela y principalmente como herramientas que faciliten el proceso diagnóstico y orienten las intervenciones.

Finalmente, cabe destacar como limitaciones del estudio el tamaño de la muestra que, debido a su carácter clínico, es más reducido que en otro tipo de trabajos. Por otra parte, sería interesante continuar esta línea de investigación complementando la escala utilizada con otras baterías que midan con mayor precisión aspectos cognitivos específicos tales como el lenguaje. De este modo, se podría profundizar sobre las diferencias encontradas. Igualmente, el análisis de dos índices alternativos creados por Labin et al. (2018) para evaluar las habilidades fluidas verbales y no verbales a partir del WISC-IV podría emplearse para llevar a cabo un estudio más exhaustivo de estos trastornos. Por último, la introducción, aunque todavía con un uso clínico poco extendido, de la nueva versión de las escalas de Wechsler (WISC-V), ofrece nuevas posibilidades para continuar esta línea de trabajo a fin de conocer si los cambios introducidos en esta versión (principalmente la reconceptualización de algunos índices, incluyendo el visoespacial -VS- y razonamiento fluido -RF-) podrían alterar o complementar el conocimiento que se tiene hasta la fecha sobre los perfiles cognitivos de estas poblaciones. Los citados índices derivan de los subtests que componen RP en el WISC-IV (Weiss et al., 2016) y dado que éste ha sido el índice que presenta más homogeneidad tanto a nivel intergrupo como intragrupo en este estudio, la replicación de los datos presentados a partir del uso de los índices alternativos del WISC-IV así como del WISC-V podría arrojar nuevas evidencias.

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# Appendix 2. Migración diagnóstica entre Trastorno Específico del Lenguaje y Trastorno del Espectro Autista: Estudio exploratorio del impacto en los Centros de Atención Infantil Temprana

Vacas, J., Antolí, A., Sánchez-Raya, A., Pérez-Dueñas, C., & Cuadrado, F. (2021). Migración diagnóstica entre Trastorno Específico del Lenguaje y Trastorno del Espectro Autista: Estudio exploratorio del impacto en los Centros de Atención Infantil Temprana. *Revista de Investigación en Logopedia, 11*(Special Issue), 77-88. https://dx.doi.org/10.5209/rlog.70221

#### Resumen

Trastorno específico del lenguaje (TEL) y trastorno del espectro autista (TEA) son trastornos del neurodesarrollo muy similares a edades tempranas, lo que dificulta el diagnóstico diferencial temprano. El fenómeno 'migración diagnóstica' alude al cambio diagnóstico por evolución del trastorno o modificación de criterios diagnósticos, instrumentos o técnicas de evaluación. El objetivo de este estudio fue analizar la incidencia y el impacto del fenómeno en los centros de atención infantil temprana (CAITs) de la provincia de Córdoba. A través de un cuestionario en formato online habilitado desde marzo hasta mayo de 2020, los responsables de los CAITs proporcionaron datos cuantitativos sobre este fenómeno y su valoración del impacto en la intervención, el pronóstico y las familias de población infantil de 0 a 6 años. Diez CAITs con financiación pública respondieron al cuestionario. Los resultados arrojaron una alta incidencia del fenómeno migración diagnóstica, principalmente desde TEA hacia TEL. También se observó una diferencia significativa en el número de casos diagnosticados en ambos trastornos, siendo TEA el diagnóstico mayoritario. El impacto de este fenómeno sobre el pronóstico del menor y la familia se estimó positivo, ya que generalmente suponía una evolución hacia un trastorno más leve. Estos resultados ofrecen una aproximación hacia un tema de importancia y escasa investigación hasta la fecha. Continuar esta línea de investigación ampliando la muestra de centros, la información

sociodemográfica de los mismos e incluyendo la perspectiva de las familias mejoraría el conocimiento de estos trastornos y favorecería la práctica clínica en el ámbito de la atención temprana.

**Palabras clave:** Trastorno específico del lenguaje, trastorno del espectro autista, migración diagnóstica, atención temprana.

### Abstract

Specific language impairment (SLI) and autism spectrum disorders (ASD) are very similar neurodevelopmental disorders mainly at early ages, which hampers early differential diagnosis. Diagnostic migration involves the shift of diagnosis either because of the development of the disorder or because of changes in diagnostic criteria or assessment tools. The aim of this study was to analyze the incidence and impact of this phenomenon in the centers of early intervention (CEIs) in Córdoba. Through an online survey available from March to May 2020, the CEIs managers provided quantitative data on the phenomenon as well as their appraisal of its impact on the intervention, prognosis, and family of children from 0 to 6 years. Ten public CEIs filled the survey. Results yielded a high incidence of diagnostic migration, this phenomenon occurred mainly from ASD to SLI. A significant disproportion of ASD and SLI diagnoses was also highlighted, being ASD the major one. The impact of diagnostic migration on child prognosis and family was deemed positive as it occurred mainly from a severe disorder to a more moderate one. Results from this study provide an initial approach to a relevant and unknown phenomenon. Continuing this line of research by increasing the sample, deepening on socio-demographic information, and including families perspective may improve our knowledge of SLI and ASD, as well as support clinical practice in early intervention.

**Keywords:** Specific Language Impairment; Developmental Language Disorder; Autism Spectrum Disorders; diagnostic migration; early childhood intervention.

## 2.1 Introducción

El Trastorno Específico del Lenguaje (TEL) es una alteración de inicio temprano en la adquisición y el uso del lenguaje, en la que las habilidades lingüísticas del niño están significativamente por debajo de su edad cronológica y van acompañadas de un impacto considerable en su funcionamiento diario. Dicha alteración no puede deberse a factores Appendix 2. Migración diagnóstica entre Trastorno Específico del Lenguaje y Trastorno del Espectro Autista: Estudio exploratorio del impacto en los Centros de Atención Infantil Temprana

como discapacidad intelectual, trastorno generalizado del desarrollo, trastornos auditivos o sensoriales, ni a cualquier otra condición médica o diagnóstica, ya que estas condiciones son criterios de exclusión del diagnóstico TEL (Asociación Americana de Psiquiatría [APA], 2014; Leonard, 2014; Organización Mundial de la Salud [OMS], 2001). En los últimos años se ha promovido una flexibilización en los criterios diagnósticos del TEL, ya que la investigación respalda que la presencia de un CI no verbal bajo o medio no conlleva diferencias significativas en el perfil clínico de estos niños y niñas, por lo que el tradicional uso del CI no verbal como criterio de inclusión/exclusión pierde validez teórica (Bishop et al., 2016, 2017; Castro-Rebodello et al., 2004; Martos & Ayuda, 2004; McGregor et al., 2020; Norbury et al., 2016; Paul, 2020; Reilly et al., 2014). Así, el TEL se ha reconceptualizado como un trastorno más amplio, que no incluye limitaciones diagnósticas como CI no verbal y que pierde el carácter de específico, ya que se asume que la afectación del lenguaje puede aparecer sola o ir asociada a otra condición biomédica (Bishop et al., 2016, 2017). Esta nueva perspectiva ha conducido, por un lado, a un cambio terminológico (principalmente en el ámbito anglosajón) que aboga por emplear el término trastorno del desarrollo del lenguaje, y por otro, a la eliminación del criterio de exclusión del nivel normativo de habilidad no verbal para diagnosticar TEL en el DSM-5 (APA, 2014). La prevalencia actual del TEL se estima en torno al 7 % de la población en edad escolar (APA, 2014; Leonard, 2014; Norbury et al., 2016; Tomblin et al., 1997). En España no existen amplios estudios de prevalencia de TEL por lo que se asumen habitualmente las cifras internacionales (Carballo, 2012).

Uno de los trastornos más similares al TEL en edades tempranas es el Trastorno del Espectro Autista (TEA), caracterizado por dificultades persistentes en la comunicación e interacción social y un repertorio restringido y repetitivo de actividades e intereses (APA, 2014; Autism Europe [AE], 2015; Charman & Baird, 2002; OMS, 2001). Al contrario de lo que ocurre en TEL, existen numerosos estudios de prevalencia en población con TEA, estimándose una proporción del 1 % a nivel global (AE, 2015; Málaga et al., 2019; Özerk, 2017).

Tanto niños y niñas con sospecha de TEL como de TEA presentan dificultades comunicativas. Tradicionalmente, se ha considerado que en el TEL estas se encuentran en las áreas fonológica, gramatical y semántica, mientras que los principales déficits en TEA se circunscriben al nivel pragmático. Sin embargo, estos criterios han sido rebatidos

por la investigación más actual que ha encontrado dificultades similares en ambos perfiles (Bishop, 2000; Durrleman & Delage, 2016; Modyanova et al., 2017; Ramírez-Santana et al., 2019; Wittke et al., 2017), demostrando así que la variabilidad del espectro autista y del propio TEL implica perfiles lingüísticos muy heterogéneos que en un porcentaje significativo de casos conduce a confusión entre ambos trastornos, lo cual dificulta el diagnóstico diferencial temprano (Durrleman & Delage, 2016; Kjelgaard & Tager-Flusberg, 2001; Taylor & Whitehouse, 2016; Williams et al., 2008). La posible etiología común de ambos trastornos ha sido objeto de gran debate. Existe una corriente que defiende que se trata de dos condiciones considerablemente diferenciadas con ciertas similitudes superficiales (Schaeffer, 2018; Taylor et al., 2014; Taylor et al., 2012; Williams et al., 2008), mientras que una posición alternativa afirma que dentro de un continuum entre ambas condiciones puede haber casos en los que coexistan rasgos de ambos trastornos (Bishop, 2000; Bishop & Norbury, 2002; Conti-Ramsden et al., 2006; Leyfer et al., 2008; Tomblin, 2011). Esta segunda posición, explicaría el porcentaje significativo de casos en los que se dibujan perfiles intermedios, que son los que presentan más dificultades tanto en evaluación, como en diagnóstico e intervención (Bishop, 2000; Bishop & Norbury, 2002; Martos & Ayuda, 2004).

Actualmente, se han alcanzado técnicas de detección temprana que pueden arrojar los primeros resultados de sospecha de TEL o TEA entre los dos y tres años (Charman, 2010; Charman & Baird, 2002; Martos & Ayuda, 2004; Morales-Hidalgo et al., 2018; Torras-Mañá et al., 2016). En edades tan tempranas, se habla de sospecha debido a que la estabilidad del diagnóstico es menor (Charman & Baird, 2002; Eadie et al., 2014) y está condicionada por las distintas trayectorias que puede seguir el curso del trastorno (Bishop & Norbury, 2002; Charman, 2010; Williams et al., 2008). Factores como la severidad del trastorno (Goodwin et al., 2017; Martos & Ayuda, 2004; Zablotsky et al., 2017) o determinadas características sociodemográficas (Mandell et al., 2005) favorecen el diagnóstico temprano, ya que permiten detectar anomalías del desarrollo con mayor antelación. Los primeros diagnósticos de sospecha son de gran utilidad, ya que conducen al inicio temprano de la intervención, lo cual se relaciona con un mejor pronóstico de la evolución (Bennett et al., 2014; Bishop, 2000; Charman & Baird, 2002; Eadie et al., 2014; Mandell et al., 2005; Martos & Ayuda, 2004; Nicholas et al., 2009; Özerk, 2017; Reilly et al., 2014; Zablotsky et al., 2017). Sin embargo, en estas primeras etapas la estabilidad del diagnóstico es menor, siendo posible que este evolucione hacia perfiles distintos repercuten en la estabilidad del diagnóstico conduciendo, en ocasiones, a un fenómeno conocido como migración diagnóstica.

El término 'migración diagnóstica' surgió para referirse al cambio de diagnóstico en algunos individuos que en el momento del diagnóstico inicial son incluidos en una categoría diagnóstica, pero con el paso del tiempo cumplen los criterios de otra, ya sea por cambios en los criterios de clasificación, los métodos y procedimientos de evaluación, o por las características individuales del sujeto y del trastorno (Cardinal & Griffiths, 2016; Özerk, 2017). Otros autores se refieren al mismo fenómeno como 'sustitución diagnóstica' (King & Bearman, 2009). Este fenómeno se ha atribuido mayoritariamente a la modificación de criterios de los principales manuales de clasificación (Bishop et al., 2008; Coo et al., 2008; Howlin, 2008; King & Bearman, 2009; Newschaffer, 2006; Shattuck, 2006) y ha sido ampliamente estudiado para entender las causas del reciente aumento de prevalencia en el diagnóstico de TEA, ya que podría explicar un alto porcentaje del auge de casos de autismo en las últimas décadas (Cardinal & Griffiths, 2016; Coo et al., 2008; Howlin, 2008; King & Bearman, 2009; Newschaffer, 2006; Özerk, 2017; Shattuck, 2006). Asimismo, este concepto justificaría que un elevado número de individuos originalmente diagnosticados con TEL en sus diferentes subtipos pudieran ser incluidos en la categoría TEA en la adultez según los criterios diagnósticos actuales (Bishop et al., 2008).

El concepto de migración diagnóstica es reciente y los estudios publicados hasta la fecha se han centrado en evaluar su incidencia, olvidando prestar atención a las consecuencias clínicas y emocionales que puedan ir asociadas a él. Dado que la investigación y la práctica clínica coinciden en señalar que el diagnóstico temprano es fundamental para el inicio y la orientación de la intervención, el despliegue adecuado de recursos y, en última instancia, la mejora del pronóstico del menor (Bennett et al., 2014; Bishop, 2000; Charman & Baird, 2002; Eadie et al., 2014; Mandell et al., 2005; Martos & Ayuda, 2004; Nicholas et al., 2009; Özerk, 2017; Reilly et al., 2014; Zablotsky et al., 2017), se podría suponer que la migración diagnóstica implicaría la inespecificidad de la

intervención y la pérdida de un tiempo muy valioso, con las consiguientes repercusiones en el pronóstico del menor.

Por otra parte, también se podría asumir que este fenómeno tiene un importante coste emocional para las familias, ya que la aceptación y el afrontamiento del diagnóstico en edades tempranas son cuestiones muy delicadas para ellas (Charman & Baird, 2002). Karst y Vaughan van Hecke (2012) destacan que tras un periodo de incertidumbre que comienza con las primeras sospechas de trastorno por parte de la familia, esta experimenta una sensación de alivio al recibir el diagnóstico, ya que empieza a comprender y dar sentido a las dificultades del menor. Igualmente, Charman (2010) señala que los falsos positivos suponen un mayor coste para los servicios, mientras que en los falsos negativos el coste incide sobre la familia y el menor. Esta afirmación junto a la reciente incorporación y expansión del modelo centrado en la familia en el ámbito de la atención temprana (AT; Escorcia-Mora et al., 2018; Fernández Valero et al., 2020; McWilliam, 2016; Serrano et al., 2017), contrastan con la falta de atención a este efecto.

En el presente trabajo se pretende analizar la incidencia y la repercusión del fenómeno migración diagnóstica en AT. La AT se define como el conjunto de acciones enfocadas a atender las necesidades puntuales o permanentes de niños y niñas de cero a seis años con trastornos del desarrollo o riesgo de padecerlos, abarcando la intervención sobre el menor, la familia y el entorno (Federación Estatal de Asociaciones de Profesionales de Atención Temprana [GAT], 2000). Desde sus orígenes y expansión en los años 60 y 70, la AT ha evolucionado al ritmo en que lo han hecho las investigaciones sobre el desarrollo. Así, en las últimas décadas ha adoptado un enfoque neuroconstructivista que, entre otros supuestos, invita a ir más allá de la conducta observable ahondando en los diferentes mecanismos cognitivos que pueden esconderse tras un mismo comportamiento, subrayando el papel activo del menor, así como la importancia de sus interacciones con el entorno para el aprendizaje y el desarrollo individual (Campos, 2018; Tolchinski, 2018). Igualmente, en los últimos años ha crecido exponencialmente el interés de la investigación por la AT (Dunst, 2018b), contribuyendo así al auge en la implantación de prácticas basadas en la evidencia (PBE) a nivel internacional. Entre estas PBE destacan, por su relevancia para la temática del estudio, aquellas centradas en la evaluación comprensiva que considera al niño en su globalidad, centrándose en las fortalezas del pequeño en lugar de en sus dificultades, haciendo

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partícipe a la familia desde el momento de la evaluación inicial, atendiendo a sus necesidades e inquietudes, y devolviendo juicios clínicos de una manera comprensible y empática (Dunst, 2018a; Division for Early Childhood [DEC], 2014; Workgroup on Principles and Practices in Natural Environments, 2008). En España los principales cambios que se están incorporando en AT para incluir PBE hacen referencia al modelo centrado en la familia, que invita a enfocar las intervenciones en las prioridades e inquietudes de la familia, aprovechando sus intereses y fortalezas y proveyéndoles de apoyos y recursos para que sean agentes activos en todo el proceso (Serrano et al., 2017). Igualmente, existen diferentes agentes sociales involucrados en la AT, siendo los Centros de Atención Infantil Temprana (CAITs) los encargados de la intervención más directa con los menores y sus familias. Por tanto, los objetivos de este estudio son (1) Cuantificar el fenómeno migración diagnóstica entre TEL y TEA en AT y (2) Evaluar su impacto en los CAITs de la provincia de Córdoba.

Basándonos en la literatura consultada, la hipótesis de partida es que se encontrará un porcentaje significativo de migración diagnóstica entre TEL y TEA en AT (H1). Igualmente, considerando las similitudes entre TEL y TEA en el desarrollo temprano, el aumento exponencial del diagnóstico de TEA y la relativa inestabilidad del diagnóstico temprano, se estima que este fenómeno se dará principalmente en diagnósticos TEA que evolucionen a TEL, mientras que a la inversa será poco frecuente (H2). De no hallar incidencia de este fenómeno, se respaldaría la idea de que las variaciones diagnósticas son marginales y se deben eminentemente a la evolución propia del desarrollo del individuo, restando validez empírica al fenómeno en cuestión. Respecto a la repercusión a nivel clínico, de pronóstico y familiar, se espera encontrar un impacto negativo significativo en los tres niveles según la percepción de los profesionales de AT (H3), ya que la investigación ha correlacionado el diagnóstico temprano y el inicio de la intervención con la mejora del pronóstico y ha resaltado el impacto del diagnóstico sobre la vida familiar.

## 2.2 Método

Esta investigación cuenta con la correspondiente aprobación por parte del Comité de Ética de la Investigación de Córdoba con fecha seis de marzo de 2019, confirmando así que cumple con los principios establecidos en la Declaración de Helsinki de 1964, en la Asociación Médica Mundial, en el Convenio del Consejo de Europa de 1996, relativo

a los derechos humanos y a la biomedicina, así como en el Real Decreto-ley 5/2018, de 27 de julio, sobre protección de datos.

# 2.2.1 Participantes

En España la regulación de la AT depende de las comunidades autónomas, siendo el Decreto 85/2016, de 26 de abril por el que se rige su organización y funcionamiento en Andalucía. Según este decreto, el pediatra de atención primaria es quien inicia el protocolo de derivación del caso a las Unidades de Atención Infantil Temprana (UAITs) ante una sospecha de trastorno del desarrollo. Este decreto también contempla que sea el Equipo Provincial de Atención Infantil Temprana quien derive directamente el caso a la UAIT cuando la detección del posible trastorno tiene lugar en el segundo ciclo de educación infantil. Las UAITs evalúan el caso, realizan un diagnóstico funcional de las necesidades del menor, la familia y el entorno y estipulan si existe necesidad de recibir intervención temprana en CAIT. Igualmente, el decreto establece que en la comunidad autónoma de Andalucía el diagnóstico se rige por la Organización Diagnóstica de Atención Temprana (ODAT; GAT, 2004), si bien todos los diagnósticos de AT son corroborados y emitidos por la Unidad de Salud Mental Infantojuvenil del Hospital Universitario Reina Sofía de Córdoba, que basa sus juicios clínicos en los criterios CIE-11 y DSM-5.

La provincia de Córdoba cuenta con 23 CAITs de financiación pública. Todos ellos fueron invitados a participar de manera voluntaria en el estudio. Diez centros enviaron sus respuestas (tres de la capital y seis de los municipios) proporcionando información de un total de 282 casos (16 TEL y 266 TEA).

# 2.2.2 Instrumentos

Para obtener los datos de interés se utilizó un instrumento de evaluación *ad hoc* de elaboración propia, configurado específicamente para el propósito de este estudio, redactado por un panel de expertos en el ámbito académico, investigación y AT, y distribuido a través de la plataforma Google formulario (Suplemento 1). El primer borrador del cuestionario fue enviado a expertos de igual perfil para que dieran su opinión. Cada experto recibió un documento que contenía una explicación de cómo se había construido y contestó a nuestra solicitud con sus propios comentarios y sugerencias. Las observaciones de los revisores contribuyeron a detectar posibles carencias y omisiones de

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alguna faceta relevante. Así, el cuestionario final consta de 13 preguntas. Dos de ellas abiertas para identificar el centro y permitir comentarios o sugerencias respecto a las cuestiones planteadas (preguntas 1 y 6). Dos preguntas de opción múltiple relativas a la incidencia del fenómeno migración diagnóstica y la valoración de los profesionales de las dificultades del diagnóstico diferencial temprano en TEL y TEA (preguntas 2 y 3, la primera dicotómica y la segunda con escala tipo Likert de 5 puntos donde 1 corresponde a "Nada difícil" y 5 a "Muy difícil"). Una pregunta abierta con seis subapartados en los que se solicitan datos cuantitativos de los últimos cinco años (2015-2020) en relación a: a) Número de diagnósticos iniciales de TEA y TEL (preguntas 4.1 y 4.4); b) Número de casos que inicialmente fueron diagnosticados de TEA y evolucionaron a TEL y a la inversa (preguntas 4.2 y 4.5); c) Número de casos que inicialmente fueron diagnosticados de TEA y TEL pero evolucionaron a otro trastorno indicando a qué trastorno o trastornos evolucionaron (preguntas 4.3 y 4.6). Finalmente, se incluye una pregunta de opción múltiple (escala tipo Likert de 5 puntos donde 1 corresponde a "Muy negativo" y 5 a "Muy positivo") con tres subapartados en los que se aborda la valoración de los profesionales sobre el impacto del cambio de diagnóstico en el proceso de intervención, el pronóstico del menor y la familia (preguntas 5.1, 5.2 y 5.3 correlativamente).

Así, las preguntas 2 a la 4 (con sus correspondientes subapartados) tratan de dar respuesta al objetivo 1, mientras que la pregunta 5 (con sus correspondientes subapartados) aborda el objetivo 2, sirviendo las preguntas 1 y 6 de control.

#### 2.2.3 Diseño y Procedimiento

La investigación realizada es un estudio descriptivo de poblaciones mediante encuesta probabilística (Montero & León, 2007). El procedimiento se llevó a cabo íntegramente de manera telemática. En un primer momento, se contactó con todos los CAITs públicos de la provincia de Córdoba para informar del propósito del estudio y solicitar su participación voluntaria, adjuntando el enlace al cuestionario en formato online. Los cuestionarios fueron cumplimentados por diferentes profesionales, todos ellos con más de cinco años de experiencia en el ámbito de la AT. La recogida de datos se llevó a cabo de manera colaborativa, contando con la participación del conjunto de profesionales de cada centro y la supervisión de la dirección del mismo (principalmente la información relativa a los ítems 5.1-5.3 se llevó a cabo de manera consensuada partiendo de la experiencia y la percepción de los diferentes profesionales). Al cabo de

unas semanas, se envió un recordatorio del estudio a aquellos centros que no habían emitido respuesta. Finalmente, tras un periodo de espera de tres meses para la recepción de respuestas (marzo-mayo de 2020), estas se recopilaron y se procedió al análisis de datos con el paquete estadístico Statistical Package for the Social Sciences, en su versión 21 (SPSS 21; IBM, 2012). Dada la naturaleza eminentemente descriptiva del estudio, se han realizado análisis de frecuencias y porcentajes para dar respuesta a las hipótesis de una manera visual y comprensiva.

### 2.3 Resultados

De los diez profesionales de AT que respondieron al cuestionario se deriva que su percepción sobre la incidencia del fenómeno migración diagnóstica en sus centros (pregunta 2, dicotómica) es desigual, ya que un 40 % indicó que sí considera que el fenómeno ocurre en sus centros, mientras que un 60 % señaló lo contrario. Por otro lado, un 60 % de los profesionales reconoció la dificultad de diferenciar entre ambos trastornos a edades tempranas (pregunta 3, escala tipo Likert de 5 puntos), un 20 % estimó que la dificultad era media, un 10 % indicó que la tarea resultaba muy difícil y otro 10 % señaló que no era difícil; en ningún caso se señaló la opción "nada difícil" en esta pregunta.

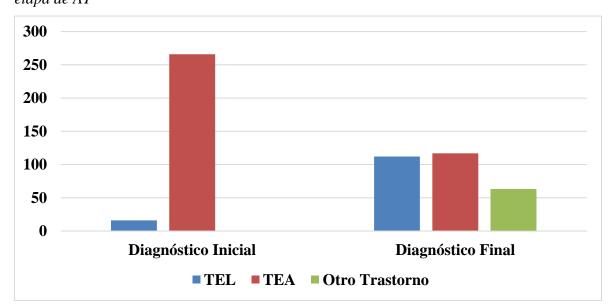
Los datos cuantitativos sobre migración diagnóstica (pregunta 4 con sus correspondientes apartados) aparecen reflejados en la Tabla A2.1 y la Figura A2.1. Se puede observar que existe una gran diferencia entre el número de diagnósticos iniciales de TEL y TEA, encontrándose una considerable mayoría de casos de TEA respecto al menor número de casos de TEL. Este dato se corrobora al comprobar que el 100 % de los centros participantes reportaron casos de TEA, mientras que solo el 50 % de ellos reportó algún caso de TEL. Igualmente, se observa que un elevado porcentaje de casos de TEA finalizan la etapa de AT con un cambio de diagnóstico (56.77 %), en su mayoría hacia TEL (33.83 %) pero también hacia otros trastornos como el retraso simple del lenguaje o el retraso evolutivo. Por otro lado, a pesar del menor número de casos de TEA también se observa un porcentaje significativo de migración diagnóstica (25 %) hacia TEA o hacia otros trastornos, en este caso retraso simple del lenguaje.

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	f	%
Casos TEA	266	100
Migración TEA-TEL	90	33.83
Migración TEA-Otro trastorno	61	22.93
Total migración TEA	151	56.77
Casos TEL	16	100
Migración TEL-TEA	2	12.5
Migración TEL-Otro trastorno	2	12.5
Total migración TEL	4	25

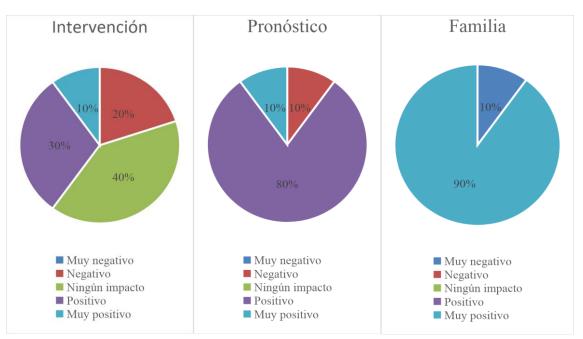
 Table A2.1. Análisis de frecuencias de los casos de migración diagnóstica

**Figure A2.1.** Evolución del diagnóstico desde la evaluación inicial hasta el final de la etapa de AT



Finalmente, los resultados de la valoración de los profesionales del impacto del fenómeno en el proceso de intervención, el pronóstico del menor y la familia (pregunta 5 con subapartados, escala tipo Likert de 5 puntos) se muestran en la Figura A2.2. Así, se observa que las opiniones de los profesionales respecto a la repercusión del fenómeno en el proceso de intervención son más heterogéneas, valorándose mayormente como inocua. Respecto al efecto sobre el pronóstico y la familia parece haber un mayor consenso en

que el cambio de diagnóstico afecta positiva y muy positivamente a estos ámbitos, lo cual se discute con más detalle en el siguiente apartado.



**Figure A2.2.** Impacto del fenómeno migración diagnóstica en el proceso de intervención, el pronóstico del menor y la familia

# 2.4 Discusión

Los objetivos de este estudio se centraban en analizar el fenómeno migración diagnóstica en los CAITs de la provincia de Córdoba a partir de la cuantificación y evaluación de su impacto. Así, hemos constatado que el fenómeno tiene una alta incidencia en la provincia de Córdoba, a pesar de la percepción inicial de los profesionales. En ese sentido, resulta llamativo que en un principio se indicara mayoritariamente que el fenómeno no tenía lugar en los centros, mientras que los datos cuantitativos respaldan una alta incidencia de migración diagnóstica, lo cual refleja el desconocimiento que existe del fenómeno, cuya investigación es relativamente escasa hasta la fecha. Por otro lado, en la segunda pregunta los profesionales asignaban una elevada dificultad a la distinción entre TEA y TEL en AT, lo cual refuerza la necesidad de seguir investigando en metodologías, técnicas e instrumentos que contribuyan al diagnóstico diferencial temprano, así como favorecer la accesibilidad del conocimiento científico para que llegue a los profesionales, promoviendo asimismo la incorporación de PBE en los CAITs.

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Igualmente, se ha observado que existe un porcentaje significativo de casos que migran de un diagnóstico a otro y que esta migración tiene lugar mayoritariamente desde TEA hacia TEL, corroborando así las H1 y H2. Sin embargo, este fenómeno no solo ocurre en TEA, donde el número de casos es elevado, sino también en TEL, a pesar de que las cifras son considerablemente menores. Algunos autores señalan que a edades tempranas se observa un mayor solapamiento de fenotipos en los patrones lingüístico y comportamental de ambas condiciones, pero con el tiempo estos evolucionan de manera que en edad escolar las diferencias entre ellos son más pronunciadas (Weismer, 2013; Williams et al., 2008). Esta evolución propia del trastorno podría explicar la alta incidencia del fenómeno migración diagnóstica, ya que a medida que el menor va creciendo se va concretando su perfil y se clarifica el diagnóstico. Por otro lado, el hecho de que más de un 50 % de los diagnósticos iniciales de TEA finalicen la etapa de AT con otro diagnóstico podría estar relacionado con el aumento exponencial en la prevalencia de este trastorno en los últimos años. La investigación señala como posibles causas de este incremento la ampliación de los criterios diagnósticos (conceptualización del TEA como un espectro de amplia variabilidad tanto en habilidades como en dificultades, en lugar de una categoría cerrada), perfeccionamiento de los servicios diagnósticos, protocolos y técnicas de evaluación e intervención, nuevos métodos de estudio que han llevado a un mayor conocimiento del trastorno, mayor conciencia social del trastorno (mayor nivel de información sobre autismo tanto en familias como en profesionales médicos, educativos y agentes sociales), sin descartar la posibilidad de cierto incremento real de la prevalencia (Baird et al., 2006; Hammad et al., 2019; Howlin, 2008; Málaga et al., 2019; Özerk, 2017; Pérez-Crespo et al., 2019). Estas razones, si bien han contribuido a la mejora del diagnóstico precoz, también podrían haber conducido a un elevado número de falsos positivos, es decir, niños y niñas que en un primer momento presentaban sospecha de TEA, pero cuya trayectoria evolutiva unida a la eficacia de las intervenciones de inicio temprano ha derivado en un perfil distinto. En futuras investigaciones se debería explorar esta hipótesis a la vista de los datos tan significativos sobre migración diagnóstica en TEA.

Respecto a la repercusión del fenómeno a nivel clínico y familiar, nuestra H3 no se ha confirmado ya que, en general, la valoración de los profesionales del impacto del fenómeno sobre el pronóstico del menor y sobre la familia ha sido positiva o muy positiva. Este dato podría explicarse por el hecho de que las migraciones diagnósticas reportadas

han sido mayoritariamente desde un trastorno más global y con mayor estigma social (TEA, trastorno global con importantes dificultades a nivel socioemocional) a uno específico y menos estigmatizado (TEL, trastorno específico con menor afectación social). Esto incidiría en el pronóstico del menor (estimándose una mejor evolución) y en la capacidad de afrontamiento de la familia. Recientes estudios tanto en familias de menores con TEA (Crane et al., 2016) como familias de menores con TEL (Ash et al., 2020) han reportado elevados índices de estrés e insatisfacción entre las familias durante el proceso diagnóstico debido, entre otras variables, a la falta de información, la imprecisión terminológica, la vía de comunicación familia-profesional o el apoyo tras el diagnóstico. Así, resulta inevitable aludir al modelo centrado en las familias que aspira a incluirlas en el proceso de AT desde los primeros contactos, asumiendo por parte del profesional un estilo de comunicación comprensible y comprensivo para ellas (DEC, 2014; Escorcia-Mora et al., 2018; Fernández Valero et al., 2020; McWilliam, 2016; Serrano et al., 2017). La aplicación de este modelo ha sido relacionada con una mejora notable en la satisfacción de las familias con el proceso y la evolución del pronóstico del menor (DEC, 2014). Por otro lado, los efectos del fenómeno sobre la intervención se perfilan más heterogéneos, ya que las valoraciones oscilaban desde 'negativo' hasta 'muy positivo', con un mayor porcentaje de respuestas de 'ningún impacto'. Esto puede deberse a que las intervenciones actuales tienen un carácter individualizado y funcional, es decir, no dependen tanto de la categoría diagnóstica en la que se ubica al menor como de sus características individuales y necesidades de apoyo, por lo que el cambio de diagnóstico no tendría el mismo impacto que en los demás componentes. Este supuesto se alinea con las PBE que resaltan la importancia de desarrollar intervenciones dinámicas centradas en las necesidades, preferencias, habilidades y valores culturales del menor y la familia, tratando de insertarse en la rutina diaria para aprovechar todas las oportunidades de aprendizaje que proporciona el contexto (Dunst, 2018a; Workgroup on Principles and Practices in Natural Environments, 2008).

En la pregunta 6 en la que se animaba a los profesionales a expresar sus comentarios o preocupaciones respecto al objeto del estudio se recopilaron comentarios que nos parece interesante reportar. Así, se indicó que los trastornos del neurodesarrollo deben considerarse como dimensiones, lo que implica que debe contemplarse cierta flexibilidad y que deben servir como orientación para profesionales y familias, no como categorías cerradas. También se manifestó que el TEL es un diagnóstico difícil de detectar

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a edades tempranas por lo que los niños y niñas a menudo llegan a los CAITs con sospecha de TEA y conforme avanzan la evaluación y la intervención se van concretando sus perfiles. Esta afirmación va en línea con los datos de este estudio que encuentran un elevado número de diagnósticos iniciales de TEA que se ven modificados al final de la etapa de AT. Por último, se señaló que el impacto de la migración diagnóstica sobre las intervenciones es menor debido a la funcionalidad e individualización de estas, lo que las hace menos vulnerables a variaciones diagnósticas. Estos comentarios refuerzan los datos discutidos en este apartado y avalados por la investigación, constatando así la dificultad del diagnóstico diferencial temprano, y la importancia de diagnósticos e intervenciones flexibles e individualizados (Bishop & Norbury, 2002; Castro-Rebodello et al., 2004; Conti-Ramsden et al., 2006; Leyfer et al., 2008; Weismer, 2013). Asimismo, es interesante señalar que estos comentarios respaldan el actual modelo neuroconstructivista que predomina en AT, según el cual las dificultades se abordan de manera funcional, trabajando en dimensiones interdependientes, respetando el proceso de especialización progresiva de los diferentes dominios a partir de la experiencia y la interacción con el ambiente, y profundizando en los mecanismos subyacentes a las conductas observables (Campos, 2018; Tolchinski, 2018). De este modo, se cuestiona la obsoleta visión mecanicista de la psicología centrada en el déficit que durante muchos años ha invadido el ámbito de AT influyendo así en las prácticas aplicadas.

Finalmente, es importante señalar como limitaciones del estudio la representatividad de la muestra, cercana al 50 % de los centros de la provincia de Córdoba. Igualmente, la decisión de no solicitar información más exhaustiva sobre menores y centros, evitando incumplir la legislación en materia de protección de datos y aligerando el proceso de cumplimentación del cuestionario, ha impedido una mayor definición de la muestra del estudio y la extracción de conclusiones más precisas sobre el fenómeno migración diagnóstica relacionándolo con otras variables sociodemográficas. Por último, el instrumento de evaluación utilizado para la obtención de datos ha sido un cuestionario creado *ad hoc* para cuantificar los aspectos específicos que son objeto de este estudio y no ha sido sometido por el momento a un proceso de validación formal de sus propiedades psicométricas. Si bien este instrumento fue elaborado por un panel de expertos que consensuó la definición del constructo migración diagnóstica, el tipo de preguntas a incluir y la redacción de las mismas, y revisado por otro grupo de expertos de perfil similar para abarcar los aspectos más relevantes del fenómeno teniendo en cuenta

la realidad de los CAITs. Las citadas limitaciones inducen la necesidad de ser precavidos a la hora de interpretar y generalizar los resultados. Si bien en un principio este estudio fue diseñado como un trabajo exploratorio; consideramos que los datos que se desprenden de él abren una nueva línea de investigación en el campo de la AT, y justifican la continuidad de su estudio ampliando el tamaño de la muestra y la información sobre sus características clínicas y sociodemográficas. Asimismo, queda justificada la necesidad de crear un instrumento validado más amplio para indagar en el constructo y comparar resultados con otras áreas geográficas. Igualmente, considerando el elevado impacto emocional del proceso diagnóstico en las familias y su papel fundamental en el proceso de AT, se debería incorporar su visión sobre el fenómeno y su impacto en aras de una mayor comprensión del mismo y su repercusión. Así, en este trabajo se ha desvelado una interesante tendencia que apunta a una elevada incidencia del fenómeno migración diagnóstica en la que conviene seguir profundizando para comprender la naturaleza de estos trastornos y facilitar la labor de los CAITs.

### 2.5 Conclusiones

El presente estudio es el primero hasta la fecha que analiza el fenómeno migración diagnóstica entre TEL y TEA en población española. En este sentido, si bien la muestra hace difícil generalizar los resultados a toda la población, los datos apuntan a que existe un porcentaje significativo de migraciones diagnósticas en AT que debería seguir explorándose en estudios futuros debido a su importante repercusión en la práctica clínica, así como en la vida del menor y la familia. Una mejor comprensión de este fenómeno ampliaría el conocimiento de la naturaleza del TEL y el TEA a edades tempranas, contribuyendo así a la labor de los CAITs. Igualmente, debería hacernos reflexionar a los profesionales de este ámbito acerca de las lagunas y los déficits de los actuales procesos de evaluación en AT con vistas a seguir trabajando en su perfeccionamiento.

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- Appendix 2. Migración diagnóstica entre Trastorno Específico del Lenguaje y Trastorno del Espectro Autista: Estudio exploratorio del impacto en los Centros de Atención Infantil Temprana development, third edition. Journal of Intellectual Disability Research, 60(5), 502-511. https://doi.org/10.1111/jir.12291
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## Suplemento 1. Cuestionario

# Estudio prevalencia de diagnóstico en trastornos del neurodesarrollo

- Indique su centro de pertenencia (esta información es exclusivamente de control, en ningún caso será publicada o considerada objeto de análisis):
- ¿Considera que en su centro existe un elevado número de casos diagnosticados inicialmente como Trastorno del Espectro Autista (TEA) que evolucionaron a Trastorno Específico del Lenguaje (TEL) (o a la inversa) antes de recibir el alta? (marque una X donde corresponda):

Sí	No	No sabe/ No contesta

 Evalúe la dificultad de diferenciar entre TEA y TEL en las primeras etapas del desarrollo, siendo 1 Nada difícil y 5 Muy difícil (marque una X donde corresponda):

1	2	3	4	5

 A continuación, le pedimos datos sobre número de niños/as con determinados diagnósticos SOLO DE LOS ÚLTIMOS 5 AÑOS:

	Número de	
	casos	
4.1. Número de diagnósticos iniciales TEA		

Appendix 2. Migración diagnóstica entre Trastorno Específico del Lenguaje y Trastorno del Espectro Autista: Estudio exploratorio del impacto en los Centros de Atención Infantil Temprana

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4.2. Número de casos que inicialmente fueron	
diagnosticados de TEA y evolucionaron a TEL	
4.3. Número de casos que inicialmente fueron	
4.5. Rumero de casos que iniciamente rucron	
diagnosticados de TEA y evolucionaron a otro trastorno	
(indique a qué trastorno o trastornos evolucionaron)	
4.4. Número de diagnósticos iniciales TEL	
4.5. Número de casos que inicialmente fueron	
diagnosticados de TEL y evolucionaron a TEA	
4.6. Número de casos que inicialmente fueron	
diagnosticados de TEL y evolucionaron a otro trastorno	
(indique a qué trastorno o trastornos evolucionaron)	

5. Evalúe el impacto que puede tener el cambio diagnóstico de TEA a TEL sobre los siguientes elementos (marque una X donde corresponda):

	Muy negativo	Negativo	Ningún impacto	Positivo	Muy positivo
5.1 Intervención					
5.2 Pronóstico					
5.3 Familia					

 Finalmente, rellene este espacio si tiene alguna sugerencia o comentario respecto a las cuestiones planteadas:

Muchas gracias por su colaboración.

# Appendix 3. Formal Approval from the Research Ethics Committee of Córdoba



Servicio Andaluz de Salud CONSEJERÍA DE SALUD

Hospital Universitario Reina Sofia

Eduardo Morán Fernández, Secretario en funciones del Comité de Ética de la Investigación de Córdoba, comité constituido a tenor de lo establecido en el Decreto 439/2010, de 14 de diciembre, por el que se regulan los órganos de ética asistencial y de la investigación biomédica de Andalucía (BOJA núm. 251 de 27 de diciembre) del que es Presidenta Inmaculada Concepción Herrera Arroyo

### CERTIFICA

Que en la reunión del Comité de Ética de Investigación de Córdoba celebrada el día 26 de febrero de 2019 (Acta nº 285, ref. 4154), se ha estudiado y evaluado el Proyecto de Investigación, titulado: "ANÁLISIS DIFERENCIAL DE LA COMPETENCIA EN RECONOCIMIENTO EMOCIONAL EN POBLACIÓN INFANTIL CON TRASTORNO DEL ESPECTRO AUTISTA Y TRASTORNO ESPECÍFICO DEL LENGUAJE", Cód. Protocolo Tesis/TEA-TEL, Protocolo versión 2 - 20/02/2019 y Hoja de Información al Paciente y Consentimiento Informado versión 1 - 13/12/2018, en el que figura como Investigadora principal Dña. Julia Vacas Ruiz, de la Universidad de Córdoba, habiendo considerado los integrantes de dicho Comité que el citado estudio respeta los principios fundamentales establecidos en la Declaración de Helsinki de 1964, de la Asociación Médica Mundial, y enmiendas posteriores, y en el Convenio del Consejo de Europa de 1996, relativo a los Derechos Humanos y a la Biomedicina, demostrando sus autores conocer suficientemente los antecedentes y el estado actual del tema que proponen investigar, estando bien definidos sus objetivos y siendo adecuada su metodología, por lo que hacen constar la viabilidad en todos sus términos del proyecto de investigación, estimando que los resultados pueden ser de gran interés.

Se hace constar, de acuerdo con el artículo 18 de la Ley 40/2015, de 1 de octubre, de Régimen Jurídico del Sector Público, que la presente certificación se emite con anterioridad a la aprobación del acta correspondiente.

En Córdoba, a 6 de marzo de 2019

EL SECRETARIO

LA PRESIDENTA INVE dian.

Fdo.: Eduardo Morán Fernández, Secretario en funciones aculada Concepción Herrera Arroyo



Servicio Andaluz de Salud CONSEJERÍA DE SALUD

Hospital Universitario Reina Sofia

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El Comité de Ética de la Investigación de Córdoba está constituido por los siguientes vocales: PRESIDENTA Dña. Inmaculada Concepción Herrera Arroyo. Jefe de Servicio Hematología del HURS VICEPRESIDENTE D. José Luis Barranco Quintana. FEA Medicina Preventiva del HURS SECRETARIO D. Eduardo Morán Fernández. FEA Medicina Intensiva H Infanta Margarita de Cabra, Secretario en funciones VOCALES D. Gregorio Jurado Cálz. Técnico de Función Administrativa. Licenciado en Derecho del HURS D. Juan Manuel Parras Rejano. Medico de Familia EBAP, Área Santaria Norte de Córdoba D. Eduardo Morán Fernández, FEA Medicina Intensiva H Infanta Margarita de Cabra D. Javier Caballero Villarraso, FEA Bioquímica Clínica del HURS Dña. Beatriz García Robredo. Farmacéutica de Atención Primaria del Área Sanitaria Norte D. Rafael Segura Saint-Gerons. Odontólogo C.S. La Carlota. Distrito Sanitario Guadalquivir Dña. María Mercedes Gil Campos. FEA Pediatria del HURS Dia. India Mercedes Gii Campos, FEA Pediatria del HURS D. Carlos José Pérula de Torres. Enfermeno de Familia de Atención Primaria. Distrito Sanitario Córdoba Dña. India José Párula de Torres. Enfermeno de HURS Dña. India Carmen Redriguez Fernández. FEA Traumatología HURS Dña. India Carmen Redriguez Garcia. Enfermena del HURS Dña. Sonia García Cabezas. FEA Oncología Radioterápica del HURS D. Antonio Díaz Valenzuela. Enfermero de la Agencia Pública Empresarial Sanitaria Hospital Alto Guadalquivir. CHARE Puente Geoli Puente Geni Dña, Eva Mª Rojas Calvo. Auxiliar Administrativo HURS. Licenciada en Derecho D. Miguel Ángel Romero Moreno. FEA Cardiología del HURS D. Manuel Jesús Cárdenas Aranzana. Farmacéutico Hospitalario del HURS D. Félix Igea Arisqueta. Médico de Familia Área Sanitaria Norte de Córdoba Dña. Maria Pleguezuelo Navarro. FEA Digestivo del HURS Que dicho Comité está constituido y actúa de acuerdo con la normativa vigente y las directrices de la Conferencia Internacional de Buena Práctica Clínica.

En Córdoba, a 6 de marzo de 2019

EL SECRETARIO LA PRESIDENTA COMITÉ DE ÉTICA DE LA INVE 302 Fdo.: Eduardo Morán Fernández, Secretario en funciones Fdo: Inmaculada Concepción Herrera Arroyo Avda, Menéndez Pidal, s/n. 14004 Córdoba Tel. 957 010 000 www.hospitalreinasofia.org

### **Appendix 4. Project Information Sheet for Professionals and Families**

#### CONSENTIMIENTO INFORMADO - INFORMACIÓN AL PACIENTE

Este documento es para su información. Por favor léalo detenidamente, y pregunte cualquier duda que le surja. Usted puede leer este documento y preguntar lo que desee, sin que por ello esté obligado a participar en el estudio.

#### 1. Objetivo del estudio

Su hijo/a ha sido seleccionado/a como candidato/a para participar voluntariamente en un estudio que investiga la atención, el reconocimiento y la comprensión de emociones, con el objetivo de explorar nuevas vías para la detección y diferenciación entre distintos trastornos del neurodesarrollo.

Las investigadoras que participan en el estudio son las psicólogas Dra. Araceli Sánchez Raya (directora del CAIT UCO y profesora de la Facultad de Ciencias de la Educación de la UCO), Dra. Adoración Antolí Cabrera (profesora de la Facultad de Ciencias de la Educación de la UCO), Dra. Carolina Pérez Dueñas (profesora de la Facultad de Ciencias de la Educación de la UCO) y la Doctoranda Julia Vacas Ruiz. Todas ellas investigadoras del Instituto Maimónides de Investigación Biomédica de Córdoba (IMIBIC) y pertenecientes al grupo de investigación HUM924.

#### 2. Metodología

Se citará a su hijo/a para realizar tres pruebas experimentales de 3.5 min. aproximadamente cada una. En ellas, su hijo/a sólo tendrá que mirar en la pantalla del ordenador una serie de estímulos que consisten en rostros de personas y objetos, mientras se registran sus movimientos oculares.

Este registro se hará mediante tecnología Eye-Tracking, en este caso se utilizará como instrumento el dispositivo Tobii 2.X que consiste en un pequeño equipo colocado en la pantalla del ordenador junto con un software encargado de captar y procesar los movimientos oculares del observador/a de manera similar a como aparece en la siguiente imagen.



Igualmente, se evaluará el nivel de vocabulario receptivo y la competencia en reconocimiento de emociones del niño/a aplicando dos breves escalas. En el primer caso, se administrará el test de vocabulario en imágenes Peabody-III y, en el segundo, la subescala "percepción social" de la batería NEPSY-II. El procedimiento completo durará entre 25 y 35 minutos aproximadamente, siendo posible dividirlo en dos sesiones según las necesidades del niño/a.

#### 3. Beneficios

La realización de esta investigación podría arrojar nuevos marcadores que faciliten la concreción de un diagnóstico diferencial con la consiguiente repercusión en la intervención y el pronóstico de niños/as con trastornos del neurodesarrollo.

#### 4. Riesgos

La participación de su hijo/a no implica ningún tipo de riesgo, el proceso experimental ha sido diseñado para evitar que resulte invasivo y garantizar la ausencia de cualquier molestia en el/la participante. Los datos serán tratados de manera confidencial conforme al Real Decreto-ley 5/2018, de 27 de julio, de medidas urgentes para la adaptación del Derecho español a la normativa de la Unión Europea en materia de protección de datos y siguiéndose de forma rigurosa los principios éticos promulgados en la declaración de Helsinki por la Asociación Médica Mundial.

#### 5. Carácter voluntario de su participación y terminación del estudio

La participación de su hijo/a en el estudio es de carácter totalmente voluntario. Si decide participar, recibirá esta hoja de información para que la conserve y se le pedirá que firme como representante legal un formulario de consentimiento.

Usted puede negarse a participar y puede retirarse del estudio en cualquier momento sin necesidad de explicar sus motivos, y sin que por ello se altere la relación con el equipo profesional que atiende o atenderá a su hijo/a, ni se produzca perjuicio en su tratamiento. Si usted decide interrumpir su participación en el estudio puede hacerlo notificando su decisión al profesional que le atiende.

El equipo asistencial que le atiende, por su parte, también podrá interrumpir su participación en el estudio si no se considerara un/a candidato/a apropiado/a para poder ser incluido en el mismo, en función de los criterios de inclusión/exclusión.

Por favor, no dude en preguntar a nuestro personal cualquier duda o pregunta que tenga. Si requiere información adicional en cualquier momento del estudio se puede poner en contacto con nuestro personal en el teléfono: 658 43 60 77 o en el correo electrónico: <u>172varuj@gmail.com</u>

Si decide que su hijo/a participe en el estudio, firme el formulario siguiente, en el que se hace constar que se le ha explicado y que entiende toda la información que se le ha proporcionado sobre el estudio.

### **Appendix 5. Informed Consent for Participants' Parents or Caretakers**

#### CONSENTIMIENTO INFORMADO - CONSENTIMIENTO POR ESCRITO DEL PACIENTE

<Análisis Diferencial De La Competencia En Reconocimiento Emocional En Población Infantil Con Trastorno Del Espectro Autista Y Trastorno Específico Del Lenguaje >

Yo (Nombre y Apellidos):.... como padre/madre, tutor/a o representante legal de (Nombre y Apellidos del niño/a):

- · He leído el documento informativo que acompaña a este consentimiento (Información al Paciente)
- He podido hacer preguntas sobre el estudio < Análisis Diferencial De La Competencia En Reconocimiento Emocional En Población Infantil Con Trastorno Del Espectro Autista Y Trastorno Específico Del Lenguaje >
- He recibido suficiente información sobre el estudio < Análisis Diferencial De La Competencia En Reconocimiento Emocional En Población Infantil Con Trastorno Del Espectro Autista Y Trastorno Específico Del Lenguaje > He hablado con el profesional sanitario informador: Julia Vacas Ruiz
- Comprendo que mi participación es voluntaria y soy libre de participar o no en el estudio.
- Se me ha informado que todos los datos obtenidos en este estudio serán confidenciales y se tratarán conforme establece el Real Decreto-ley 5/2018, de 27 de julio, de medidas urgentes para la adaptación del Derecho español a la normativa de la Unión Europea en materia de protección de datos.
- Se me ha informado de que la información obtenida sólo se utilizará para los fines específicos del estudio.
- Deseo ser informado/a de los resultados de la investigación una vez que termine el mismo. Deseo ser informado/a de mis datos de carácter personal que se obtengan en el curso de la investigación, incluidos los descubrimientos inesperados que se puedan producir, siempre que esta información sea necesaria para evitar un grave perjuicio para mi salud o la de mis familiares biológicos.

No

Comprendo que puedo retirarme del estudio:

- Cuando quiera
- · Sin tener que dar explicaciones
- · Sin que esto repercuta en el trato recibido hacia mi persona o en cualquier otra circunstancia

Si

Presto libremente mi conformidad para participar en el proyecto titulado < Análisis Diferencial De La Competencia En Reconocimiento Emocional En Población Infantil Con Trastorno Del Espectro Autista Y Trastorno Específico Del Lenguaje > y autorizo al equipo al frente del proyecto a obtener los datos de carácter personal de mi hijo/a necesarios para completar el estudio, habiendo sido informado/a de que éstos serán tratados de forma anónima y con el estricto cumplimiento de la normativa vigente.

Firma del participante (o representante legal en su caso)

Firma del profesional informador

 Nombre y apellidos: Julia Vacas Ruiz Fecha: .....

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