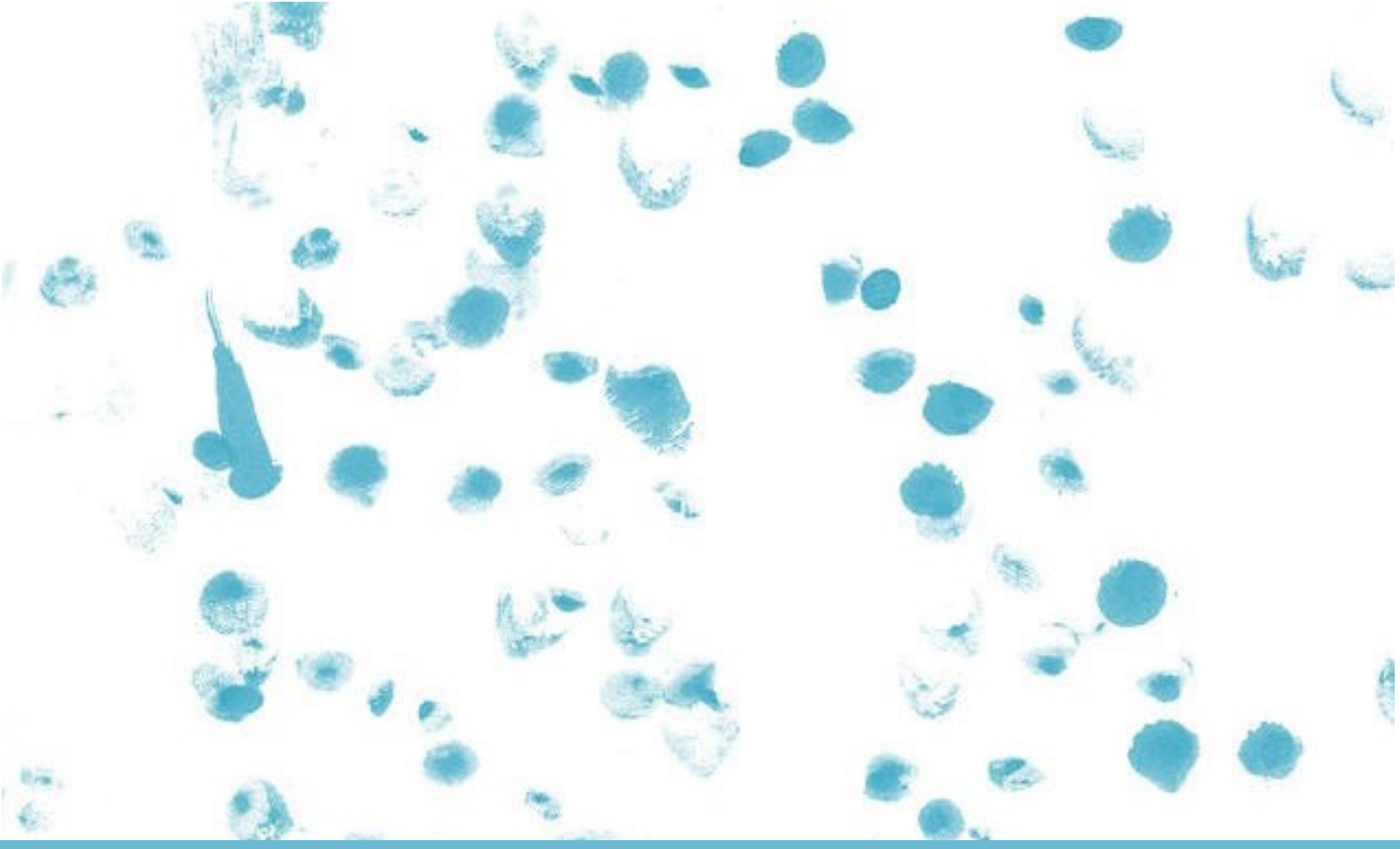

TESIS DOCTORAL

AUTISMO EN EDADES TEMPRANAS: DETECCIÓN DE NECESIDADES E INTERVENCIÓN PRECOZ

Álvaro Bejarano Martín

VNiVERSiDAD D SALAMANCA
CAMPUS DE EXCELENCIA INTERNACIONAL

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UNIVERSIDAD DE SALAMANCA, INSTITUTO UNIVERSITARIO DE INTEGRACIÓN EN LA COMUNIDAD (INICO), CENTRO DE ATENCIÓN INTEGRAL AL AUTISMO (INFOAUTISMO), DEPARTAMENTO DE PERSONALIDAD, EVALUACIÓN Y TRATAMIENTO PSICOLÓGICOS

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CENTRO DE ATENCIÓN INTEGRAL AL AUTISMO (INFOAUTISMO)
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CERTIFICA

Que D. Álvaro Bejarano Martín ha realizado, bajo su dirección, la Tesis Doctoral titulada: "Autismo en edades tempranas: detección de necesidades e intervención precoz" y que esta cumple con los requisitos de calidad, originalidad y presentación requeridos en una investigación científica para optar al grado de Doctor por la Universidad de Salamanca. La presente Tesis Doctoral se presenta en la modalidad de Tesis por Compendio de Artículos/Publicaciones, y opta a la mención de Doctor Internacional.

Para que así conste, y tenga los efectos oportunos, el director firma este certificado en Salamanca, a 28 de mayo de 2021.

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De acuerdo con el RD 99/2011, de 28 de enero, por el que se regulan las enseñanzas oficiales de doctorado, la Comisión de Doctorado y Posgrado de la Universidad de Salamanca establece como posible formato de presentación de Tesis Doctoral, la modalidad de Tesis por Compendio de Artículos/Publicaciones, publicados o aceptados en revistas especializadas y de prestigio. Así, la presente Tesis Doctoral se presenta bajo esta modalidad, optando a la mención de Doctorado Internacional.

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Resumen

Los avances en los cuidados intensivos neonatales han mejorado mucho la tasa de supervivencia de los niños prematuros. Sin embargo, la incidencia de trastornos del neurodesarrollo en este grupo, como el Trastorno del Espectro Autista, es uno de estos problemas de comportamiento observados. El objetivo de la intervención temprana es anticiparse a estas alteraciones, con el fin de aumentar las habilidades de comunicación social y reducir los síntomas en este ámbito. Sin embargo, ninguno de estos programas de intervención temprana ha estudiado su eficacia en prematuros con riesgo de TEA.

Con este fin, se ha llevado a cabo una intervención socio-comunicativa basado en la evidencia científica en base a un estudio de revisión sistemática y metaanálisis, y a un estudio de encuesta sobre las perspectivas de padres y profesionales de menores con TEA. Este programa se enfocó a investigar los resultados en el funcionamiento social, cognitivo, lingüístico y adaptativo en menores prematuros y a término con riesgo de TEA.

En la intervención, se hizo hincapié en la incorporación de estrategias dirigidas a la imitación, la atención conjunta y el juego en las rutinas diarias y las actividades lúdicas. Las variables de resultados de los niños se recogieron mediante medidas estandarizadas y de observación.

Estos datos demostrarán si se pueden producir cambios en los problemas centrales del desarrollo de los menores prematuros y a término con riesgo de TEA con una intervención de baja intensidad dirigida a las habilidades sociales y de comunicación. Por lo tanto, este estudio se centrará en la necesidad de incrementar y mejorar las intervenciones tempranas en menores prematuros con riesgo de TEA dirigidas a las habilidades sociales-comunicativas. Es necesario investigar los efectos a largo plazo de la intervención, además de examinar los posibles mediadores y moderadores de los resultados de la intervención en las habilidades sociales y comunicativas de los menores.

Abstract

Advances in neonatal intensive care have greatly improved the survival rate of premature infants. However, the incidence of neurodevelopmental disorders in this group, such as Autism Spectrum Disorder, is one of these observed behavioral problems. The goal of early intervention is to anticipate these disturbances, in order to increase social communication skills and reduce symptoms in this area. However, none of these early intervention programs has studied their efficacy in preterm infants at risk for ASD.

To this end, an evidence-based social-communication intervention was conducted based on a systematic review and meta-analysis study, and a survey study on the perspectives of parents and professionals of children with ASD. This program focused on investigating outcomes in social, cognitive, language and adaptive functioning in preterm and full-term children at risk for ASD.

In the intervention, emphasis was placed on incorporating strategies aimed at imitation, joint attention and play into daily routines and play activities. Child outcome variables were collected using standardized and observational measures.

These data will demonstrate whether changes in the core developmental problems of preterm and term infants at risk for ASD can occur with a low-intensity intervention targeting social and communication skills. Therefore, this study will focus on the need to increase and improve early interventions in preterm children at risk for ASD targeting social-communication skills. There is a need to investigate the long-term effects of the intervention, in addition to examining possible mediators and moderators of intervention outcomes on children's social and communication skills.

ÍNDICE DE CONTENIDOS

ÍNDICE DE TABLAS

Introducción y justificación de tesis

CONCEPTUALIZACIÓN DEL TRASTORNO DEL ESPECTRO AUTISTA

DEFINICIÓN, CARACTERÍSTICAS Y CLASIFICACIÓN

El trastorno del espectro autista (TEA) es una alteración del neurodesarrollo en la infancia temprana. Se prolonga durante toda la vida y presenta una amplia variabilidad desde su inicio, existiendo también una gran heterogeneidad entre los casos y a lo largo del desarrollo. Las características principales del trastorno mantienen en lo esencial los rasgos que destacó Kanner (1943), sintetizados actualmente en dos características que son un deterioro persistente de la comunicación social recíproca y la interacción social, y patrones de conducta, intereses o actividades restrictivos y repetitivos (Asociación Americana de Psiquiatría – APA, 2013). Estas dos características tienen consecuencias significativas en la vida diaria de la persona y, junto con estos síntomas centrales, pueden concurrir otros trastornos psiquiátricos o neuroevolutivos, entre los que la discapacidad intelectual, el trastorno por déficit de atención con o sin hiperactividad, las alteraciones del lenguaje, la ansiedad, la depresión y la epilepsia son relativamente frecuentes (Hodges et al. 2020).

El autismo es un trastorno heterogéneo cuyo término se ha utilizado de diversas formas para describir tanto una condición más amplia, como un diagnóstico específico cuando se consideraba un subgrupo dentro de la categoría de diagnóstico general de "trastornos generalizados del desarrollo (TGD)", introducido en el Manual diagnóstico y estadístico de trastornos mentales, tercera edición (DSM III) en 1980 sobre la construcción de un espectro más amplio de déficits de comunicación social. Debido a la falta de límites claros en los TGD y a las dificultades que presentan, los sistemas de clasificación categorial actuales, como la Clasificación Internacional de Enfermedades, 11ª Revisión (CIE-11) (OMS, 2018) y el DSM-5 utilizan el término general *TEA*, y diferencian a las personas utilizando indicadores clínicos. Ver Tabla 1.

Tabla 1. Criterios del DSM-5 para el trastorno del espectro autista

A. Déficits persistentes en la comunicación e interacción social en múltiples contextos, que se manifiestan en lo siguiente, en la actualidad o en la historia; debe cumplir los 3 criterios:

Déficits	Ejemplos
1. En reciprocidad socioemocional	- Acercamiento social anormal y fracaso en la conversación normal en ambos sentidos, o disminución en intereses, emociones o afectos compartidos
2. En conductas comunicativas no verbales utilizadas en la interacción social	- Comunicación verbal y no verbal poco integrada, o anomalías en contacto visual el lenguaje corporal o deficiencias en la comprensión y el uso de gestos,
3. En el desarrollo, mantenimiento y comprensión de relaciones	- Dificultades para ajustar el comportamiento a los diversos contextos sociales; o para compartir juegos imaginativos o para hacer amigos

B. Patrones restrictivos y repetitivos de comportamiento, intereses o actividades, que se manifiestan en dos o más de los siguientes criterios, actualmente o por los antecedentes:

Déficits	Ejemplos
1. Movimientos, uso de objetos o habla estereotipados o repetitivos	- Estereotipias motoras simples, alinear juguetes o girar objetos, ecolalia, frases idiosincrásicas
2. Insistencia en la monotonía, excesiva inflexibilidad en rutinas o patrones de comportamiento verbal o no verbal ritualizados	- Gran angustia frente a cambios pequeños, dificultades con las transiciones, patrones de pensamiento rígidos, rituales de saludo, necesidad de tomar el mismo camino o de comer los mismos alimentos cada día.
3. Intereses muy restringidos y fijos que son anormales por su intensidad o foco	- Fuerte apego o preocupación por objetos inusuales, intereses excesivamente circunscritos o perseverantes
4. Hiper- o hiporreactividad a los estímulos sensoriales o interés inhabitual por aspectos sensoriales del entorno.	- Aparente indiferencia al dolor o a la temperatura, o respuestas adversas a sonidos o texturas específicas

C. Los síntomas deben estar presentes desde las fases iniciales del desarrollo, aunque pueden no expresarse claramente hasta que las demandas sociales superen las capacidades de la persona (pero pueden no manifestarse totalmente hasta que la demanda social supera las capacidades limitadas, o pueden estar enmascarados por estrategias aprendidas en fases posteriores de la vida).

D. Los síntomas causan un deterioro clínicamente significativo en lo social, laboral u otras áreas importantes del funcionamiento habitual.

E. Este trastorno no se explica mejor por una discapacidad intelectual (trastorno del desarrollo intelectual) o el retraso del desarrollo global. La discapacidad intelectual y el TEA frecuentemente ocurren conjuntamente; para hacer diagnósticos de comorbilidad del TEA y discapacidad intelectual, la comunicación social debe estar por debajo de lo esperado para el nivel de desarrollo general.

El manual DSM-5 también incluye criterios para determinar la gravedad (ver Tabla 2). La clasificación de la severidad expresa el deterioro que producen los síntomas del TEA y las consiguientes necesidades de apoyo de la persona. Sin embargo, los niveles de gravedad no son cuantificables y no deben usarse para indicar los servicios que deben proporcionarse a la persona (APA, 2013). Estos niveles son el reflejo del impacto de las dificultades en las actividades cotidianas.

Tabla 2. Niveles de gravedad del trastorno del espectro autista

Nivel de gravedad	Comunicación social	Comportamiento restrictivo y repetitivo
Grado 3 "Necesita ayuda muy notable"	Deficiencias graves en comunicación social, verbal y no verbal, que causan alteraciones graves del funcionamiento, inicio muy limitado de interacciones sociales y respuesta mínima a las aperturas sociales de otras personas.	Inflexibilidad del comportamiento, extrema dificultad para hacer frente a los cambios y comportamientos restringidos y repetitivos que interfieren notablemente con el funcionamiento en todos los ámbitos. Ansiedad intensa o dificultad para cambiar el foco o la acción.
Grado 2 '"Necesita ayuda notable"	Deficiencias notables en habilidades de comunicación social, verbal y no verbal; problemas sociales obvios incluso con ayuda *in situ*; inicio limitado de interacciones sociales, y respuestas reducidas o anormales a la apertura social de otras personas.	La inflexibilidad del comportamiento, la dificultad para hacer frente a los cambios y los comportamientos restringidos o repetitivos son evidentes para un observador casual e interfieren con el funcionamiento en diversos contextos.
Grado 1 "Necesita ayuda"	Sin ayuda *in situ* las deficiencias en comunicación social causan problemas importantes. Dificultad para iniciar interacciones sociales; respuestas atípicas o infructuosas a aperturas sociales de otros. Puede parecer que tiene poco interés en las interacciones sociales.	La inflexibilidad del comportamiento causa una interferencia significativa con el funcionamiento en uno o más contextos. Dificultad para alternar actividades. Los problemas de organización y de planificación dificultan la autonomía.

Los estudios epidemiológicos basados en datos administrativos y comunitarios sugieren que el autismo es un trastorno más común de lo que se penaba en los primeros

20 años desde su identificación por Kanner (1943). Se ha constatado que es más frecuente en hombres que en mujeres, con proporciones que oscilan entre 2:1 – 5:1. Estos resultados han estimulado a muchos investigadores a plantear que el TEA puede manifestarse de manera diferente, de forma más sutil, en las mujeres (Dworzynski et al., 2012; Glidden et al., 2016). Los estudios basados en búsqueda activa de casos han reportado la tendencia a una prevalencia similar en ambos sexos a medida que desciende el cociente intelectual de las personas, encontrándose el mismo número de hombres y mujeres cuando la discapacidad intelectual es moderada. Es posible que en los casos más leves y sin discapacidad intelectual asociada las mujeres sean más capaces de camuflar los síntomas y pasar así desapercibidos (Dean et al., 2017).

Las estimaciones de prevalencia de TEA en la población difieren en gran medida según los métodos de verificación empleados. Se estima que globalmente la prevalencia de TEA es del 1% (Fombonne et al., 2021). Diferentes estudios han reportado una escasa variación de prevalencia entre regiones, etnias y servicios (Baxter et al, 2014; Elsabbagh et al., 2012) y, aunque la prevalencia ha registrado un aumento en los últimos 20 años, se está produciendo una estabilización. Este incremento puede deberse a que en las dos últimas décadas han mejorado los sistemas de detección y diagnóstico, lo que permitiría la identificación de casos más leves y/o sin discapacidad intelectual.

La realidad actual es que los casos de menores con TEA crecen cada día. La sociedad es más sensible a este tipo de alteraciones, los profesionales tienen un nivel de especialización mayor, y los sistemas de detección temprana son más específicos para el TEA (Siu et al., 2016). Este aumento en la detección de casos, de forma cada vez más temprana, plantea una serie de problemas que a día de hoy la sociedad no ha podido resolver (Bejarano-Martín et al., 2020b). Si los casos aumentan, la respuesta sanitaria, educativa y social debe dar una respuesta eficaz. Sin embargo, las familias con menores

con TEA reportan mayores dificultades para acceder a los servicios, mayores costes asociados y falta de información y apoyo durante el proceso (Hodgetts, Zwaigenbaum y Nicholas, 2015; Kogan et al., 2008; Thomas, Parish, Rose y Kilany, 2012a; Wang, Mandell, Lawer, Cidav y Leslie, 2013).

Un estudio muy reciente (Crane et al., 2018) sobre las opiniones de familias, profesionales y adultos con autismo sobre el proceso de detección y diagnóstico encontró que existía una organización inadecuada de los programas de atención, que afectaba en gran medida a la satisfacción con los servicios. Las familias, profesionales y sociedad en general están reclamando más orientación, consejos y apoyo emocional que les ayuden a comprender el significado y las implicaciones del diagnóstico recibido, a fin de poder evitar crisis en la familia y manejar adecuadamente el estrés (Crane et al., 2018).

Los servicios de salud, educativos y sociales deben responder ante este aumento de casos y los crecientes niveles de insatisfacción de familias y profesionales ante dichos servicios. Si se detectan más casos, los servicios deben aumentar, y acompañar en el proceso una vez detectado el trastorno. La detección de TEA no tiene ningún sentido si esta no se acompaña de un programa de intervención temprana que aminore las dificultades (Siu et al., 2016). Las familias están demandando una respuesta eficaz ante este aumento de prevalencia. Reclaman participar más activamente en la evaluación de las necesidades del niño y de la familia, y que los profesionales adopten un enfoque proactivo para identificar tales necesidades (Bejarano-Martín et al., 2020b; Casagrande e Ingersoll, 2017). Las familias que informan que participan activamente en las decisiones y tienen una buena comunicación con los profesionales también informan una mayor satisfacción con los servicios, menos brechas en los servicios, menos demoras en el acceso al tratamiento y los servicios, menos estrés y menores costes generales

relacionados con el TEA (Burke y Goldman, 2015; Kuo, Bird, & Tilford, 2011; Moh y Magiati, 2012;).

ETIOLOGÍA, FACTORES DE RIESGO

Es imprescindible conocer cuáles son las causas y los factores de riesgo de desarrollo de TEA. Estudiar los factores relacionados con el TEA permite conocer mejor las manifestaciones asociadas tempranas que se producen, lo que redunda en una reducción en la edad del diagnóstico del trastorno, y por consiguiente en una reducción en la edad de acceso a programas de intervención (Salomone et al., 2015). El estudio de factores de riesgo como la prematuridad permiten conocer características y manifestaciones tempranas del autismo, que pueden hacerse extensibles al resto de la población. El objetivo es detectar con mayor eficacia aquellas manifestaciones que ya conocemos y a edades más bajas, además de la posibilidad de descubrir nuevas vías de acceso al trastorno. El TEA se explica principalmente por una suma de factores ambientales y genéticos. La investigación en curso continúa profundizando nuestra comprensión de los posibles mecanismos etiológicos en los TEA, pero actualmente no se ha dilucidado una sola causa unificadora.

Factores ambientales

Diversos estudios de revisiones y metaanálisis (Modabbernia et al., 2017) han puesto de manifiesto que algunos factores ambientales tienen riesgo potencial para el TEA. Un estudio de revisión (Wu et al., 2017) sobre la asociación entre edad parental avanzada y TEA encontró que por cada 10 años de aumento de la edad materna y paterna se incrementa el riesgo de TEA en la descendencia en un 18 y 21% respectivamente.

Además, la obesidad materna y la diabetes gestacional (Lyall et al., 2014), un intervalo corto entre embarazos (Zerbo et al., 2015), y el uso de valproato durante el embarazo (Gentile, 2014) se han asociado con un mayor riesgo de autismo. Se debe tener en cuenta que estos factores no son causales del TEA, pero sí podrían considerarse reactivos del trastorno. Estos factores podrían ser la base de riesgo de autismo, acompañados de mecanismos subyacentes como la genética o la epigenética (Lord et al., 2020).

Existen además diferentes estudios que han evaluado diferentes factores de riesgo, reportando una ausencia de asociación con el TEA. Factores como la vacunación, diferentes formas de parto (prolongado, por cesárea, asistido, con rotura de membrana, por reproducción asistida), no se asociaron con un mayor riesgo de TEA (Lord et al., 2020). Por último, el uso de suplementos de ácido fólico durante el embarazo se ha asociado con un menor riesgo de TEA (Schmidt et al., 2012).

Respecto al parto, el trauma o una lesión en el momento del nacimiento, gran prematuridad, complicaciones por un peso inferior a 1,500 kg al nacer y dificultades neonatales de alimentación se han asociado con un mayor riesgo 18. La prematuridad y/o bajo peso al nacer puede aumentar la vulnerabilidad del cerebro en desarrollo y, junto con otras exposiciones asociadas con el parto prematuro, pueden actuar como vías casuales para el TEA (Joseph et al., 2017). Existe una relación inversa entre meses de gestación y riesgo de TEA: a menor edad de gestación mayor riesgo de TEA (Hernandez-Fabian et al., 2018). Un metaanálisis reciente informó de una prevalencia de TEA del 7% en esta población (Agrawal et al., 2018), en comparación con la población general, en la que la prevalencia general de TEA es del 1% (Lord et al., 2020).

Los nacimientos prematuros son cada vez más generalizados, y el 10,6% de los nacimientos mundiales son prematuros, lo que representa 15 millones de bebés cada año en todo el mundo (Chawanpaiboon et al., 2019). Además, aquellos bebés que presentan

bajo peso suelen tener un parto prematuro (Savitz et al., 2000). Si aumentan los nacimientos prematuros, los casos de TEA aumentarán a su vez. Según la alta prevalencia encontrada en este grupo, este hecho se ha consolidado como uno de los factores de riesgo más importantes a tener en cuenta. La gran proporción de recién nacidos prematuros que posteriormente desarrollan TEA o que son seleccionados como en riesgo de TEA con los instrumentos existentes requiere una respuesta adecuada que mejore los criterios de sospecha y posibilite el diagnóstico precoz y el tratamiento oportuno de estos niños (Chernego et al., 2018). Sin embargo, hasta la fecha, ningún programa de intervención temprana ha estudiado su eficacia en niños prematuros que finalmente reciben un diagnóstico de TEA. Por otro lado, actualmente no hay evidencia que demuestre que este grupo sea comparable con los bebés a término con TEA, o incluso que las deficiencias de los bebés prematuros que muestran características de TEA puedan reducirse mediante un programa de intervención temprana.

Factores genéticos

En los últimos años se ha producido un avance tecnológico sustancial que ha permitido establecer asociaciones genéticas con el TEA. Los estudios realizados con gemelos y hermanos de menores con TEA han demostrado que los aspectos genéticos contribuyen de forma significativa al autismo, calculándose una heredabilidad que ronda entre el 40% y el 90% (Gaugler et al., 2014). Parece que el TEA se encuentra entre las alteraciones neuroevolutivas más heredables (Wang et al., 2017). Hasta la fecha, los estudios han identificado más de 100 genes y regiones genómicas que se han asociado al autismo (Sanders et al., 2015), basándose principalmente en el análisis de mutaciones heterocigotas, de novo (es decir, aparecen en el niño, pero no en los padres). Estos cambios tienen un efecto funcional en las regiones del genoma que codifican proteínas

presentando una asociación más sólida y fiable con el riesgo de autismo (Bourgeron, 2015).

Aunque hay una sólida evidencia de que el TEA tiene una carga genética, el trastorno no se puede explicar única y exclusivamente desde esta perspectiva, teniendo en cuenta que en la población general la contribución de las mutaciones de novo al riesgo de autismo es pequeña (en torno al 3%) (Gaugler et al., 2014), y muchos de estos nunca llegarían a desarrollar síntomas. La aparición del trastorno parece depender también de las interacciones entre los genes y el medio ambiente. Los mecanismos epigenéticos que establecen la relación entre las influencias genéticas y ambientales son procesos relevantes en el desarrollo de la persona y pueden verse afectados por los factores ambientales vistos anteriormente, aumentado así el riesgo de TEA (Meek et al., 2013).

Los aspectos genéticos del trastorno son importantes factores de riesgo. Sin embargo, representan una pequeña parte de la explicación del TEA. Si los genes fueran los únicos capaces de explicar el comportamiento de las personas, por ejemplo, los gemelos serían idénticos no solo físicamente si se han criado en un mismo entorno (Iacono y McGue, 2002). Por consiguiente, aunque un rasgo sea altamente heredable, las influencias del entorno pueden ser importantes a la hora de que los efectos de los genes medien en la conducta. Los factores ambientales juegan un papel muy importante a la hora de explicar los rasgos morfológicos y psicológicos (Hustvedt, 2017). Un hecho altamente contrastado es que el TEA no aparece debido a una única causa, si no a la suma de factores que van incrementando el riesgo de desarrollar el trastorno (Hodges et al., 2020; Lord et al., 2020). En este sentido, el estudio de grupos de riesgo en base a factores ambientales como la prematuridad incrementará el conocimiento que se tiene a día de hoy sobre las causas del TEA.

CARACTERÍSTICAS Y DESARROLLO DE MENORES CON TEA

Los síntomas de TEA se reconocen durante el segundo año de vida (Hodges et al., 2020), aunque hay algunos indicadores que se pueden observar en torno a los 12 meses en algunos casos (APA, 2013). Las características conductuales del trastorno empiezan a ser evidentes en la primera infancia, presentando dificultades de interacción social y comunicación durante el primer año de vida. Estas dificultades durante la primera infancia son propias del TEA, y raramente aparecen en otros trastornos (APA, 2013). Por tanto, el estudio de estos rasgos que aparecen de forma temprana es imprescindible para entender cómo se desarrolla el TEA, para así intervenir de una manera adecuada en consecuencia. Entender cómo aparece y se desarrolla el TEA implica comprender su origen y, por tanto, encontrar soluciones terapéuticas más eficaces.

En los últimos años se está produciendo un avance notable en el descenso de la edad de identificación de menores con riesgo de trastorno del espectro autista (TEA) (Mazurek et al., 2014). Esta reducción en la edad se ha logrado gracias a la generalización de nuevas experiencias de detección precoz y también a que los servicios de atención a la infancia son ahora más sensibles hacia los signos precoces de TEA. También han proliferado los estudios de seguimiento de grupos de riesgo, por ejemplo, de hermanos de niños con TEA o de niños con nacimiento prematuro, que están permitiendo identificar en niños menores de 2 años alteraciones muy tempranas en su desarrollo comunicativo y social.

Es antes de cumplir los dos primeros años de vida cuando el bebé desarrolla las habilidades básicas para establecer relaciones de reciprocidad. El concepto de reciprocidad hace referencia a la correspondencia mutua entre el bebé y sus progenitores y es el marco en el que se desarrolla la comunicación social, generalmente afectada en los niños pequeños con TEA. En ese marco de reciprocidad se integra el uso social de la mirada, los actos de atención conjunta, como por ejemplo la conducta de señalar con

función declarativa, el uso de gestos y vocalizaciones, la imitación y el juego (Charman et al., 2003; Landa, Holman, & Garrett-Mayer, 2007; Ozonoff et al., 2008 Yoder, Stone, Walden, & Malesa, 2009). También, los comportamientos motores repetitivos y el uso peculiar de objetos se ha encontrado que puede ser indicador de riesgo en el segundo año de vida (Ozonoff et al., 2008; Watt, Wetherby, Barber, & Morgan, 2008). También se han identificado otras alteraciones tempranas en otros dominios del desarrollo relevantes para el autismo, como son las respuestas afectivas, la sonrisa recíproca, la reactividad comportamental y el desarrollo sensoriomotor (Bryson et al., 2007).

La imitación juega un papel fundamental en el desarrollo de las habilidades sociales, las funciones comunicativas socioemocionales y la teoría de la mente, así como las habilidades de lenguaje y juego (Contaldo, Colombi, Narzisi y Muratori, 2016; Cooley, 2017; Dohmen, Bishop, Chiat y Roy, 2016). La imitación promueve una orientación social hacia los demás (Contaldo et al., 2016), y los niños mejoran la atención a los adultos en lugar de los objetos, y sonríen más a medida que realizan sus acciones. Reconocen este "juego de imitación" desde los 9 meses de edad (Agnetta y Rochat, 2004).

El contacto visual, que se ve afectado en niños con TEA (Jones y Klin, 2013), es un componente esencial de las relaciones sociales diarias y proporciona la base para el desarrollo de habilidades más complejas, como el compromiso social (Franchini, Glaser, Wood, Gentaz, Eliez & Schaer, 2017). Los niños de 1 a 2 meses de edad que luego fueron diagnosticados con TEA mostraron un nivel de contacto visual equivalente a los niños con desarrollo típico, disminuyendo la frecuencia a medida que crecían (Jones y Klin, 2013). Esta disminución en el contacto visual, en lugar de una ausencia, ofrece una oportunidad prometedora para la intervención temprana.

La atención conjunta es otro déficit significativo en niños con TEA (Mundy, 2018). La atención conjunta se correlaciona con el desarrollo del lenguaje y otras habilidades de

comunicación social (p. Ej., Juego, imitación) (Adamson, Bakeman, Suma & Robins, 2019; Bottema-Beutel, 2016; Kasari et al., 2015; Pickard & Ingersoll, 2015). Esta sólida correlación mejora muchos aspectos del desarrollo del lenguaje, como el lenguaje expresivo y receptivo, el vocabulario y los gestos. Más importante aún, al enseñar habilidades de atención conjunta a niños con TEA, podrían aprender a prestar más atención al comportamiento social de los demás e incorporar a su repertorio el uso de habilidades sociales como gestos, vocalizaciones o expresiones emocionales, lo que promueve su participación social en rutinas diarias (Meindl y Cannella-Malone 2011). De esta manera, los niños con TEA comenzarían a tener en cuenta a los demás y su compromiso social aumentaría al poder compartir la atención sobre objetos o eventos y personas.

En relación a los gestos, los niños con TEA tienen dificultades para usar estos para reconocer un objeto o sus propiedades (Gordon y Watson, 2015; Mastrogiuseppe, Capirci, Cuva y Venuti, 2015). Estas habilidades de comunicación no verbal son la base para el desarrollo de habilidades como atención conjunta (señalar), imitación, juego y lenguaje (Ingersoll y Lalonde, 2010; Özçaliskan, Adamson y Dimitrova, 2016; Paparella y Freeman, 2015).

Los niños con TEA encuentran muchas dificultades en el juego, especialmente cuando se trata de realizar secuencias de juego simbólico y simulado (Wilson et al., 2017). Las habilidades de juego son predictores importantes de las habilidades de lenguaje, sociales y de comunicación posteriores (Kasari, Freeman y Paparella, 2008) y del desarrollo de la teoría de la mente (Lin, Tsai, Li, Huang y Chen, 2017). Además, el juego tiene una relación recíproca con el desarrollo de habilidades como el lenguaje. Los niños que mejoran sus habilidades de juego también mejoran el desarrollo de otras habilidades (Pierucci et al., 2015).

En la Tabla 3 se presenta un resumen de los signos de alarma más relevantes comúnmente presentes en la mayor parte de los instrumentos desarrollados para la detección precoz.

Tabla 3. Signos precoces de alarma de TEA más relevantes

	A partir de los:		
Conducta	**12 meses**	**18 meses**	**24 meses**
Social/ Emocional	- Escaso contacto ocular - Escasez de sonrisa - No orientación al nombre - No sigue la mirada de otros - Iniciativa social pobre - Escasa expresión facial - Poca regulación emocional - Escasez de posturas anticipatorias	- Aversión a la mirada - Escasa expresión emocional - No respuesta al nombre - Menos cambios de atención entre objetos y personas - Poca atención a gestos y/o cambios de atención de otros - Falta de imitación - Poca atención al malestar de otros	- Falta de interés social y en otros niños - Contacto ocular muy breve - Poca variedad de expresiones afectivas - No ofrece consuelo
Comunicativa/ Simbólica	- Poca frecuencia de vocalizaciones - Escasa respuesta a la atención conjunta - Escasez de actos de señalar - Ausencia de actos de mostrar - Retraso en el balbuceo	- No señala para pedir - Pocas respuestas e inicios de atención conjunta - Pocas consonantes comunicativas - Pocos gestos y/o poco variados - Retraso en lenguaje receptivo y expresivo - Poco juego y poco variado	- Pocas respuestas a la atención conjunta - Poca integración de mirada y comunicación - No busca a otros para compartir intereses - Pocos gestos - Escaso vocabulario - Prosodia atípica
Atencional/ Sensoriomotora	- Movimientos poco variados/atípicos - Déficit en el desenganche atencional - Hipotonía - Anormalidades de activación y en respuestas sensoriales - Escasa coordinación - Pasividad y escasa conducta exploratoria - Patrón atencional anormal	- Conductas estereotipadas	- Conductas repetitivas e intereses restrictivos

Adaptada de Canal et al. (2013)

El autismo sigue planteando un gran número de preguntas sobre cómo empieza a manifestarse, cómo evitar su aparición o cómo reconducir el desvío que inicialmente se

produce. Aun así, uno de los hechos más aceptados es que la detección de estas manifestaciones tempranas ayuda a iniciar programas de intervención precoces, que tienen efectos muy positivos sobre el pronóstico (Dawson, 2008; Dawson et al., 2010; Reichow, et al., 2012). Los estudios constatan beneficios sustanciales de la atención temprana para el desarrollo cognitivo, del lenguaje y de la conducta adaptativa, en comparación con los resultados logrados para menores que recibieron una intervención más tardía (Bejarano-Martín et al., 2020a). Por eso, se considera fundamental detectar los casos tan pronto como sea posible, incrementando así las oportunidades de los menores con TEA para alcanzar mejores niveles adaptativos (Zwaigenbaum et al., 2015). La atención temprana puede evitar el agravamiento del problema, así como la aparición de síntomas secundarios del trastorno y minimizar algunos síntomas, como los intereses restrictivos, la insistencia en rutinas, o las dificultades graves de socialización (Landa, 2018; Rogers & Vismara, 2014). Además, si se inicia la intervención antes de que el niño pierda totalmente sus habilidades comunicativas y sociales la eficacia de la atención temprana es mucho mayor (Barbaro y Dissanayake, 2010). Las mejoras que se logren por efecto de la atención temprana reducirán también la carga de enfermedad y mejorarán la calidad de vida de los afectados y de sus familias, pudiendo reducir los costes sociales y económicos que las familias de las personas con TEA han de afrontar a lo largo de toda la vida (Rogers et al., 2016). Por tanto, se considera muy relevante el progreso en el conocimiento sobre cómo detectar lo más pronto posible el riesgo de TEA.

Si los programas de detección identifican retrasos o desviaciones en habilidades como contacto visual, juego, imitación o atención conjunta, es posible implementar programas dirigidos a esas habilidades, involucrando activamente a las familias para que comiencen a trabajar con sus hijos/as, incluso antes de que reciban un diagnóstico formal de TEA. Esto haría que los programas de detección temprana fueran más útiles y

socialmente válidos. Los programas de intervención aumentarían así el éxito de los programas de detección y reducirán los síntomas del TEA (MacDonald, Parry-Cruwys, Dupere y Ahearn, 2014; Orinstein et al., 2014).

El nivel y el tipo de manifestaciones tempranas de los menores con TEA pueden cambiar el efecto de la eficacia de los programas de intervención. Los estudios informan diferencias significativas en las habilidades de comunicación social en aquellos casos en los que la capacidad no verbal de los participantes es mayor antes de empezar la intervención (Virués-Ortega, 2010). La mejora de dichas habilidades parece ser un predictor de una reducción en la gravedad de los TEA y un aumento en las habilidades de adaptación (Zachor y Ben-Itzchak, 2017). Esto significa que se debe estudiar el papel de las habilidades socio-comunicativas, con el fin de recomendar programas de intervención adecuados que tengan en cuenta estas características en los participantes. Si la implementación de programas de intervención temprana se basa en una cantidad suficiente de información sobre las manifestaciones tempranas de cada participante, los resultados serán más efectivos (Bejarano-Martín et al., 2020a). Asimismo, los profesionales podrían seleccionar con mayor precisión los procedimientos potencialmente más efectivos y factibles.

NECESIDADES DE APOYO A MENORES CON TEA Y SUS FAMILIAS

Las necesidades de apoyo a menores con TEA pasan por la implementación de programas eficaces de detección e intervención de forma temprana. Como se ha comentado, la reducción de la edad y mejora de los sistemas de detección han hecho posible detectar signos de riesgo en niños de 12 a 14 meses (Jones et al., 2014). Como resultado, la demanda de estrategias efectivas diseñadas para ser implementadas en programas de intervención temprana está aumentando (Zwaigenbaum et al., 2015). El

objetivo de la detección temprana es identificar signos de riesgo o síntomas tempranos de TEA en el desarrollo del niño, con el fin de iniciar un tratamiento más temprano, incluso antes de que el niño reciba un diagnóstico formal. Por lo tanto, los menores con TEA y sus familias no solo necesitan programas de detección temprana, necesitan además tratamientos tempranos que den sentido a la detección y que hayan demostrado su eficacia.

Las acciones sistemáticas de detección temprana asociadas con el inicio de actividades de intervención están vinculadas a resultados positivos para los niños en riesgo de TEA, ya que los menores que comienzan el tratamiento antes tienen mejores resultados (MacDonald, Parry-Cruwys, Dupere y Ahearn, 2014; Orinstein et al., 2014). Por lo tanto, el objetivo final es que la intervención se produzca durante el período crítico de desarrollo del niño, cuando la plasticidad neuronal es mayor y se pueden lograr resultados positivos a largo plazo (Crais y Watson, 2014). Además, el inicio de actividades de intervención temprana también podría tener consecuencias positivas para la familia, que puede encontrar respuestas a sus inquietudes y aprender estrategias para hacer frente a las dificultades de desarrollo de su hijo (Ingersoll, Shannon, Berger, Pickard y Holtz, 2017; Kasari et al., 2015).

Si atendemos a las manifestaciones tempranas, la comunicación e interacción social constituye una de las principales áreas de dificultad de los menores con TEA, por lo que las necesidades en esta área son múltiples, y en muchas ocasiones complejas de atender (APA, 2013). Los menores con TEA necesitan disponer de diferentes recursos y destrezas para la participación social, la comunicación y el juego. Hacer uso de habilidades como iniciar actos comunicativos y responder con espontaneidad a las comunicaciones de otros suponen dificultades en este grupo. Por tanto, requieren apoyos para guiarles en los inicios sociales antes de actuar.

Los menores con mayor afectación o síntomas más graves muestran un perfil de inicios sociales que incluyen principalmente interacciones positivas pero muy básicas, breves, con pocas conductas de búsqueda de atención y algunas conductas de evitación y de rechazo (APA, 2013). Si se les deja solos con frecuencia se centran en comportamientos ritualizados; por su parte lo niños con TEA de mejor funcionamiento suelen tener conductas de interacción social, pero muestran principalmente comportamientos sociales pasivos con bajo nivel de implicación como el contacto visual a cierta distancia, no combinado con una sonrisa (Bauminger-Zviely, 2014). Las interacciones sociales de los menores con TEA no solo son menos frecuentes, sino que además son percibidas por los demás como cualitativamente diferentes. En el apoyo a estas dificultades se debe prestar atención especial a las aproximaciones sociales menos eficientes de los menores con TEA que son ignoradas por sus compañeros, porque el fracaso en los intentos de aproximación por parte del menor con TEA puede llevarle a la creencia de errónea de que no es correspondido o que es rechazado por sus compañeros. Además, algunos de estos comportamientos sociales pueden ser inapropiados (porque usas frases aprendidas de memoria (ecolalias) o comportamientos repetitivos para aproximarse a otros).

Los menores con TEA encuentran muchas dificultades para leer las señales del entorno, tanto las indicaciones sociales de otros (gestos, expresiones, sonidos) como del medio físico. Si el niño es capaz de captar, por sí mismo o por medio de apoyos, estas señales y de establecer intercambios recíprocos con los otros, tendrá la capacidad de, por ejemplo, cambiar de una actividad a otra de manera flexible, de participar activamente en juegos y aprendizajes, o de establecer interacciones de ida y vuelta. Sería indicativo de que dispone de recursos básicos para la reciprocidad social y para la comunicación efectiva con los demás que facilitarán el desarrollo de las habilidades conversacionales,

por lo que el apoyo para lograr al máximo nivel de reciprocidad es un elemento necesario en los programas de intervención.

Por último, debemos tener en cuenta las dificultades de lenguaje que puede tener un menor con TEA. Las necesidades que se derivan de esta dificultad pasan por el nivel de expresión y comprensión lingüísticas que tenga. Para que la comunicación sea eficaz, el lenguaje que usen todas las personas de su entorno tiene que adecuarse a su nivel comprensivo y expresivo. En términos de expresión, sus necesidades serán similares, el nivel de comunicación que muestre será el que se le exija, pero necesitará de nuestra ayuda para que progrese en su habilidad expresiva. En este sentido, si el niño habla con palabras, nosotros expandiremos sus palabras sueltas en frases sencillas, indicando comprensión y atendiendo al mensaje que el niño trata de comunicar. Debemos tener en cuenta además otros elementos de la comunicación, como hacer que nuestro lenguaje sea redundante, concreto, algo repetitivo y que se centre, al menos en los primeros momentos del desarrollo, en los centros de interés del niño.

Familias y profesionales también demandan necesidades relativas a los servicios y procesos de detección e intervención. Como se ha comentado, la participación activa de los padres aumenta la satisfacción familiar con los servicios. Diversos estudios muestran que la participación de los padres es fundamental para la satisfacción con los programas de intervención (McIntyre & Zemantic, 2017; Stadnick, Drahota & Brookman-Frazee, 2013). En los últimos años, también se ha demostrado que la participación activa no solo aumenta la satisfacción del servicio, sino que también mejora los resultados de la intervención, por ejemplo, aumentando el progreso en la adquisición de habilidades (Ingersoll y Wainer, 2013; Kasari, Gulsrud, Paparella, Hellemann, y Berry, 2015; Pickles et al., 2016). Además, involucrar a los padres reduce los costes de los programas de intervención al disminuir el número de horas con profesionales y aumentar el desarrollo

de habilidades en contextos naturales (Ingersoll, Shannon, Berger, Pickard y Holtz, 2017; Pickles et al., 2016). Todos estos factores significan que la participación de los padres en las intervenciones reduce la carga económica sobre la familia, el sistema de salud y la sociedad, junto con el estrés asociado con tener un hijo con TEA (Kasari et al., 2015).

INTERVENCIÓN TEMPRANA

Como se ha comentado, los sistemas de detección y la observación de profesionales y familiares, en la mayoría de los casos, están alertando de las primeras preocupaciones de un problema en el desarrollo del menor en torno a los 18 meses (Bejarano-Martín et al., 2020b). Debido a esta reducción y a la implementación de programas de dirigidos la detección precoz de este trastorno, se hacen necesarios programas de intervención cuyo objetivo sea reducir las dificultades asociadas, incluso antes de recibir un diagnóstico formal de TEA que, en muchos casos, tarda en llegar hasta 6 meses o más (Bejarano-Martín et al., 2020b), debido a barreras socioeconómicas o por presentar una menor severidad en los síntomas o un nivel de funcionamiento intelectual alto (Mazurek et al., 2014). El informe de la Task Force (Siu et al., 2016) aboga por la implementación de programas que complementen la detección temprana, ya que de otra manera esta perdería el sentido para el que fue creada. Por ello, la intervención debe realizarse de la forma más temprana que sea posible, y siempre unida al proceso de detección de signos de alarma de TEA.

Las intervenciones que focalizan en las habilidades sociales y comunicativas se han convertido en el eje central de la mayoría de las terapias en menores con TEA, ya que la demanda de este tipo de intervenciones ha crecido para esta población (Wang, Parrila and Cui, 2013. Los déficits en las habilidades comunicativas y sociales se han reconocido como uno de los problemas clave en menores con TEA desde los primeros casos

reportados por Kanner (1949). Por tanto, los modelos de intervención deben ayudar a los niños con TEA a incrementar sus habilidades sociales y comunicativas y reducir el mayor número de síntomas de esta área.

El objetivo de la intervención temprana es anticiparse a las deficiencias para iniciar un tratamiento más temprano, incluso antes de recibir un diagnóstico formal. Este es un momento crítico, cuando el cerebro aún se está desarrollando y los síntomas del TEA están emergiendo (Crais y Watson, 2014). La intervención temprana mejora las habilidades sociales y comunicativas, el desarrollo cognitivo, el lenguaje y la conducta adaptativa (Bejarano-Martín et al., 2020a; Gates, Kang & Lerner, 2017). También puede reducir la sintomatología del TEA, así como los síntomas secundarios (Reichow, Barton, Boyd & Hume, 2012). La intervención temprana también reducirá la carga de TEA y puede mejorar la calidad de vida de los menores y sus familias, reduciendo los costes sociales y económicos que las familias y los servicios tienen que enfrentar a lo largo de toda la vida (Bejarano-Martín, 2020b; Salomone et al., 2016; Fletcher-Watson et al., 2017).

En los últimos años se han desarrollado diferentes programas de intervención temprana. Un grupo de revisiones (Bradshaw, Steiner, Gengoux, & Koegel, 2015; Odom, Boyd, Hall, & Hume, 2010; Reichow, Barton, Boyd, & Hume, 2012; Virués-Ortega, 2010; Waddington, Meer, & Sigafoos, 2016) han seleccionado estudios sobre diferentes procedimientos, organizados para intervenir en una amplia variedad de dificultades, todas generalmente presentes en menores con TEA, con el objetivo de lograr un aprendizaje amplio y un impacto global en los déficits centrales del trastorno. Revisiones recientes (Ryberg, 2015; Waddington et al., 2016) y ensayos controlados aleatorios (ECA) como los de Estes et al. (2015) y Pickles et al. (2016) indican además que estos tratamientos aplicados de forma temprana logran resultados favorables y estables en la reducción de

síntomas de TEA. Además, aspectos como la participación activa de los padres, la intensidad y duración de la intervención, o selección de contenidos concretos relativos a las dificultades de los niños con TEA, son características a tener en cuenta para lograr mayor eficacia en los programas de intervención (Bradshaw et al., 2015; Debodinance, Maljaars, Noens, & Van den Noortgate, 2017; McConachie & Diggle, 2007; Reichow, 2012).

Las intervenciones sociales y comunicativas han sido reconocidas como uno de los programas más prometedores en TEA (Watkins, Kuhn, Ledbetter-Cho, Gevarter y O'Reilly, 2017; Zwaigenbaum et al., 2015). Además, las habilidades de comunicación social son clave para el desarrollo de habilidades complejas, como el lenguaje (Bradshaw, Koegel y Koegel, 2017; Hampton y Kaiser, 2016). En consecuencia, la intervención temprana debería ayudar a los niños en riesgo de TEA a aumentar sus habilidades de comunicación social y reducir los síntomas en esta área (Gates et al., 2017; Murza, Schwartz, Hahs-Vaughn & Nye, 2016; Schreibman et al., 2015 ; Zwaigenbaum et al., 2015).

La identificación y selección de contenidos específicos de la intervención como ingredientes activos del tratamiento es muy interesante desde un punto de vista clínico y puede ser clave, ya que la selección del contenido de intervención es una cuestión de gran importancia cuando se consideran las dificultades principales en los TEA. Además, debido a las diferencias individuales entre los niños pequeños con riesgo o con TEA, la elección de una estrategia de intervención integral, dirigida a una variedad amplia de aspectos, podría no responder a las necesidades reales de cada caso. Así, decidir qué elementos del modelo integral de intervención sería necesario descartar por no ser adecuados para un cierto menor es una tarea compleja. Ese esfuerzo de configuración del modelo integral a las características específicas de cada individuo con riesgo o con TEA

podría modificar la aplicación del modelo. Por estas razones, para determinados casos, sería más sencillo desarrollar una estrategia de intervención basada en la selección de procedimientos focalizados en las dificultades concretas detectadas (imitación, atención conjunta, juego, etc.), con objetivos más individualizados (Wong, Huertas-Ceballos, Cowan, & Modi, 2014), para ser desarrollados en un periodo de tiempo relativamente corto, en comparación con los programas integrales, hasta que el niño con riesgo o con TEA haya desarrollado la habilidad (Odom et al., 2010). La aplicación de intervenciones focalizadas en dificultades específicas también podría proporcionar una evaluación clara del proceso y de los resultados del aprendizaje, utilizando herramientas particularmente diseñadas para medir el progreso en la habilidad objeto de intervención.

Los programas actuales se dirigen a un conjunto de habilidades muy extensas, cuyo objetivo es la mejora integral del menor, que en muchos casos llega a durar un mínimo de un año, con unos costes asociados muy altos (Wang et al., 2013). Por otro lado, las prácticas de intervención focalizada son prácticas o estrategias de instrucción individuales que se utilizan para enseñar habilidades específicas a niños con TEA en un período de tiempo relativamente corto (p. Ej., 0-3 meses) (Odom, Boyd, Hall, Hume, 2010a; Odom, Klinbergerg , Rogers y Hatton, 2010b; Wong et al., 2015). Se utilizan con frecuencia para mejorar las habilidades sociales y de comunicación y se han convertido en el foco de la mayoría de las terapias para niños pequeños (6 años o menos) con TEA para satisfacer las demandas de la sociedad (Schreibman et al., 2015; Wang et al., 2013; Zwaigenbaum et al. al., 2015). Las estrategias de comportamiento, las intervenciones naturalistas, los sistemas de comunicación de intercambio de imágenes, el entrenamiento de respuesta fundamental y los apoyos visuales son todos ejemplos de estas prácticas (Boyd, Odom, Humphreys y Sam, 2010). En los últimos años, se han realizado revisiones de investigaciones empíricas que cubren estudios de diseño de casos individuales y grupales

de prácticas focalizadas destinadas a mejorar las habilidades sociales y de comunicación (Bradshaw, et al., 2015; French & Kennedy, 2018; Reichow et al., 2012 ; Waddington et al., 2016). Sin embargo, estas revisiones no siempre incluyen una evaluación de la calidad del estudio. A pesar del uso extensivo de las prácticas focalizadas, se han realizado pocas investigaciones para examinar su eficacia general (Kasari, Shire, Factor y McCracken, 2014).

Los principales modelos de intervención que han mostrado mayores niveles de evidencia científica son los denominados "intervenciones conductuales naturalistas del desarrollo" (Bejarano-Martín et al. 2020a), aunque existe cierta incertidumbre y desacuerdo para evaluar qué niño debe recibir qué tratamiento o la intensidad del mismo. Este vacío deja a los padres y a los profesionales a merced de lo que está disponible y a veces se comercializa en su región. De hecho, el acceso a los servicios de intervención temprana es variable en la mayoría de las comunidades, incluso en los países de ingresos altos, y en su mayoría son llevados a cabo por personas no especializadas supervisadas por profesionales especialmente formados (Bejarano-Martín et al., 2020b).

Es esencial que los programas de intervención temprana en menores con autismo se basen en la evidencia científica de su efectividad. Este requisito es particularmente importante no solo para menores con autismo, también para sus familias. Muchos proveedores de programas de "tratamiento" han afirmado que sus programas o prácticas pueden mejorar la vida de los niños con autismo o incluso sugerir que tienen una cura (Siri & Lyons, 2014). Aunque hay mucha discusión sobre terminología y aplicación (McGrew et al., 2016), hay poco desacuerdo sobre la importancia de seleccionar y utilizar intervenciones que tengan evidencia empírica de eficacia. El grupo de la National Clearinghouse on Autism Evidence and Practice Review Team ha realizado una revision de la literatura desde 1990 hasta 2017, donde han seleccionado diferentes prácticas

basadas en la evidencia científica que han demostrado ser eficaces (Ver Tabla 4). Los programas de intervención temprana deben basarse en uno o varios de estos principios que han confirmado su eficacia a través de diferentes estudios (Steinbrenner et al., 2020).

Tabla 4. Prácticas basadas en la evidencia científica

		Apoyo empírico		
Práctica basada en la evidencia científica	**Definición**	**1990-2011 (n)**	**2012-2017 (n)**	**1990-2017 (n)**
Intervenciones basadas en antecedentes (ABI)	Arreglo de eventos o circunstancias que preceden a una actividad o demanda con el fin de aumentar la ocurrencia de un comportamiento o llevar a la reducción de los comportamientos desafiantes / interferentes.	29	20	**49**
Comunicación aumentativa y alternativa (AAC)	Intervenciones que utilizan y / o enseñan el uso de un sistema de comunicación que no es verbal / vocal que puede ser asistido (por ejemplo, dispositivo, libro de comunicación) o sin ayuda (por ejemplo, lenguaje de señas)	9	35	**44**
Intervención de movilización conductual (BMI)	La organización de las expectativas de comportamiento en una secuencia en la que las respuestas de baja probabilidad, o las más difíciles, están integradas en una serie de respuestas de alta probabilidad, o de menor esfuerzo, para aumentar la persistencia y la ocurrencia de respuestas de baja probabilidad.	8	4	**12**
Estrategias cognitivas conductuales / instruccionales (CBIS)	Instrucción sobre el manejo o control de procesos cognitivos que conducen a cambios en el comportamiento social, académico o conductual.	7	43	**50**
Refuerzo diferencial de comportamientos alternativos, incompatibles u otros (DR)	Un proceso sistemático que aumenta el comportamiento deseable o la ausencia de un comportamiento indeseable al proporcionar consecuencias positivas para la demostración / no demostración de tal comportamiento. Estas consecuencias se pueden proporcionar cuando el alumno: a) se involucra en un comportamiento deseado específico que no sea el comportamiento indeseable (DRA), b) participar en un comportamiento que es físicamente imposible de hacer mientras exhibe el comportamiento indeseable (DRI), o c) no participar en el comportamiento indeseable (DRO).	27	31	**58**
Instrucción directa (DI)	Un enfoque sistemático de la enseñanza utilizando un paquete de instrucción secuenciado con protocolos o lecciones con guiones. Enfatiza el diálogo entre el maestro y el estudiante a través de respuestas corales e independientes de los estudiantes y emplea correcciones de errores sistemáticas y explícitas para promover el dominio y la generalización.	2	6	**8**
Entrenamiento de prueba discreto (DTT)	Enfoque de instrucción con ensayos masivos o repetidos, cada uno de los cuales consta de la instrucción / presentación del maestro, la respuesta del niño, una consecuencia cuidadosamente planificada y una pausa antes de presentar la siguiente instrucción.	16	22	**38**
Ejercicio y movimiento (EXM)	Intervenciones que utilizan esfuerzo físico, habilidades / técnicas motoras específicas o movimiento consciente para enfocarse en una variedad de habilidades y comportamientos.	6	11	**17**

Extinción (EXT)	La eliminación de las consecuencias reforzantes de un comportamiento desafiante para reducir la aparición futura de ese comportamiento.	13	12	**25**
Evaluación funcional del comportamiento (FBA)	Una forma sistemática de determinar la función o el propósito subyacente de un comportamiento para que se pueda desarrollar un plan de intervención eficaz.	11	10	**21**
Entrenamiento en comunicación funcional (FCT)	Un conjunto de prácticas que reemplazan un comportamiento desafiante que tiene una función de comunicación con comportamientos o habilidades de comunicación más apropiados y efectivos.	12	19	**31**
Modelado (MD)	Demostración de un comportamiento objetivo deseado que resulta en el uso del comportamiento por parte del alumno y que conduce a la adquisición del comportamiento objetivo.	10	18	**28**
Intervención mediada por música (MMI)	Intervención que incorpora canciones, entonación melódica y / o ritmo para apoyar el aprendizaje o el desempeño de habilidades / comportamientos. Incluye musicoterapia, así como otras intervenciones que incorporan música para abordar las habilidades específicas.	3	4	**7**
Intervención naturalista (NI)	Una colección de técnicas y estrategias que están integradas en actividades y / o rutinas típicas en las que el alumno participa para promover, apoyar y alentar naturalmente las habilidades / comportamientos objetivo.	26	49	**75**
Intervención implementada por los padres (PII)	Entrega por parte de los padres de una intervención a su hijo que promueva su comunicación social u otras habilidades o disminuya su comportamiento desafiante.	13	42	**55**
Instrucción e intervención basadas en pares (PBII)	Intervención en la que los compañeros promueven directamente las interacciones sociales de los niños autistas y / u otras metas de aprendizaje individuales, o el maestro / otro adulto organiza el contexto social (p. Ej., Grupos de juego, grupos de redes sociales, recreo) y, cuando es necesario, proporciona apoyo (p. Ej., Indicaciones, refuerzo) a los niños autistas y sus compañeros para participar en interacciones sociales.	19	25	**44**
Indicaciones (PP)	Asistencia verbal, gestual o física que se brinda a los alumnos para ayudarlos a adquirir o participar en un comportamiento o habilidad específicos.	55	85	**140**
Refuerzos (R)	La aplicación de una consecuencia después del uso por parte de un alumno de una respuesta o habilidades que aumenta la probabilidad de que el alumno utilice la respuesta / habilidades en el futuro.	53	53	**106**
Interrupción de respuesta/ Redirección (RIR)	La introducción de una indicación, comentario u otros distractores cuando se está produciendo un comportamiento interferente diseñado para desviar la atención del alumno se aleja de la conducta que interfiere y da como resultado su reducción	13	16	**29**
Autogestión (SM)	La instrucción se centra en que los alumnos discriminen entre comportamientos apropiados e inapropiados, controlen y registren con precisión sus propios comportamientos y se recompensen por comportarse de manera adecuada.	14	12	**26**
Integración sensorial® (SI)	Intervenciones que se dirigen a la capacidad de una persona para integrar información sensorial (visual, auditiva, táctil, propioceptiva y vestibular) de su cuerpo y entorno para responder utilizando un comportamiento organizado y adaptativo.	1	2	**3**

Narrativas sociales (SN)	Intervenciones que describen situaciones sociales para resaltar características relevantes de una conducta o habilidad objetivo y ofrecen ejemplos de respuestas apropiadas.	15	6	**21**
Entrenamiento en habilidades sociales (SST)	Instrucción grupal o individual diseñada para enseñar a los alumnos formas de participar de manera adecuada y exitosa en sus interacciones con los demás.	18	56	**74**
Análisis de tareas (TA)	Un proceso en el que una actividad o comportamiento se divide en pasos pequeños y manejables para evaluar y enseñar la habilidad. Otras prácticas, como el refuerzo, el modelado de video o el retardo de tiempo, se utilizan a menudo para facilitar la adquisición de los pasos más pequeños.	9	4	**13**
Instrucción e intervención asistida por tecnología (TAII)	Instrucción o intervención en la que la tecnología es la característica central y la tecnología está específicamente diseñada o empleada para apoyar el aprendizaje o desempeño de un comportamiento o habilidad para el alumno.	10	30	**40**
Tiempo de espera (TD)	Una práctica que se utiliza para desvanecer sistemáticamente el uso de indicaciones durante las actividades de instrucción mediante un breve retraso entre la instrucción inicial y las instrucciones o indicaciones adicionales.	16	15	**31**
Video Modelado (VM)	Una práctica que se utiliza para desvanecer sistemáticamente el uso de indicaciones durante las actividades de instrucción mediante un breve retraso entre la instrucción inicial y las instrucciones o indicaciones adicionales.	35	62	**97**
Apoyos Visuales (VS)	Una demostración grabada en video de la conducta o habilidad específica que se le muestra al alumno para ayudarlo a aprender o participar en una conducta o habilidad deseada.	34	31	**65**

*Traducción de Steinbrenner et al. (2020)

RELACIÓN ENTRE ARTÍCULOS Y JUSTIFICACIÓN DE LA TESIS DOCTORAL

El principal objetivo de tesis ha sido la creación y puesta en marcha de un programa de intervención temprana para menores con TEA. Para la consecución de este objetivo, el cual se ha llevado a cabo en el tercer artículo presentado en la tesis cuyo nombre es "Effect of a Focused Social and Communication Intervention (SCI) on Preterm Children at Risk for ASD: A Pilot Study", se tuvieron en cuenta, tanto el campo de la investigación como a todos los grupos implicados.

Primero, se realizó una revisión sistemática y metaanálisis "Efficacy of focused social and communication intervention practices for young children with autism spectrum disorder: A meta-analysis" para conocer qué técnicas focalizadas a la mejora de habilidades de comunicación social son más eficaces, además del estudio de características como la edad, la dosis de tratamiento, o la participación de los padres en las sesiones.

Una vez que se extrajeron las técnicas más eficaces, se realizó un estudio de encuesta "Early Detection, Diagnosis and Intervention Services for Young Children with Autism Spectrum Disorder in the European Union (ASDEU): Family and Professional Perspectives" para conocer las opiniones y perspectivas de familias de menores con TEA y profesionales que trabajan con este grupo en relación con los servicios. Este estudio permitió conocer aquellos puntos en los que las familias y profesionales tenían mayor o menor satisfacción.

Después de analizar lo que reporta, tanto la investigación, por un lado, como los grupos implicados por otro, se creó e implementó un programa de intervención temprana. Este programa contó con las técnicas que resultaron más eficaces en el primer estudio de

metaanálisis, además de focalizar en los aspectos reportados por familias y profesionales, como la edad de inicio o la participación de los padres.

Cuando los padres comienzan a preocuparse por las dificultades de desarrollo de su hijo, deben hacer un esfuerzo considerable para buscar respuestas a sus preguntas y obtener un diagnóstico y un programa de intervención preciso. Varios estudios indican que los padres de niños con TEA informan niveles más altos de estrés y menor satisfacción con los servicios que los padres de niños con otras discapacidades (Baker-Ericzén, Brookman-Frazee y Stahmer, 2005; Gray, 2006; Griffith, Hastings, Nash, & Hill, 2010; Hayes y Watson, 2013). Las familias con un niño pequeño con TEA reportan mayores dificultades para acceder a los servicios, mayores costes asociados y falta de información y apoyo durante el proceso (Hodgetts, Zwaigenbaum y Nicholas, 2015; Kogan et al., 2008; Thomas, Parish, Rose y Kilany, 2012a; Wang, Mandell, Lawer, Cidav y Leslie, 2013).

En los últimos años, los esfuerzos para mejorar los servicios de intervención temprana para niños con TEA han prestado más atención a las opiniones de las familias y los profesionales, lo que refleja la creencia de que las estrategias de mejora deben centrarse en el niño y su familia (McConachie et al., 2015; Pellicano, Dinsmore y Charman, 2014). El propósito es asegurar que las familias participen más activamente en la evaluación de las necesidades del niño y de la familia, y que los profesionales adopten un enfoque proactivo para identificar tales necesidades. Las familias que informan que participan activamente en las decisiones y tienen una buena comunicación con los profesionales también informan una mayor satisfacción con los servicios, menos brechas en los servicios, menos demoras en el acceso al tratamiento y los servicios, menos estrés y menores costos generales relacionados con el TEA (Kuo, Bird, & Tilford, 2011; Moh y Magiati, 2012; Burke y Goldman, 2015). Asimismo, estudios recientes han demostrado

que, cuando los profesionales responden con prontitud a las inquietudes de los padres, las demoras en el acceso a los servicios se reducen y la satisfacción general aumenta (Zablotsky et al., 2017; Zuckerman, Lindly & Sinche, 2015).

Aunque las opiniones y la satisfacción de las familias y los profesionales con los servicios de intervención temprana parecen haber jugado un papel fundamental en el cambio de políticas y la mejora de los servicios para la comunidad de TEA, las perspectivas de estos dos grupos diferentes rara vez se han considerado juntas. Por lo tanto, es importante obtener información detallada sobre el tipo de servicios que reciben los niños pequeños con TEA, con el fin de desarrollar e implantar un programa de intervención temprana.

El objetivo de la intervención temprana es anticiparse a todas estas preocupaciones y dificultades para iniciar un tratamiento más temprano, incluso antes de que el niño reciba un diagnóstico formal. Este es un momento crítico, cuando el cerebro aún se está desarrollando y los síntomas del TEA están emergiendo (Crais y Watson, 2014). La intervención temprana mejora las habilidades sociales y comunicativas, el desarrollo cognitivo, el lenguaje y el comportamiento adaptativo (Bejarano-Martín et al., 2020; Gates, Kang & Lerner, 2017). También puede reducir la sintomatología del TEA, así como los síntomas secundarios (Reichow, Barton, Boyd & Hume, 2012). La intervención temprana también reducirá la carga de TEA y puede mejorar la calidad de vida de los niños prematuros y sus familias, reduciendo los costos sociales y económicos que las familias y los servicios tienen que enfrentar a lo largo de la vida (Bejarano-Martín, 2020; Fletcher-Watson et al., 2017).

La intervención social y comunicativa ha sido reconocida como uno de los programas más prometedores en TEA (Watkins, Kuhn, Ledbetter-Cho, Gevarter y O'Reilly, 2017; Zwaigenbaum et al., 2015). Además, las habilidades de comunicación

social son clave para el desarrollo de habilidades complejas, como el lenguaje (Bradshaw, Koegel y Koegel, 2017; Hampton y Kaiser, 2016), pero las deficiencias en estas habilidades no parecen mejorar con el tiempo en niños con TEA. letreros (Gates, Kang y Lerner, 2017). En consecuencia, la intervención temprana debería ayudar a los niños en riesgo de TEA a aumentar sus habilidades de comunicación social y reducir los síntomas en esta área (Gates et al., 2017; Murza, Schwartz, Hahs-Vaughn & Nye, 2016; Schreibman et al., 2015; Zwaigenbaum et al., 2015).

Existen varios programas de intervención social y comunicativa dirigidos a niños en riesgo de TEA que buscan reducir los síntomas antes de un diagnóstico formal (Landa, 2018; Rogers & Vismara, 2014). Por otro lado, existen varios programas de intervención temprana destinados a mejorar diferentes habilidades de los bebés prematuros (p. Ej., Chernego et al., 2018). Sin embargo, ninguno de estos programas de intervención temprana ha estudiado su eficacia en niños con riesgo de TEA. Tampoco está claro si los menores con riesgo de TEA pueden disminuir sus deficiencias después de participar en un programa de intervención temprana, lo que reduciría las diferencias con los menores sin riesgo de TEA.

Dentro de los programas de intervención social y comunicativa encontramos las prácticas de intervención focalizada. Estas son prácticas o estrategias de instrucción individuales que se utilizan para enseñar habilidades específicas a niños con TEA en un período de tiempo relativamente corto (p. Ej., 0-3 meses) (Odom, Boyd, Hall, Hume, 2010a; Odom, Klinbergerg , Rogers y Hatton, 2010b; Wong et al., 2015). Se utilizan con frecuencia para mejorar las habilidades sociales y de comunicación y se han convertido en el foco de la mayoría de las terapias para niños pequeños (6 años o menos) con TEA para satisfacer las demandas de la sociedad (Schreibman et al., 2015; Wang et al., 2013; Zwaigenbaum et al., 2015). Aunque existe evidencia que demuestra que los FIP son

efectivos para el desarrollo de habilidades sociocomunicativas en niños pequeños con TEA, los resultados varían según la habilidad a la que se dirige (Zwaigenbaum et al., 2015).

En general, existe una gran heterogeneidad en los resultados obtenidos por diferentes estudios. Esta heterogeneidad puede deberse al efecto de determinados factores, como las características de los participantes (edad cronológica, sexo, nivel de gravedad de los síntomas) o de la intervención (individual / grupal) que podrían estar actuando como variables moderadoras que influyen en la fuerza y / o dirección de la relación entre el tratamiento y los resultados (Kazdin, 2007; Lerner, White y McPartland, 2012; Spielmans y Flückiger, 2018). El estudio de las posibles variables moderadoras puede ayudar a identificar y explicar los mecanismos involucrados en el resultado del tratamiento. Los estudios de eficacia deben tener en cuenta estos mediadores y mecanismos cuando intervienen en el resultado del tratamiento, ya sea para explicar, al menos teóricamente, cómo se relacionan los mediadores con las variables de resultado, o para considerar cómo las variables del tratamiento influyen en los mediadores (Gottfredson et al., 2015)

Objetivos:
general y específicos

OBJETIVO GENERAL

El objetivo general de la Tesis Doctoral consiste en el desarrollo, puesta en marcha y evaluación de un programa de intervención temprana, específico para menores con TEA, que incluya todos los procedimientos que han probado ser eficaces en esta población en base a la evidencia científica, y que tenga en cuenta las opiniones de las familias y profesionales que ofrecen servicios a esta población, cuyo fin último es la mejora de las habilidades socio-comunicativas de los menores con TEA y sus familias.

OBJETIVOS ESPECÍFICOS

La consecución del objetivo general de la presente Tesis Doctoral se ha llevado a cabo a través de tres estudios de investigación, los cuales se presentan por medio de tres publicaciones científicas. Cada una de estas investigaciones cuentan con una serie de objetivos. Estos objetivos se centran en el análisis de los programas de intervención existentes, el estudio de las opiniones de las familias y profesionales, y la puesta en marcha de un programa de intervención temprana en menores con TEA.

Artículo I:

Efficacy of focused social and communication intervention practices for young children with autism spectrum disorder: A meta-analysis.

a) Examinar las prácticas de intervención focalizada para menores con TEA en términos de: su efectividad en relación con las habilidades básicas de

comunicación social; y sus beneficios según el participante seleccionado y las características del programa.

b) Examinar si diferentes aspectos, como las características del participante y del programa (p. ej., edad, coeficiente intelectual, nivel de lenguaje, dosis de intervención y participación activa de la familia y / o del maestro en la intervención) influirán en los resultados del tratamiento.

Artículo II:

Early detection, diagnosis and intervention services for young children with Autism Spectrum Disorder in the European Union (ASDEU): family and professional perspectives.

a) Identificar los tipos de servicios que reciben los niños con TEA en Europa.
b) Examinar el grado de satisfacción de las familias y los profesionales con los servicios en toda Europa.
c) Explorar las variaciones de edad en el momento de la detección, el diagnóstico y la intervención y las demoras en el acceso a los servicios, reportado por padres y profesionales.
d) Identificar las variables que predicen la satisfacción de los servicios en ambos grupos.

Artículo III:

Effect of a focused Social and Communication Intervention (SCI) on preterm children with ASD: a pilot study.

a) Examinar el efecto de un programa de intervención en comunicación social en menores prematuros y a término con TEA.
b) Explorar si hay diferencias en los beneficios de la intervención entre menores prematuros con TEA, menores a término con TEA (grupo de comparación) y menores prematuros sin TEA (grupo de control).
c) Investigar el efecto individual de la intervención.
d) Evaluar la viabilidad y aceptabilidad del programa de intervención en comunicación social en niños prematuros y a término con TEA.

Publicaciones científicas

ARTÍCULO I: METAANÁLISIS DE TÉCNICAS DE INTERVENCIÓN SOCIOCOMUNICATIVAS

Bejarano-Martín, Á., Canal-Bedia, R., Magán-Maganto, M., Fernández-Álvarez, C., Lóa-Jónsdóttir, S., Saemundsen, E., Vicente, A., Café, C., Rasga, C., García-Primo, P., & Posada, M. (2020a). Efficacy of focused social and communication intervention practices for young children with autism spectrum disorder: A meta-analysis. *Early Childhood Research Quarterly*, *51*, 430-445. https://doi.org/10.1016/j.ecresq.2020.01.004

Resumen

Introducción. Las prácticas de intervención focalizadas (FIPs) se utilizan ampliamente para mejorar las habilidades de comunicación social, ya que están dirigidas específicamente a mejorar las habilidades identificadas como problemáticas en los menores con Trastorno del Espectro Autista (TEA), como la imitación, el contacto visual, los gestos, la atención conjunta y el juego. ***Objetivos.*** Este metaanálisis se realizó para determinar la efectividad general de las FIP en menores con TEA de 6 años o menos. ***Método.*** Se realizaron cinco búsquedas electrónicas, se recuperaron 1.828 referencias y se incluyeron 43 estudios (59 medidas de resultado) en el metaanálisis. Los estudios incluyeron 785 participantes (41,6 meses) con TEA. El tamaño del efecto sociocomunicativo global para cada habilidad específica (imitación, atención conjunta y juego) se calculó utilizando la g de Hedges (g) para los estudios de diseño de grupo, y el

Nonoverlap of All Pairs (NAP) para los estudios de diseño de caso único. También se utilizaron modelos de metarregresión de efectos aleatorios y correlaciones para evaluar si los resultados eran diferentes según las características de la población y la intervención. Se analizó el impacto del posible sesgo de publicación. ***Resultados.*** Los resultados sugieren que, mientras que las FIP tienen efectos positivos de medianos a grandes (g=0,51; NAP=0,86), aquellos en los que los cuidadores o profesores desempeñan un papel activo (g=0,50; NAP=0,89) tienen tamaños de efecto medianos. Todos los resultados de las habilidades sociales y comunicativas de las FIP tienen tamaños de efecto medios (Imitación: g=0,42, NAP=0,90; Atención conjunta: g=0,54, NAP=0,86; Juego: g=0,47, NAP=0,81). ***Discusión.*** Los tamaños de los efectos fueron mayores cuando la edad de los participantes antes de la intervención era menor y la dosis de tratamiento era mayor. A la hora de conseguir mejoras sustanciales, los factores a destacar fueron el papel de los cuidadores y la adaptación del programa a las características del menor. La aplicación de los programas de intervención temprana debe estar fundamentada en una cantidad suficiente de información sobre las características de cada participante. Los profesionales deben tener en cuenta esta información para seleccionar con la mayor precisión posible los procedimientos más eficaces y factibles.

Palabras clave. Trastorno del espectro autista; habilidades sociocomunicativas; intervención; meta-análisis; eficacia.

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Review

Efficacy of focused social and communication intervention practices for young children with autism spectrum disorder: A meta-analysis

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ABSTRACT

Focused intervention practices (FIPs) are widely used to improve social communication skills, as they are specifically aimed at enhancing skills identified as being problematic in children with autism spectrum disorder ASD, such as imitation, eye contact, gestures, joint attention and play. This meta-analysis was performed to ascertain the overall effectiveness of FIPs in children with ASD 6 years of age and younger. Five electronic searches were conducted, 1828 references were retrieved, and 43 studies 59 outcome measures were included in the meta-analysis. Studies included 785 participants 41.6 months with ASD. The overall socio-communicative effect size for each specific skill imitation, joint attention, and play was calculated using the Hedges' g (*g*) for group design studies, and the Nonoverlap of All Pairs (NAP) for single case design studies. Random-effects metaregression models and correlations were also used to assess whether the results were different according to population and intervention characteristics. The impact of possible publication bias was analysed. The results suggest that, whereas FIPs have medium to large positive effects (*g* = 0.51; NAP = 0.86), those where caregivers or teachers play an active role (*g* = 0.50; NAP = 0.89) have medium effect sizes. All social and communicative skills outcomes of FIPs have medium effect sizes (Imitation: g = 0.42, NAP = 0.90; Joint attention: g = 0.54, NAP = 0.86; Play: g = 0.47, NAP = 0.81). Effect sizes were greater when participants' preintervention ages were lower and treatment dosage was higher. When it comes to achieving substantial improvements, factors to be highlighted are the role of caregivers and adaptation of the programme to the characteristics of the child. Implementation of early intervention programmes should be substantiated by a sufficient amount of information about the characteristics of each participant. Professionals should take this information into account in order to select as accurately as possible those procedures that are most effective and feasible.

Contents

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1. Introduction

Impairment in social communication skills has been recognised as one of the key problems in children with autism spectrum disorder (ASD) (Watkins, Kuhn, Ledbetter-Cho, Gevarter, & O'Reilly, 2017; Zwaigenbaum et al., 2015), and is given as one of the criteria for diagnosis of ASD in the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) (American Psychiatric Association, 2013). Furthermore, social communication skills are fundamental for the development of more complex abilities, such as language (Bradshaw, Koegel, & Koegel, 2017; Hampton & Kaiser, 2016), but impairments in these skills do not seem to ameliorate with time in children with ASD (Gates, Kang, & Lerner, 2017). Consequently, intervention models should help children with ASD to increase their social communication skills and reduce the greatest number of severe symptoms in this area (Gates et al., 2017; Murza, Schwartz, Hahs-Vaughn & Nye, 2016; Schreibman et al., 2015; Zwaigenbaum et al., 2015. The overall ASD prevalence was 17 per 1000 children aged 4 years in 2014 for Early Autism and Developmental Disabilities Monitoring Early ADDM sites Christensen, 2019).

Focused intervention practices (FIPs) are individual instructional practices or strategies used to teach specific skills to children with ASD in a relatively short period of time (e.g., 0–3 months) (Odom, Boyd, Hall, & Hume, 2010; Odom, Collet-Klingenberg, Rogers, & Hatton, 2010; Wong et al., 2015). They are frequently used to improve social and communication skills and have become the focus of most therapies for young children (6 years and younger) with ASD to meet society's demands (Schreibman et al., 2015; Wang, Parrila, & Cui, 2013; Zwaigenbaum et al., 2015). Behavioural strategies, naturalistic interventions, picture exchange communication systems, pivotal response training and visual supports are all examples of FIPs (Boyd, Odom, Humphreys, & Sam, 2010). In recent years, there have been empirical research reviews covering both single-case and group-design studies of FIPs aimed at improving social and communication skills (Bradshaw, Steiner, Gengoux, & Koegel, 2015; French & Kennedy, 2018; Reichow, Barton, Boyd, & Hume, 2012; Waddington, van der Meer, & Sigafoos, 2016). However, these reviews do not always include an evaluation of study quality. Despite the extensive use of FIPs, little research has thus been done to examine their overall efficacy (Kasari, Shire, Factor, & McCracken, 2014).

1.1. Current evidence in support of FIPs in ASD

There are some meta-analytical studies that have rigorously assessed and corroborated the effectiveness of FIPs, and reported a medium effect. Reichow et al. (2012) reported outcomes that support these types of programmes with behavioural approaches (g = 0.47). Similarly, Murza et al. (2016) reviewed the efficacy of FIPs, specifically in the case of joint attention, and their results substantiate the efficacy of this type of intervention (g = 0.66), as do those of Gates et al. (2017), who also provide substantial supporting evidence (g = 0.51). Despite these promising results, these studies focused exclusively on behavioural approaches (Reichow et al., 2012), in youth individuals with ASD (>5 years) (Gates et al., 2017), or on a specific targeted skill (Murza et al., 2016), thereby underscoring the limited nature of the literature on the effectiveness of different FIPs (not only behavioural approaches) in young children with ASD (under 6 years) on a wide range of targeted skills.

1.2. FIPs in targeted social and communication skills

Although there is evidence to show that FIPs are effective for the development of socio-communicative skills in young children with ASD, outcomes vary depending on the skill being targeted (Zwaigenbaum et al., 2015). These differences highlight the complexity of social-communication skills, including behaviours such as imitation, eye contact, joint attention, gestures and play, which are also skills evaluated for diagnosis of ASD via the ADOS-2 (Autism Diagnostic Observation Schedule, Second Edition) "gold standard" tool (Lord, Rutter, DiLavore, Risi, Gotham & Bishop, 2012).

Imitation plays a fundamental role in the development of social skills, social-emotional communicative functions, and theory of the mind, as well as language and play skills (Contaldo, Colombi, Narzisi, & Muratori, 2016; Cooley, 2017; Dohmen, Bishop, Chiat, & Roy, 2016). Imitation promotes a social orientation toward others (Contaldo et al., 2016), and children improve attention to adults instead of objects, and smile more as they perform their actions (Carpenter et al., 2002). They recognise this "imitation play" from the age of 9 months (Agnetta & Rochat, 2004). Therefore, imitation has become a critical ability to work on different intervention therapies, both to reduce the symptoms of ASD and to increase the skills that are associated (Ingersoll, 2010; Landa, 2018; McDuffie et al., 2007).

For example, eye contact, which is impaired in children with ASD (Jones & Klin, 2013), is an essential component of daily social relations and provides the basis for the development of more complex skills, such as social engagement (Franchini et al., 2017). Children of 1–2 months of age who were later diagnosed with ASD showed a level of visual contact equivalent to children with typical development (Jones & Klin, 2013), decreasing the frequency as they grew. This decline in eye contact, rather than an absence, offers a promising opportunity for early intervention.

Joint attention is another significant deficit in children with ASD (Mundy, 2018) and is one of the key FIP components (Kasari, Gulsrud, Paparella, Hellemann, & Berry, 2015; Murza et al., 2016). Joint attention correlates with language development and other social communication skills (e. g., play, imitation) (Adamson, Bakeman, Suma, & Robins, 2019; Bottema-Beutel, 2016; Kasari et al., 2015; Pickard & Ingersoll, 2015). This robust correlation improve many aspects of language development, such as expressive and receptive language, vocabulary, and gestures. More importantly, by teaching joint attention skills to children with ASD they could learn to pay more attention to the social behaviour of others and incorporate into their repertoire the use of social skills such as gestures, vocalizations or emotional expressions, which promotes their social involvement in daily routines (Meindl & Cannella-Malone, 2011). In this way, children with ASD would begin to take others into account and their social engagement would increase by being able to share attention on objects or events and people.

In addition, children with ASD have difficulties using gestures to acknowledge an object or its properties (Gordon & Watson, 2015; Mastrogiuseppe, Capirci, Cuva, & Venuti, 2015), and these nonverbal communication movements have been included in intervention programmes to improve the development of skills such as joint attention (pointing), imitation, play, as well as language (Ingersoll & Lalonde, 2010; Özçalışkan, Adamson, & Dimitrova, 2016; Paparella & Freeman, 2015).

Children with ASD encounter many difficulties in play, especially when it comes to performing sequences of symbolic and pretend play (Wilson et al., 2017). Play skills are important predictors of later language, social, and communication skills (Kasari, Paparella, Freeman, & Jahromi, 2008) and of the development of theory of mind (Lin, Tsai, Li, Huang, & Chen, 2017). Moreover, play has a reciprocal relationship with the development of skills such as language. Children who improve their play skills also improve the development of other skills (Pierucci, Barber, Gilpin, Crisler, & Klinger, 2015), therefore, it is justified that intervention programmes promote the development of play skills.

Hence, examining change in these targeted skills as intervention outcomes, in order to compare them, both separately and overall, may be useful for the purpose of understanding FIP outcomes according to the specific skills targeted.

1.3. Plausible moderators of FIP effects

In general, there is a wide heterogeneity in the results obtained by different studies. This heterogeneity may be due to the effect of certain factors, such as the characteristics of the participants (chronological age, sex, level of symptom severity) or of the intervention (individual/group) that could be acting as moderating variables that influence the strength and/or direction of the relationship between treatment and outcomes (Kazdin, 2007; Lerner, White, & McPartland, 2012; Spielmans & Flückiger, 2018).

Examining the potential moderator variables may help explain the heterogeneous results within meta-analysis studies about treatment efficacy and may help identify and explain mediating variables and mechanisms involved in treatment outcome. Efficacy studies should take these mediators and mechanisms into account as they intervene in treatment outcome, either to explain, at least theoretically, how mediators relate to outcome variables, or to consider how treatment variables influence mediators (Gottfredson et al., 2015). The authors of the studies are therefore expected to identify and describe, within the approach underpinning the treatment they propose, the role of possible mediators. Alternatively, the description of treatment characteristics and the type of measures used in the studies should include data on mediating variables, as well as information on the moderating effect of other variables and the possible effect that the treatment might have on the identified mediators (i.e. indirect effects of socio-communicative programs on variables such as symptomatology level, language, IQ, adaptive behavior). This would make it possible to identify the characteristics of the intervention that are most central in explaining the observed change.

Taking into consideration these elements, the study on the efficacy of a given treatment could allow some kind of proposal to be made about the mechanism that produces the result of the treatment. On the other hand, a meta-analysis study could analyse the specific influence of different moderating variables, since it would combine the results of several studies, in which there will be certain heterogeneity with respect to the characteristics of the participants, as well as variations in the characteristics of the treatment. Thus, by means of a meta-analysis, it would be possible to obtain conclusions on the generability of the treatment.

Participants and FIPs' characteristics (e. g. age or treatment dosage) were chosen as moderators of the effect. These moderating variables have been studied extensively as they are the ones that most influence the intervention outcome. (Kazdin, 2007). Another characteristic that has been studied lately is the active participation of parents in the intervention, increasing the effect of the intervention and reducing parental stress and family burden, suggesting that family participation should be taken into account when studying the effect of a treatment. (de Veld et al., 2017; McIntyre & Zemantic, 2017; Stadnick, Drahota, & Brookman-Frazee, 2013).

1.4. Participant characteristics

The age that FIPs are implemented can vary widely. Although Gates et al. (2017) have shown how these interventions are effective for youth (5–21 years) with ASD (g = 0.51), no study has investigated whether these results would be consistently main-

tained in children with ASD from very early ages until they reach the age of 6 years. Significant progress in reducing ASD identification age has been observed in recent years (Mazurek et al., 2014), as it is now possible to detect signs of risk in children aged 12–14 months (see Jones et al., 2014), as a result, the demand for effective strategies designed to be implemented in early intervention programmes is increasing (Zwaigenbaum et al., 2015). The aim of early detection is to identify signs of risk or early symptoms of ASD in the child's development, in order to initiate earlier treatment, even before the child receives a formal diagnosis. Thus, early detection only makes sense if there is early treatment available that has proven its efficacy.

Systematic early detection actions associated with the initiation of early intervention activities are linked to positive outcomes for children at risk for ASD, as children who begin treatment earlier have better outcomes (MacDonald, Parry-Cruwys, Dupere, & Ahearn, 2014; Orinstein et al., 2014). Hence, the ultimate goal is for intervention to occur during the child's critical period of development, when neuronal plasticity is greater and long-term positive results can be achieved (Crais & Watson, 2014). In addition, the initiation of early intervention activities could also have positive consequences for the family, who may find answers to their concerns and learn strategies to cope with the developmental difficulties of their child (Ingersoll, Shannon, Berger, Pickard, & Holtz, 2017; Kasari et al., 2015).

Participants' cognitive development, verbal ability, and IQ may change the effect of FIPs' efficacy. Studies report significant differences in social communication skills in those cases where participants' IQ or verbal ability is higher at baseline (Virués-Ortega, 2010). Improvement in such abilities at baseline seems to be a predictor of a reduction in ASD severity and an increase in adaptive abilities (Zachor & Ben-Itzchak, 2017). This means that the role of IQ and verbal ability in the effect of FIPs must be studied, in order to recommend suitable intervention programmes taking into account these characteristics in the participants.

1.5. FIP characteristics

One of the most controversial variables is the dosage of the intervention. Numerous studies recommend that in order to yield positive effects on the targeted skills, treatment should be as long (duration of the intervention in weeks/months/years) and intense (hours per week) as possible (Granpeesheh, Dixon, Tarbox, Kaplan, & Wilke, 2009; Klintwall, Eldevik, & Eikeseth, 2015; Linstead, Dixon, French et al., 2017; Zwaigenbaum et al., 2015). However, the use of programmes that can be taught in a short period of time (FIPs) reduce costs and waiting time delays (Ingersoll et al., 2017; Kasari et al., 2015). The findings are mixed: whereas one meta-analysis reported that the intensity (hours per week, ranged from 6 to 45) seemed to partially influence the outcome of the intervention (Virués-Ortega, 2010), other studies reported that this had a strong influence on the outcome (hours per month, ranged from 20 to 198) (Eldevik et al., 2010; Linstead, Dixon, French et al., 2017, Linstead, Dixon, Hong et al., 2017). It is therefore necessary to study whether there is a significant variation in the effect of FIPs depending on the dosage of the intervention.

Another aspect that must be taken into account is the fidelity of the intervention, which is the extent to which the intervention is delivered as it was intended (Gearing et al., 2011). Reporting the intervention fidelity in the published product is crucial in order to assess the quality of the study and to understand how different factors may have influenced the outcome of the study (Murphy & Gutman, 2012). If treatment fidelity is not sufficiently assured, significant uncontrolled variability in effect sizes may appear. (Mandell et al., 2013)

The people involved in the intervention program must be considered because they too influence the effectiveness of the FIPs. Several reviews suggest that the active participation of parents is an aspect that must be taken into account when evaluating the effectiveness of intervention outcomes (Bradshaw et al., 2015; Debodinance, Maljaars, Noens, & Van den Noortgate, 2017; Reichow, 2012; Zwaigenbaum et al., 2015). Several studies have shown the effectiveness of intervention programmes where parents and/or teachers actively participate with the main therapist, after receiving specific training (Lawton & Kasari, 2012; Schertz, Odom, Baggett & Sideris, 2013; Wong, 2014).

Finally, parental involvement is also fundamental for considering the satisfaction with intervention programme (McIntyre & Zemantic, 2017; Stadnick et al., 2013). In addition, involving parents reduces intervention costs by decreasing the number of treatment hours with professionals and increasing skill development in natural contexts (Ingersoll et al., 2017; Pickles et al., 2016). All these aspects mean that parental involvement in interventions could significantly reduce the economic burden for the family, health-care system, and society, and it could also decrease the stress associated with raising a child with ASD (Kasari et al., 2015). Despite showing positive effects in some studies, it is not clear whether or not parental involvement in intervention programmes increases the effect of FIPs, and thus, it is important to identify the characteristics of parent participation procedures that achieve significantly positive outcomes.

1.6. Measuring FIPs

A key concept when examining the effectiveness of interventions such as FIPS is whether the comparison measures are adequate and accurate. Within the group-design studies, Randomised Control Trials (RCTs) and Quasi-Experimental Designs studies (QEDs) use posttest measures to calculate standardised mean difference. RCTs are considered a Ġold Standardänd the ideal option in research on treatment efficacy because the randomization of RCTs increases the probability that the groups are equivalent and comparable in terms of the variables of interest (e.g., participant characteristics), allow better control of possible biases, and provide greater security in the determination of causality. However, for practical or ethical reasons, it is not always possible to carry out RCTs and some researchers opt for QEDs or Single Case Design studies (SCDs) when there are few participants. In QEDs and SCDs, as they do not include randomisation procedures, there may be a selection bias that can interact with independent variables and a priori the equivalence of the groups (QEDs) is not guaranteed or there are no groups to compare (SCDs), which poses a threat to the internal validity of the study. Therefore, since in QEDs and SCDs the effect of the intervention could be due to uncontrolled variables, rather than to variables considered independent, it is critical to make an analysis of the quality of the QEDs and SCDs that includes, among other aspects (experimental mortality, testing effects, etc.), information on the groups selection process, type of measures, etc. In QEDs, only those studies that have proved that there are no significant differences between the comparison groups at baseline would be incorporated into the meta-analysis. That is to say, we will select those QEDs where quality analysis establishes that the study method includes procedures to ensure the equivalence of the different groups in terms of general and specific criteria (age, sex, symptoms, IQ, etc.), which may lead to biases in the effects of the intervention (French & Kennedy, 2018; Wong et al., 2015). In SCDs, only studies that have proved to meet quality standards (See Kratochwill et al., 2010), such as interobserver agreement (IOA) of at least 80%, a second observer measured the variable 20% of all phases, and provided repeated measures across different conditions. Hence, we will select those SCDs whose quality analysis

establishes that the study method includes procedures to ensure that the effect of the intervention is produced exclusively by the FIPs, and allow the replicability of the study.

1.7. Current study

The first aim of this study was to examine FIPs for young children with ASD in terms of: their effectiveness in relation to basic social-communication skills; and their benefits according to selected participant and programme characteristics. The second objective was to examine whether different aspects, such as participant and FIP characteristics (e.g., age, IQ, language level, dosage of intervention, and family and/or teacher actively participation in the intervention) will moderate treatment outcomes. In addition, the methodological rigor on the final set of studies selected for the meta-analysis was also measured.

2. Methods

2.1. Identification and selection of studies

An electronic search was conducted in the following databases: PsycINFO; PUBMED; Educational Resource Information Centre; and Cumulative Index to Nursing and Allied Health Literature. Five different searches were made, one for each targeted skill, using the following search criteria: (autism OR autism spectrum disorder OR ASD OR autistic disorder OR Pervasive Developmental Disorder OR Asperger) AND (imitation; eye contact; joint attention; gestures; play) AND (training OR treatment OR intervention OR teach OR teaching OR development OR improving OR therapy).

2.2. Study-selection and literature search

The design and development of the meta-analysis was carried out in two phases. The first phase consisted in the screening of references (selection of the studies by eligibility criteria) retrieved from the electronic search. In this phase, the studies were first screened by title, then by abstract and finally by full text, according to the eligibility criteria (Appendix A). The second phase focused on the methodological quality of the studies selected in the first phase. Eligibility criteria for this phase were different for studies with group and single case design.

2.2.1. First stage of literature search

The systematic literature search was conducted from January 2000 to July 2018. Fig. 1 gives a detailed layout of the study-identification and selection process. The search goals were established according to the recommendations provided by the Cochrane Collaboration (Higgins & Green, 2011), based on the PICOS structure (Participants, Interventions, Comparison, Outcomes, Study design).

2.2.2. Inclusion and exclusion criteria

The study was selected and reviewed if: (a) was empirical; (b) was published in a peer-reviewed journal; (c) written in English; ; (d) participants were six years of age or younger; (e) participants were diagnosed with ASD, according to the criteria of DSM-5 (American Psychiatric Association, 2013) or DSM-IV-TR (APA, 2000); and (f) used FIPs involving behavioural, developmental, or educational interventions to improve communication and social interaction skills (imitation, eye contact, joint attention, gestures, and play). The exclusion criteria were defined as studies that: (a) did not use "gold standard" measures, such as ADOS (Lord et al., 2000, 2012) in their diagnostic assessment; and, (b) included medical or pharmacological interventions.

2.2.3. Study-selection procedure

A total of six reviewers participated in all selection phases (from title review to full text) independently applying the eligibility criteria (inclusion/exclusion). The criteria were formulated as specific questions for each phase of the selection process, where the reviewers had to code "Yes" or "No". The intercoder agreement focused on the proportion of observed concordance between reviewers in relation to the eligibility criteria. Studies had to meet all the inclusion and exclusion criteria to move on to the next review phase (See Appendix A). All the studies were screened by two independent reviewers who belonged to the research group associated with the ASDEU project (Autism Spectrum Disorder in the European Union, 2015–2018ASDEU, 2020Autism Spectrum Disorder in the European Union, 2015–2018). A third reviewer was consulted to settle any disagreements between reviewers during the various selection stages.

Taking into account the above search criteria, 2894 studies were identified once duplicates had been removed. The title and abstract screening were completed with the mentioned eligibility criteria with excellent reliability *Kappa*: 1.00, 0.96 respectively; Cohen, 1968).

The full texts were screened again applying the following additional criteria (see Appendix A): (a) studies that included quantitative data outcomes in respect of the targeted skills analysed; (b) studies that included pre and postquantitative data; and (c) original studies with group (RCT or QED) or single-case designs (withdrawal of treatment (e.g., ABAB), multiple baseline, multiple probe, alternating treatment, and the changing-criterion design) with excellent reliability (*Kappa* = 0.92). This process yielded 116 studies eligible for the second stage of the review.

2.2.4. Quality-selection procedure

In the second phase, a review of the quality of the studies selected in the first phase (116 studies) was conducted. Single case and group design studies were screened. To include a study in the final meta-analysis, it had to meet the quality criteria of the second phase. The purpose of this revision was to ensure the methodological quality and rigor of intervention studies (single case and group design) and that they complied with specific standards that would ensure that the size of the effect would have been produced by the use of the FIPs. We conducted this quality review of all full-text studies selected in the first phase using the EBP Update Workgroup Reviewer Training criteria (Wong et al., 2015) of the National Professional Development Centre on Autism Spectrum Disorders. The EBP Inclusion Criteria Checklist (https://autismpdc.fpg.unc.edu/sites/autismpdc.fpg.unc.edu/files/imce/documents/EBP-InclusionCriteriaDesignChecklists.pdf) consist of 10 items for group design and 9 items for single case design studies. For a study to be included with this criterion, all items must be answered with a "yes". Examples for the Group design EBP Inclusion Criteria Checklist are: Does the study have experimental and control/comparative groups? Was the control/comparison condition(s) described? Was attrition NOT a significant threat to internal validity? Examples of Single Case Design EBP Inclusion Criteria Checklist are: Did a secondary observer collect data on the dependent variable for at least 20% of sessions across conditions? Was mean interobserver agreement (IOA) 80% or greater OR *kappa* of 0.60 or greater?

In this second stage 43 studies were selected for quantitative synthesis. Some of these studies measured outcomes from more than one skill. For example, a study could present results from two skills, one on imitation and one on joint attention, using the same FIP. Thus, 59 outcome measures were identified from all of the 43 studies for all the different skills analysed in this research (see Appendix C. Table 1 for details). Each of these 59 outcome measures only reported results in one of the social communication

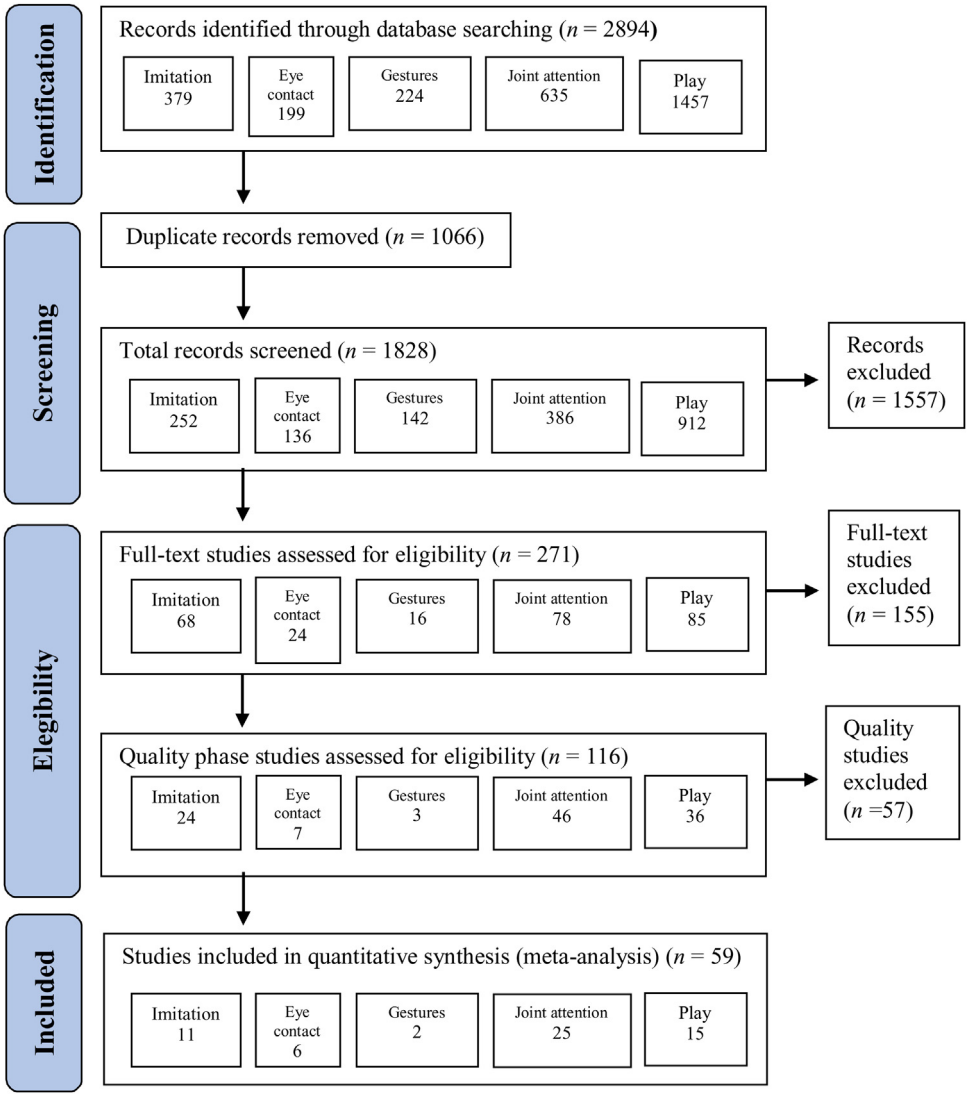

Fig. 1. PRISMA flowchart representing the identification and selection of the studies.

skills analysed in our study. The mean Cohen's *Kappa* coefficient for quality reviews was excellent (>0.98).

2.2.5. Data extraction

The 43 studies were double-coded by two independent reviewers (see Appendix B on Supplemental material). For this stage, a variable extraction protocol was created with the corresponding operational definitions and coding values. This protocol had four sections with different variables to complete according to the information from the study. These sections were the following: (a) participant characteristics at the time of inclusion in the corresponding study (e. g., age, gender, race, diagnosis, cognitive development, language level); (b) intervention characteristics (treatment dosage, parents' participation, specific FIP, professional profile); (c) study design (Randomized Control Trial (RCT), Quasi-experimental design (QED), Multiple baseline (MB), Alternating treatment (AT), etc.); and (d) outcomes based on specific social-communication skills were collected in at least one of the following two ways: (a) through general standardised measures of development, such as communication and social skill scales; and/or (b) through quantitative data on the various behaviours targeted by the intervention, as observed by two or more therapists via video recordings (frequency/percentage/steps of acts). In addition, we included data gathered throughout the different intervention stages: (a) baseline; (b) intervention; and, (c) postintervention/follow-up. The results of the studies were subcategories of the dependent variables under study (e.g., motor imitation, symbolic play) (for further details see Appendix K on Supplemental material). To settle any disagreements during the reviewing process, a third reviewer was consulted. Agreement among coders was excellent, *Kappa* > 0.89.

The specific outcome measures for each social and communicative skills in the studies (see Appendix C, Tables 1 and 2 in Supplementary material) was used to obtain the effect sizes, both total and of each studied ability. However, the different articles did not rely on the same research study dataset, so that each article represented a different study. Variables related to the characteristics of the participants (age, IQ, language level), as well as the treatment dosage, were also key analytic variables used in the meta-analysis.

This study did not describe the models that support each FIP, but the interested reader can review the following references: Odom, Boyd et al., 2010, Odom, Collet-Klingenberg et al., 2010; Wong et al., 2015; and see the Table 1 - Appendix D on Supplemental material for further information.

2.3. Meta-analytical procedure

The meta-analytical procedures used in this study adhered to the guidelines contained in the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (Moher, Liberati, Tetzlaff, Altman, & Group, 2009). PRISMA is an evidence-based minimum set of items for reporting in meta-analyses. PRISMA focuses on the reporting of reviews evaluating randomized trials, but can also be used as a basis for reporting of other types of research, particularly evaluations of interventions (PRISMA, 2019; http://www.prisma-statement.org/).

The current study included five separate meta-analyses: the first included social and communication overall measures from each study (group design, n = 18; single case design, n = 25); and the second explored the intervention programmes where caregivers and/or teachers played an active role (group design, n = 9; single case design, n = 7), whether individually after receiving training, or jointly along with the main therapist. In order to examine the different social and communication skills separately, the other meta-analyses consisted of the different outcome measures reported for each social and communication skill, i.e., imitation (group design, n = 4; single case design, n = 7), joint attention (group design, n = 14; single case design, n = 10), and play (group design, n = 7; single case design, n = 9). The meta-analysis for gestures and eye-contact abilities was not included, due to the limited number of studies obtained from both group and single-case designs.

2.4. Statistical analyses

To calculate the effect size of the different interventions, the studies were separated as follows: (a) group design; and, (b) single-case design.

The random effect model was used for the point estimate of effect size in the group-design studies, due to the difference in the progression of results (pre — posttest, pretest — follow-up) among the experimental and the control/comparison groups. The effect size (g), was calculated using the difference between means of the treatment group and the control group, divided by the standard deviation, and weighted for sample size to correct for small sample bias (Hedges & Olkin, 1985) (95% CI). The Cohen convention Cohen (1988) was used to interpret the results, since these two statistical variables are extremely similar and comparable (e.g., 0 < g < 0.30 = small; 0.30 < g < 0.80 = moderate; g > 0.80 = large). Effect sizes were calculated for each outcome measure, and then averaged together to make an overall effect size for each study. Some studies measured the effect of treatment on more than one skill (e.g., Van der Paelt et al. (2014) analysed the effect of a FIP on imitation, joint attention and play skills). In our study we have differentiated the overall effect and the specific effect of the outcome measures. The overall effect has been obtained by aggregating all treatment effects on all outcome measures in the study. The specific effect of the outcome measures is the effect of the treatment on each skill measured in the studies. However, each article represented a different study. The standardised mean difference using Hedges' g for small sample correction was calculated using Comprehensive Meta-Analysis software (CMA) version 3 (Borenstein, Hedges, Higgins, & Rothstein, 2005).

Random-effects meta-regression models (Thompson & Sharp, 1999) were also used to assess whether the results were different according to population and intervention characteristics, such as age, cognitive development, language preintervention, and treatment dosage. In every case where heterogeneity was detected (Q: I^2), a moderator analysis was conducted. The Q test inspects for heterogeneity by adding the squared deviation of the effect size for each study of the overall effect size and weighting each study by variance (Higgins & Green, 2011). The I^2 statistic describes the percentage of variation across studies caused by heterogeneity rather than chance, regardless of the treatment effect metric (Higgins & Thompson, 2002). Values of around 25%, 50% and 75% refer to low, medium and high heterogeneity respectively. Although I^2 was developed to be independent of the number of studies, it should be interpreted with caution in cases where few studies are meta-analysed (Huedo-Medina, Sánchez-Meca, Marín-Martínez, & Botella, 2006). To evaluate whether it would be appropriate to conduct a moderator analysis on any given effect, the following criteria were established (from Gates et al., 2017): (a) at least 10 studies had to be included in the analysis; and, (b) in the absence of significant Q statistics, there had to be evidence of at least a nontrivial amount of heterogeneity according to the I^2 statistic ($\geq$20%). The moderators of interest were age, treatment dosage, overall cognitive ability and language. In addition, correlation analyses were performed among those moderating variables that were significant in the effect of the intervention.

For single-case-design studies, the nonoverlap of all pairs (NAP) index was used (Parker & Vannest, 2009). Nonoverlapping data were analysed as an indicator of the performance differences between the different stages of visual analysis during single-case research (SCR) (Sidman, 1960). The NAP index has been included in recently proposed standards for evaluating SCR (Horner et al., 2005) and summarises all overlapping points within each stage (baseline-intervention, baseline-follow-up). It is equivalent to the number of comparison pairs showing no overlap, divided by the total number of comparisons. Moreover, the index can be calculated manually from an SCR graph, where individual graphs of each study can be extracted. These graphs were introduced into a software programme (Digitizelt, version 2.2.2), which extracts all points of the graph numerically. After extracting these numbers, we calculated the NAP index using the NAP calculator (http://www.singlecaseresearch.org/calculators/nap). For single-case studies, correlations were made between the effect size of the programmes and the respective study characteristics, such as age, cognitive development, language preintervention and treatment dosage.

Group and single case design studies with larger effect sizes were selected to see what commonalities they shared in terms of moderators. To do this, studies whose effect size were higher than the overall effect size calculated later were selected. Descriptive analyses of these variables were conducted (*mean*; *SD*). In addition, the Coefficient of Variation (*CV*) of the mean was calculated. The *CV* inform about the homogeneity of the data (if a set of data shares similarities). A *CV* below 20% indicates that those set of data is homogeneous and does not deviate from the mean.

2.5. Test for publication bias

The impact of possible publication bias was also analysed in cases where the number of studies was appropriate (k > 10), using funnel plots and a combined tandem method, as suggested by Ferguson and Brannick (2012) and used by Gates et al. (2017). This method includes Egger's regression test, in which significant findings suggest publication bias (Egger & Davey Smith, 1998), the trim-and-fill method (Duval & Tweedie, 2000), and Orwin's Fail Safe N (Ferguson & Brannick, 2012). When all three criteria were met, indicating the presence of bias, publication bias was deemed "probable"; when one or two criteria were met, bias was deemed "possible"; and when no evidence of bias was found, publication bias was deemed "unlikely".

To investigate publication bias for single-case studies, the correlation between standard error and observed effect size was calculated. We performed this analysis because in small studies (with a large standard error), only very large observed effect sizes are statistically significant and thus more likely to be published,

resulting in a positive correlation between standard error and effect size (Egger & Davey Smith, 1998)

3. Results

3.1. Descriptive characteristics

43 of the studies met the inclusion criteria. Several studies were selected by two or more screening reviews of the specific social-communication skills, since the same study could have measured the effect on more than one outcome measure (Fig. 1). A description of the studies included in the meta-analysis can be found in Appendix C (see Supplemental material). In the group-design studies, a total of 669 participants, mean age 41.2 months (range 24.6–56.2), were included in the experimental intervention groups, and a total of 617 participants, mean age 41.5 months (range 27.5–59.7), were included in the control groups; the percentage of male participants ranged from 75.0–91.7%. The single-case design studies included a total of 116 participants (range 1–16) with a mean age of 43.2 months (range 30–72); the percentage of male participants ranged from 50 to 100%. Total treatment dosage ranged from 8 to 259 h in the group-design studies and from 5.0–40.2 h in the single-case design studies.

3.2. Overall analysis

Interventions on social communication skills showed positive outcomes in the 18 group-design studies reviewed. The individual effect sizes, covering a total of 669 participants, ranged from g = 0.10 to g = 1.54, with positive effects indicating increases in ratings of social and communicative competence (see Table 1). Fig. 2 shows the individual effect size for this analysis (g = 0.51, K = 18, 95% [CI 0.37, 0.65], Z = 7.22, p < 0.001). This was a medium effect. The I^2 (23.01) value was nontrivial, and there were at least 10 studies included. Exploratory moderator analyses were conducted (see Moderator analysis section).

Interventions on social communication skills showed positive outcomes in the 25 single-case design studies reviewed. The individual effect sizes, covering a total of 116 participants, ranged from NAP = 0.58 to NAP = 1.00, with positive effects indicating increases in ratings of social and communicative competence. Fig. 3 shows the individual effect size for this analysis. Participants who received treatment made significant improvements in social and communicative abilities (NAP = 0.86, K = 25, 90% [CI 0.59, 0.98], Z = 62.12, p < 0.001). This was a medium effect.

3.3. Publication bias analysis

Evidence of publication bias was found by Egger's regression test (b = 2.07, p < 0.01), showing asymmetry in the funnel plot graph (see Appendix E on Supplemental material). Publication bias was not evident according to the trim-and-fill analysis, where six studies were moved to the right of the mean, making the adjusted effect of intervention smaller (g = 0.42, 95% [CI 0.29, 0.54]), though still significantly different from zero. In contrast, no evidence of publication bias was found according to the Fail-Safe N (99 studies) method. The combined tandem criteria suggested that there was a possibility of publication bias.

For single-case designs, no significant correlation was found between standard error and effect size, indicating the absence of publication bias (r = 0.07, p = 0.72).

3.4. Targeted skills analysis

The effect size (g) for imitation group-design studies ranged from g = 0.11 to g = 0.91, and the effect size for single-case design studies ranged from NAP = 0.79 to NAP = 1.00 (see Table 1 and Appendix F and G on Supplemental material). The sample of studies was not sufficiently large and I^2 statistic (6.62) did not meet the criteria to proceed with moderator or publication bias analyses.

The effect size (g) for joint attention group-design studies ranged from g = 0.21 to g = 1.26, and the effect size for joint attention single-case design studies ranged from NAP = 0.67 to NAP = 1.00 (see Table 1 and Appendix F and G). While the number of studies included in the analysis was sufficient, the I^2 statistic (19.83) was not large enough to proceed with moderator analyses. No evidence for publication bias was found using the tandem method, suggesting that such bias was unlikely.

The effect size (g) for play group-design studies ranged from g = 0.01 to g = 2.12, and the effect size for play single-case design studies ranged from NAP = 0.72 to NAP = 1.00 (see Table 1 and Appendix F and G). The I^2 statistic (73.56) was sufficient to support exploratory moderator analyses but the sample of studies was too small to allow for this. As a result, neither publication bias nor moderator analyses were conducted.

3.5. Caregivers/teachers included in the treatment programme analysis

Nine studies with group design included caregivers or teachers as active components in treatment programmes. The effect sizes for programmes where, in addition to the main therapist, the caregivers or teachers had an active role in the intervention, ranged from g = 0.11 to g = 1.02. Fig. 4 shows the individual effect size for this analysis (g = 0.50, K = 9, 95% [CI 0.32, 0.68], Z = 5.39, p < 0.001). This was a medium effect. The sample of studies was not sufficiently large and the I^2 statistic (0.00) did not meet the criteria to proceed with moderator or publication bias analyses.

Seven studies with single-case design included caregivers or teachers in treatment programmes. The effect sizes for these programmes ranged from NAP = 0.75 to NAP = 0.99. Fig. 4 shows the individual effect size for this analysis (NAP = 0.89, K = 7, 90% [CI 0.66, 0.99], Z = 62.83, p < 0.001). This was a medium effect. The sample of studies was not sufficiently large to proceed with publication bias analyses.

3.6. Moderator analysis

Metaregression analyses of moderators were conducted for the study as a whole. All the descriptive moderators (overall cognitive ability, verbal ability, and treatment dosage) were nonsignificant, except preintervention age (Table 2), which did prove to be significant, Q (1) = 6.95, p = 0.008. Effect sizes were greater when participants' preintervention ages were lower (see Appendix H for more information). Treatment dosage increased with increasing participants' age, although this relationship was not significant (r = 0.271, p = 0.076). In addition, when we eliminated the two studies where treatment dosage was much higher than the rest of the studies, the relationship between the dosage and the effect was significantly positive (see Fig. 6 in Appendix I).

The correlation analyses for single-case studies showed significant effects according to the treatment dosage moderator (Table 2), with the effect being greater when the treatment dosage was increased (number of sessions X hours). The correlations did not show effects in terms of age, IQ or language (see Appendix I for more information).

Correlation analyses were performed with treatment dosage and age of intervention (significant moderating variables of the effect of the intervention). The correlation was positive, but not significant. In addition, when eliminating the two outlier studies, the significance of the model was reduced (r Pearson = 0.199, p = 0.206).

Table 1
Aggregate effect for all analysis.

Meta-Analysis	*k*		Total *N*		Effect size		*Q*	I^2
	GD	SCD	GD	SCD	GD	SCD		
Overall	18	25	669	116	0.51**	0.86**	21.11	23.01
Caregiver/teacher	9	7	253	29	0.50**	0.89**	5.15	0.00
Imitation	4	7	77	27	0.43**	0.89*	3.21	6.62
Joint Attention	14	10	396	44	0.55**	0.85*	16.14	19.83
Play	7	9	160	25	0.47**	0.81**	28.08	73.56

Note. *Q* and I^2 were only calculated for group design studies.
GD: group design; SCD: single case design.
* $p < 0.05$.
** $p < 0.001$.

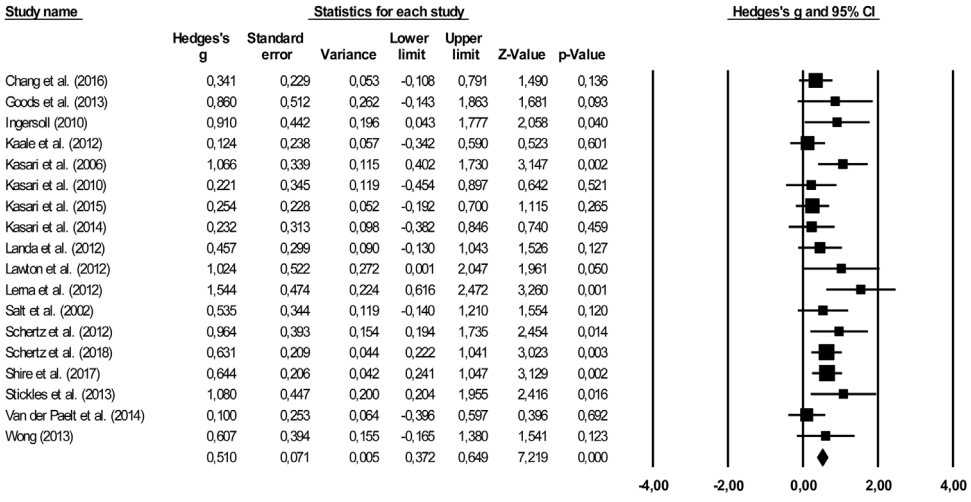

Fig. 2. Overall effect sizes for all measures of social and communicative skills from group design studies. All effect sizes are Hedges' g. 2b.
* The size of the icons represented by the Hedge's g values represent the weight of the study within the meta-analysis
In some studies, the effect outcome measures of the different skills were aggregated to obtain an overall effect of the studies. To see the analysis of the specific outcome measure in each study, see Appendix F.

Table 2
Meta-regression for group design studies and correlations for single case design studies according to the intervention features used in the social-communicative studies.[a]

Intervention feature	Meta-regressions for group design studies in the overall analysis			Correlations for single case design studies in the overall analysis		
	No. Studies	Effect size (95% CI)	*p* value	No. Studies	rPearson	Sig.
Pre-intervention age	24	**−0.02 (−0.03-0.00)**	**0.008**	31	−0.268	0.144
Pre-intervention IQ	14	0.00 (−0.02–0.03)	0.678	17	0.062	0.813
Pre-intervention exp. language	11	0.00 (−0.02–0.02)	0.946	7	0.228	0.712
Pre-intervention rec. language	11	0.00 (−0.01–0.02)	0.620	5	0.219	0.705
Treatment dosage (total hours)	16	0.00 (−0.00–0.01)	0.366	27	**0.380**	**0.050**

CI: confidence Interval. Exp.: expressive. Rec.: receptive.
[a] Pooled effect sizes were estimated from random-effects meta-regression models including the indicator variables for each intervention feature category for group design studies. Significance was estimated using correlation models that included the indicator variables of each intervention feature category for single case design studies.

In studies with larger effect sizes, the mean age of the participants was 45.6 months (*SD* = 13.4, *CV* = 29.5%). The mean IQ of the participants was 22.1 months (*SD* = 9.53, *CV* = 43.07%). The mean of expressive and receptive language was 19.7 and 19.8 months respectively (*SD* = 7.1, *CV* = 35.8%, *SD* = 8.6, *CV* = 43.4%). Finally, the total average of intervention hours was 33.2 (*SD* = 44.0, *CV* = 132.5%) (See Appendix J for more information). In 45.5% of these studies, parents and teachers played an active role in the intervention programmes.

4. Discussion

This study sought to contribute to the literature that evaluates the effectiveness of FIPs in terms of enhancing specific social and communication skills among young children with ASD. The results suggest that intervention programmes in experimental studies, having a group- or a single-case design, which focus on improving social-communication skills, produce medium positive effects. Specifically, such medium positive effects are encountered in FIPs where parents and/or teachers participate actively alongside the main therapist. Furthermore, the effect of the intervention is increased among participants who start participating in FIP programmes at an early age. FIPs have shown a positive effect on the development of communication skills. Therefore, the use of FIPs would allow professionals and parents to choose these types of programmes for the development of skills of children with ASD, since they produce positive effects, reducing costs and waiting times, two fundamental aspects in the satisfaction of parents and professionals of children with ASD (Bejarano-Martín et al., 2019). The results obtained of the overall effect size of FIPs are similar to other

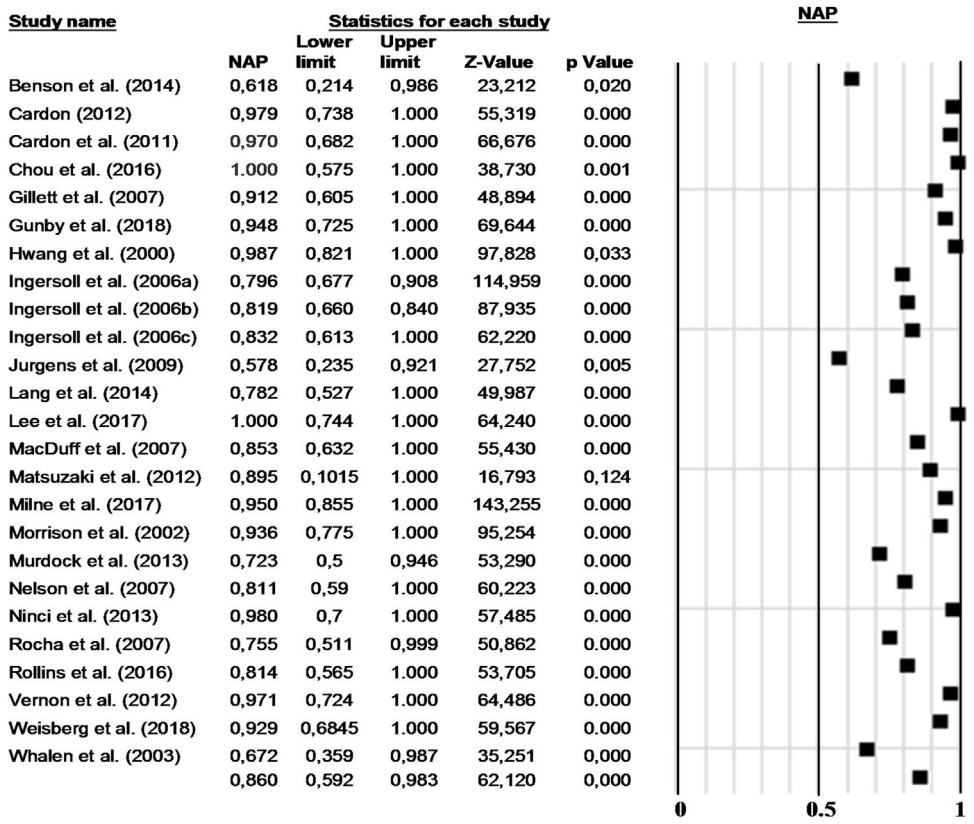

Fig. 3. Overall effect sizes for all measures of social and communicative skills from single case design studies. All effect sizes are NAP.
* In some studies, the effect outcome measures of the different skills were aggregated to obtain an overall effect of the studies. To see the analysis of the specific outcome measure in each study, see Appendix G.

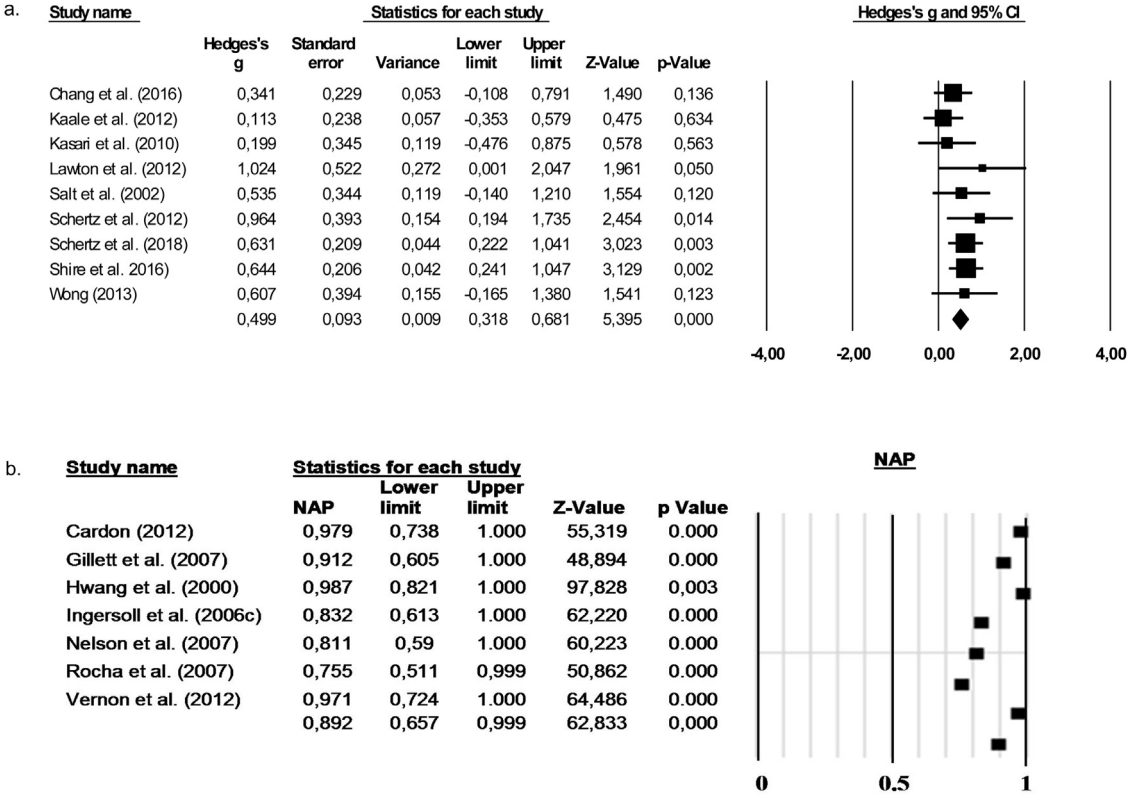

Fig. 4. (a) Effect sizes for programs in which caregivers play an active role from group design studies. All effect sizes are Hedges' g. (b) Effect sizes for programs in which caregivers play an active role from single design studies. All effect sizes are NAP.
* The size of the icons represented by the Hedge's g values represent the weight of the study within the meta-analysis.

intervention programmes studied extensively in the literature (e.g. Applied Behavior Analysis (ABA), Denver Model, PECS).

4.1. FIPs' efficacy to improve social and communication skills in children with ASD

The effect size found in the overall meta-analysis of FIPs (*g*= 0.52) was comparable to that of other meta-analyses of social and communicative interventions to date (*g*= 0.51, Gates et al., 2017; *g*= 0.47, Reichow et al., 2012). Gates et al. (2017) analysed the effectiveness of social communicative interventions from ages 5–20 years. This study examined the effectiveness of interventions for children 6 years of age and younger and obtained similar results. The effectiveness of social communicative interventions has thus been demonstrated from the first stages of development to the beginning of adulthood.

In single case design studies, the overall effect of FIPs was a medium effect (NAP = 0.86). These results were similar to those obtained by Schneider, Goldstein, and Parker, (2008) (percentage of all nonoverlapping data (PAND) = 0.91). NAP and PAND are similar and they can be directly compared (Parker et al., 2009; Rakap, Snyder, & Pasia, 2014). These results could not be compared with the results obtained in the group design studies, since the NAP and Hedge's measurements are not directly comparable. Future studies should analyse both designs with comparable measures to know which type of FIPs designs obtain better results.

In addition to finding an overall effect of FIPs on social-communication skills, it is possible that, by aggregating the data, important information about each specific skill in the studies could be lost. Therefore, an individualized analysis of each skill was carried out by calculating the effect sizes for each of the different types of social-communication skills. Although the effects were positive for all social-communication skills, the results for joint attention in the group-design studies were higher than those obtained for play and imitation skills, with medium effect sizes. In single case studies, the results of joint attention and imitation were higher than those obtained for play skills. These joint attention results are consistent with those reported by Murza et al. (2016). This is particularly noteworthy because joint attention is one of the main deficits in children with ASD, and is one of the skills which have the strongest relationship with the development of communication and cognitive development at later ages (Charman et al., 2003; Mundy et al., 2007; Poon, Watson, Baranek, & Poe, 2012). In play skill there were two studies with higher effect size. We found that the dependent variable in these studies (Cooperative play) is less specific than those specified in the other studies (functional play, symbolic play). Also, the authors measure it as the seconds that the child plays with the adult. By having such a general variable and measuring it as seconds instead of frequencies, the authors could get very good results. Therefore, the results of this meta-analysis should be interpreted with caution because there was significant variation among the FIPs used across the different studies.

Randomised group assignment was possible in the majority of group-design studies (19). Further, the general quality standards of clinical studies that included randomisation (or group equivalence in QEDs), blindness, intention-to-treat analysis, and the use of prospective -as opposed to retrospective- designs, were consistently observed in all the group design studies through the quality selection procedure (EBP inclusion criteria). Therefore, all the group design studies included in the analyses met the standards of methodological quality. Similarly, all single case design studies included in the analysis met the methodological quality standards (EBP inclusion criteria) based on Wong et al. (2015). These quality standards are similar to those proposed by Romeiser Logan, Hickman, Harris, and Heriza, (2008). However, several studies, both group and single case designs, did not carry out a post evaluation of measures, such as cognitive development, language level, or level of symptomatology, which were only recorded at baseline. Despite this, it was possible to perform a meta-analysis of the random effects by partially compensating for the effect sizes, which were calculated only for the social communication skills.

The level at which the intervention was implemented was not considered as a moderator in this meta-analysis. However, one of the inclusion criteria for the selection of studies in phase two had to ensure the description of the control group. This guaranteed that this group did not follow any specific programme or follow a "treatment as usual". However, the "treatment as usual" conducted in some studies could not be controlled, since some studies did not report this information. Despite this, most studies reported that in these types of programmes followed in the control group, the same principles of intervention group were not followed.

Although presenting a measure of treatment fidelity was not a criterion for inclusion in the selection of studies, all intervention programmes presented optimal fidelity measures (>80% or Inter Correlation Coefficient (ICC) >0.80) (See Appendix C). Only two studies with group design and two studies with single case design did not report the specific data of treatment fidelity in their studies, although they reported having complied with the programme's fidelity standards. Therefore, we can attest that the therapists collaborating in the studies conducted the intervention programmes as they were designed.

4.2. Effects of participant and FIP characteristics

Metaregression of moderator analyses of group-design studies has afforded evidence of the importance of considering participants' characteristics in order to improve the effect of the programmes implemented. These results support the suggestions of other studies (see Siu et al., 2016) that interventions should be implemented as soon as possible. However, these data show that, while the increase in treatment dosage had a positive impact on the effect of the intervention, the results were not statistically significant. These results are consistent with Virués-Ortega (2010). In addition, when considering the range of treatment dosage (10–259 h), it is still unclear what the most appropriate number of sessions or hours should be for the respective interventions that focused on the development of specific social-communication skills. By eliminating studies with higher standard deviation, the relationship between dosage and effect was significantly positive, consistent with other studies (Eldevik et al., 2010; Linstead, Dixon, French et al., 2017, Linstead, Dixon, Hong et al., 2017). These results should be interpreted cautiously, bearing in mind that the number of hours that children may have received outside the intervention-programme studies is not known.

The correlation analyses of the single-case design studies showed that increases in treatment dosage also led to more effective programme outcomes. However, the lack of a statistically significant relationship between the effect of the programme outcomes in the group-design studies and the duration of the intervention may be due to the great variability in the number of hours employed. In addition, a significant relationship was observed in single-case design studies because the range in the number of intervention hours was narrower.

As indicated above, we have performed a correlation analysis of the treatment dosage with the age of the intervention as these two variables probably have a significant moderating role on the effect of the intervention). These variables correlated positively, as the age of the participants increased, the treatment dosage increased, but not significantly, and the significance of the model decreased when we eliminated two studies with outliers (disproportionately high).

The descriptive analyses of the moderators in the selected studies with larger effect sizes showed CVs greater than 20%. This shows us that in these studies no similarities were found in terms of age, IQ, and language of the participants, or in treatment dosage, that is, no homogeneity was found in the studies. Future studies should analyse possible similarities of different FIPs, in order to find those characteristics of the participants or programmes that produce the greatest effects.

Although it has been found that these variables influenced the effect of the intervention, it was not possible to verify whether FIPs produced indirect effects on the language, level of symptomatology, adaptive behavior and / or IQ because the studies did not provide information on these aspects in the postintervention phase. In these studies, FIPs focused on the improvement of communication and social skills, so it appears that they did not consider it necessary to report information on measures about the characteristics of participants postintervention. Therefore, it was not possible to evaluate whether the intervention programmes had an indirect impact on these variables. Future studies should analyse the indirect effect of FIPs on this type of variables, so that they are taken into account when selecting the type of programme.

4.3. Effectiveness of FIPs when families and/or teachers actively participate in the intervention

Pooled effect sizes of FIPs were significant where, in addition to the therapist, parents, caregivers or teachers participated in the intervention. These results are in line with the results of other studies where parents' or caregivers' role in the intervention was evaluated (Estes et al., 2014; Pickles et al., 2016), and which also provide evidence of a reduction in family stress levels (Keen, Couzens, Muspratt, & Rodger, 2010). The participation of parents or caregivers has been proposed as one of the key intervention components by numerous studies (Casagrande & Ingersoll, 2017; Zwaigenbaum et al., 2015), since these are the persons with whom children with ASD spend most of their time. Considering the priorities and preferences of caregivers produces positive results (Leadbitter et al., 2017), as they have a better understanding of the challenges that the child faces in his/her daily life. In addition, the inclusion of caregivers would most likely lower the costs incurred by public services, by reducing the number of intervention hours that therapists devote each child. Accordingly, giving caregivers an active role would not only help the child with ASD, but also lead to a reduction in long-term parental stress and the other ASD-related burdens (Keen et al., 2010). Programmes that included caregivers in the intervention were selected for this study, regardless of the number of hours devoted by parents/caregivers to the task or the way where they participated in the intervention.

4.4. Implications for research and clinical practice

The following are some of the implications of this work for professional practice, as this study is part of a European Union project (ASDEU, 2020) on Autism Spectrum Disorders, which one of the main objectives was to promote the improvement of early intervention programmes in the European context.

Firstly, the use of FIPs in early socio-communication skills interventions has resulted in a medium positive effect. Therefore, the use of FIPs has proven its effectiveness. An average positive effect is an indicator of sufficient quality to recommend the use of a programme. FIPs are not only based on scientific evidence (Odom, Boyd et al., 2010, Odom, Collet-Klingenberg et al., 2010), but they have also shown to be effective in the development of socio-communication skills. Professionals and service providers should take into account the use of these programmes in a clinical context, as they have demonstrated positive effectiveness for children between 0 and 6 years.

Second, we suggest that the implementation of early intervention programmes be based on a sufficient amount of information about the characteristics of each participant and that this information be taken into account in the publication of results. This information should include pre - post measures of behaviour, cognition, communication and social functioning that would allow the effectiveness of the intervention, and its applicability to different groups of children, to be evaluated that subsequently would allow to compare the results with other programmes that pursue similar objectives and/or replicate the same methodology. Likewise, the professionals directly involved must take into account the characteristics of the children for whom they are going to develop an intervention programme in relation to the children who have participated in studies that prove the effectiveness of the programme. In this way, they would be able to select with greater precision the most potentially effective and feasible procedures.

Third, in relation to the context of the intervention, assuming that: (a) the effect of the intervention is significantly positive when parents are actively involved in well-structured activities, and (b) that it may be more effective if carried out in the child's natural environment, it is desirable that both intervention programmes and studies on treatment effectiveness clearly define the role of caregivers as active agents with interests in the process (Webb, Jones, Kelly, & Dawson, 2014). In addition, according to Leadbitter et al. (2017), intervention programmes and effectiveness studies should specify the type of relevant information or training that parents should receive, considering both the total number of hours that family members should devote to learning the techniques they must use to teach specific skills, and the time they should devote to conducting teaching activities themselves. In addition, it is important to keep in mind the fidelity of the intervention, something that few studies provide with concrete measures to achieve such fidelity specifically in relation to the application of the procedures by the parents (most studies only provided information on how they ensured the fidelity of treatment for therapists). It is also expected that assigning an active role to parents will also help reduce their long-term stress levels (Keen et al., 2010).

Forth, since various targeted interventions have provided evidence of efficacy, efforts can be made to make these techniques complementary to early detection programmes for signs of ASD risk. If screening programmes identify delays or deviations in skills such as eye contact, play, imitation, or joint attention, it is possible to implement FIPs aimed at those hindered skills, actively involving families to begin working with their children, even before they receive a formal diagnosis of ASD. This would make early detection programmes more useful and socially valid. FIPs will increase the success of screening programmes and reduce the symptoms of ASD (MacDonald et al., 2014; Orinstein et al., 2014).

Finally, increasing the dose of treatment may heighten the effect of the intervention. Therefore, policy makers, as well as intervention services, should seek ways to provide resources to families and professionals so that they can initiate and sustain treatments for as long as possible (duration of intervention in weeks/months/years) and as intense (hours per week) as possible.

4.5. Limitations and future research

Evidence of possible publication bias was found in the overall analysis of the studies, suggesting that the "true" effects may be smaller than what has been reported in the literature to date. There is the possibility that studies with a small effect size are not published, which, if true, would support the contention that the most relevant aspect of the study is to demonstrate that the particular FIP was studied in a sufficiently large sample. This evidence of pos-

sible publication bias was shown with a combined tandem method as this makes it easier to visually identify publication bias (Terrin, Schmid & Lau, 2005). Even so, the limited sample size of most studies suggests that evidence of publication bias may simply be the by-product of small-sample-size studies rather than genuine publication bias (Whitehead, 2002). No evidence of publication bias was found in correlation analyses of single case design studies. However, given the nature of these small studies, with subsequently high standard errors, in which multiple subjects participated with varying time intervals before and after the intervention, these results should be taken with caution. Consequently, more studies with larger number of participants are needed to increase their practical relevance.

In those studies that reported results in more than one outcome measure, the effect size was calculated by aggregating these measures to obtain a global effect in each study. These analyses allowed to obtain an overall effect. However, adding the individual effects to a global effect could lead to a loss of information from the data. Therefore, individual analyses of each of the outcome measures were conducted, and it was possible to conduct an analysis of the effect of the FIPs for imitation (n = 11), joint attention (n = 25) and play (n = 15) skills. The total number of studies found for skills such as eye contact (n = 6) and gestures (n = 2) was low. The outcomes of specific skills must thus be interpreted with caution. As a consequence, a meta-analysis could not be performed to determine the effect size in the case of the above skills.

In addition to the relatively small number of studies, analysis of the outcomes of the interventions reviewed was also limited, due to the diverse number of methods and designs used in the different papers. This limitation was further exacerbated by the degree of variability in the characteristics of each intervention programme and the characteristics of the children reported by each study (French & Kennedy, 2018; Spreckley & Boyd, 2009). In order to study the moderating effect of contextual factors, additional research studies are required to better describe the characteristics of the participants and the intervention settings.

Future research should examine whether parents' participation is equally important for teaching different types of skills or whether such active participation is more relevant for developing and/or evaluating the acquisition of specific skills. A further area to be addressed is the possible influence that parents' involvement in the intervention may have on their degree of satisfaction with the programme, as suggested by some studies (Bearss, Burrell, Stewart, & Scahill, 2015; Gulsrud, Hellemann, Shire, & Kasari, 2016). Research should be undertaken to define the best procedure to be implemented for the purpose of fostering the active participation of the various stakeholders involved. In this regard, an evaluation of the reproducibility of both the teaching methods and the support and reinforcement required, would facilitate learning and the generalisation of the procedures by such stakeholders (Debodinance et al., 2017).

5. Conclusion

This study provides information that may prove useful for debating whether currently available Focused Intervention Practices (FIPs) aimed at improving the social communicative skills generally affected in young children with ASD (imitation, joint attention and play), are effective, adequate, and suitable enough to be used as treatment in early-intervention services. Our research confirms that studies using FIPs to improve social-communication skills show promising results, and therefore supports the contention that caregivers and/or teachers could play an active role in FIPs when it comes to obtaining positive effects. Moreover, active participation by caregivers or teachers could reduce the costs incurred by public services, by reducing the number of intervention hours that therapists devote to each child. In addition, since these FIPs focus on specific skills, application time would become shorter, something that could, in turn, lead to lowering the cost of treatment and thus rendering it more affordable. Future research should provide results that would allow for their inclusion in services aimed at the identification, diagnosis and treatment of early neurodevelopmental disorders, by identifying the characteristics, which increase the intervention's chances of success.

Author contribution

ABM, RCB, MMM y CFA designed the study; ABM y RCB wrote the manuscript; ABM, SLJ, ES, AV, CR, CC conducted the literature review and executed the quality review process; ABM and MMM extracted the data from the selected articles; ABM, MMM and RCB carried out statistical analyses and interpreted the results; MMM, CFA, PGP and MP collaborated in writing the manuscript. All authors have read and approved the final manuscript.

Declarations of interest

All authors declare they have no conflicts of interest.

Ethical approval

This article does not contain any studies with human participants or animals performed by any of the authors.

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Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.ecresq.2020.01.004.

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Appendix A. Inclusion criteria of the screening selection stages

Title Review Form: Focused intervention practised

First author, Year: _____ ID: _____ Initials: __ __ __

1. The study focus on ASD early intervention on the prelinguistics skills (imitation, eye contact, joint attention, gestures, pointing and play)?	Yes	No
2. The study **does not** address? ▪ Drug therapy ▪ Biomedical/biotechnological/pharmacological interventions ▪ Genetics ▪ Medications ▪ Medical trials ▪ Biochemical processes • Comorbid conditions	Yes	No
3. The study **is not** a systematic review or meta-analysis?	Yes	No

*All questions have to be YES to pass to the next stage

Abstract Review Form: Focused intervention practised

First author, Year: _____ ID: _____ Initials: __ __ __

1. Does the study address early intervention on prelinguistics skills (imitation, eye contact, joint attention, gestures, pointing and play) for ASD?	Yes	No
2. Is the study original research (peer reviewed; not included systematic review or meta-analysis)?	Yes	No
3. Does the study include individuals in the target population? (Children aged 1 month to 6 years).	Yes	No

*All questions have to be YES to pass to the next stage

Full Text Review Form: Focused intervention practised

1. Does the study address early intervention on prelinguistics skills (imitation, eye contact, joint attention, gestures, pointing and play) for ASD?	Yes	No
2. Is the study original research (peer reviewed; not included systematic review or meta-analysis)?	Yes	No
3. Does the study include individuals in the target population? (All children must be between 1 and 6 years).	Yes	No
4. Does the study include individuals with ASD diagnosis according to DSM-IV/5 or ICD-10 or individuals with risk of ASD? (All children must be an ASD diagnosis or to have ASD risk)	Yes	No
5. Does the study provide dates about the intervention on at least one of the following ways? a. Through general standardized measures of development or IQ, like (Merril-Palmer, WIPPSI-IV…); Vineland (Social and adaptative skills), ADOS (Communication and social skills); scales of daily living skills; scales of motor development; etc. b. Trough quantitative data of specifics evaluations of the skills to work in the intervention (Frequency/percentage/steps completed of acts of imitation, eye contact, joint attention, gestures, pointing, play).	Yes	No
6. Does the study provide baseline information, intervention and post-intervention/generalization and/or follow-up results?	Yes	No
7. Is the study a? a. Experimental design or Randomized controlled trial b. Quasi-experimental design	Yes	No

*All questions have to be YES to pass to the next stage

Appendix B. References included in the meta-analysis

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Appendix C. Descriptive information for each study

Table 1. Summary of the studies included in the articles. Group design

Focused skill	Study	Tx n	Cnt n	% Male	Mean age (months)		Symptoma-tology		Cognitive development		Language			Social-adaptive		Intervention program					
					Tx	Cnt	Test	Score	Test	Score	Test	Exp.	Rec.	Test	Score	Type of treatment	Professional profile	Fidelity	Parents participation	Intensity (h/week)	Duration (weeks)
Imitation	Landa et al. (2012)	24	24	83.3	28.6	28.9		-	1	27.5[a]	2	23.92[a]			-	Discrete trial teaching (DTT) and Pivotal Response training (PRT)	-	87%	1.5 hours	10	6
	Ingersoll (2010)	11	10	90.9	41.4	37.2		-	2	21.73[c]		-			-	Contingent imitation	-	90%	-	3	10
	Salt et al. (2002)	12	5	91.7	42.4	37.7		-	2	17[c]	4	4.73[a]	62.55[a]	1	49.5[b]	Contingent imitation	Therapist and parents	-	1 hour	4	44
	Van der Paelt et al. (2016)	30	35	80	51.8	49.9	2	5.97[a]		-	1	27.03[a]	29.27[a]	1	61.2[a]	Contingent imitation	-	90%	-	259 (Total)	-
Joint Attention	Wong (2013)	14	9	85.7	56.2	59.7	3	35.93[a]	1	36.25[c]	2	29.73[c]	38.55[c]		-	DTT plus social interaction	Teachers	>80%	4 weeks	2	4
	Kaale et al. (2012)	34	27	76.5	47.6	50.3		-	1	25.6[c]	1	18.8[c]	21[c]		-	DTT plus social interaction	Teachers	85%	-	2	8
	Kasari et al. (2014)	60	52	83.3	41.9	42.8	2	7.2[a]	1	23.6[c]	2	20.1[c]	22.1[c]		-	DTT plus social interaction	-	>80%	Parents give intervention	21	12
	Schertz et al. (2013)	11	12	-	24.6	27.5	1	11[a]		-	2	24.6[c]	21[c]		-	Mediated learning with active engagement	Parents	92%	30 minutes	-	-
	Landa et al. (2012)	24	24	83.3	28.6	28.9		-	1	27.5[a]	2	23.92[a]			-	DTT plus PRT	-	87%	1.5 hours per month	10	6
	Kasari et al. (2006)	20	17	75	43.2	41.9		-	1	26.29[c]	1	20.6[c]	20.55[c]		-	DTT plus social interaction	-	92%	-	-	6
	Kasari et al. (2010)	19	19	78.9	30.3	31.3		-	1	19.83[c]		-			-	DTT plus social interaction	Psychologist and parents	ICC 0.86	10 minutes	2	8
	Lerna et al. (2012)	9	9	-	38.8	41.1	1	16.78[a]	3	73.56[b]		-			-	Picture exchange system	-	-	-	1.5	24
	Lawton et al. (2012)	9	7	-	46	43		-	1	30.3[c]		-			-	DTT plus social interaction	Teachers	ICC 0.90	11 half-hour sessions	-	-
	Salt et al. (2002)	12	5	91.7	42.3	37.7		-	2	17[c]	4	4.73[a]	62.55[a]	1	49.5[b]	Contingent imitation	Therapist and parents	-	1 hour	4	44
	Van der Paelt et al. (2016)	30	35	80	51.8	49.1	2	5.97[a]		-	1	27.03[a]	29.27[a]	1	61.2[a]	Contingent imitation	-	90%	-	259 (Total)	-
	Shire et al. (2017)	56	59	78.6	31.7	31.5		-		-	2	17.32[c]	16.44[c]		-	DTT plus social interaction	Teachers and psychologist	91.7%	6-7 weeks of training	3.5	10
	Chang et al. (2016)	38	28	78.9	48.9	51.6		-	1	34.53[c]	2	32[c]	30.76[c]		-	DTT plus social interaction	Teachers	86.9%	8 weeks of training	-	8
	Schertz et al. (2018)	73	71	79.5	24.5	24.8	2	16.36[a]	-		2	26.7[a]	25.3[a]		-	Mediated learning with active engagement	Therapist and parents	99%	30 minutes per session	1	32

Gestures	Goods et al. (2013)	7	8	-	48.7	54.7		-	1	17.21[c]	1	13.63[c]	12.14[c]		-	DTT plus social interaction	Psychologist	88.3%	-	1	12
Play	Kasari et al. (2006)	21	17	75	42.7	41.9		-	1	24.55[c]	1	21.43[c]	21[c]		-	DTT plus social interaction	Psychologist	92%	-	-	6
	Goods et al. (2013)	7	8	-	48.7	54.7		-	1	17.21[c]	1	13.63[c]	12.14[c]		-	DTT plus social interaction	Psychologist	88.3%	-	1	12
	Kasari et al. (2015)	48	43	81.4	30.7	32.3		-	1	68[b]	1	14.09[c]	16.09[c]		-	DTT plus social interaction	Psychologist	91.4%	10 hours – 10 hours	1	10
	Lerna et al. (2012)	9	9	-	38.8	41.1	1	16.78[a]	3	73.56[b]		-			-	Picture exchange system	-	-	-	1.5	24
	Van der Paelt et al. (2016)	30	35	80	51.8	49.1	2	5.97[a]		-	1	27.03[a]	29.27[a]	1	61.2[a]	Contingent imitation	-	90%	-	259 (Total)	-
	Chang et al. (2016)	38	28	84.2	48.9	51.6		-	1	34.53[c]	2	32[c]	30.76[c]		-	DTT plus social interaction	Teachers	94.2%	8 weeks of training	-	8

Tx: Treatment. Cnt: Control. Exp: Expressive. Rec: Receptive. ICC: Inter Correlation Coefficient. [a] Raw score. [b] Composite score. [c] Months. (-): Not reported.
Test symptomatology: 1- Childhood Autism Rating Scale (CARS) (Schopler, Reichler & Rochen, 1988); 2- Autism Diagnostic Observation Schedule (ADOS) (Lord et al., 2000); 3- Gilliam Autism Rating Scale (GARS) (Swift, 1999).
Test cognitive development: 1- Mullen Scales of Early Learning (Mullen, 1995); 2- Bayley Scales of Infant Development (Bayley, 1993); 3- Griffiths Mental Development Scale (Griffiths, 1996)
Test language: 1- Reynell III (Edwars et al.,1997); 2- Mullen Scales of Early Learning (Mullen, 1995); 3- Preschool Language Scales (Zimmerman, Steiner & Pond, 2002); 4- MacArthur-Bates Communicative Development Inventories (Fenson et al., (2007)
Test social-adaptive: 1- Vineland Adaptive Behaviour Scales (Sparrow, Balla & Cicchetti, 1984; Sparrow, Cicchetti, Balla, 2005)
Studies that reported data from the same population in different follow-up periods (Kaale, Morten, Fagerland, Martinsen & Smith, 2014, Kasari, Paparella & Freeman, 2008, Lerna, Esposito, Conson & Massagli, 2014) were analysed within the original study as a single study.

Table 2. Summary of the studies included in the articles. Single Case design

Focused skill	Study	Tx n	% Male	Mean Age (months)	Symptomato-logy		Cognitive development		Language				Social adaptive		Intervention program				Mean Treatment dosage (hour per session)	
					Test	Score	Test	Score	Test	Total	Exp.	Rec.	Test	Score		Professional profile	Fidelity	Parents participation	Baseline	Tx + Follow-up
Imitation	Matsusaki et al. (2012)	1	100	40		-	3	5[c]		-				-	Pivotal response training (PRT)	Psychologist	-	-	3 (1)	14 (1)
	Cardon et al. (2011)	6	100	34.2	1	37.5[a]		-	9	67.5[a]			1	76.3[b]	Video modelling	-	99%	-	6.3 (0.5)	14 (0.5)
	Cardon (2012)	4	50	36	1	35[a]		-	3		78[b]	72[b]	1	83.2[b]	Video modelling	Parents	>80%	2 hours of training	6.5 (0.5)	12 (0.7)
	Ingersoll et al. (2006a)	5	80	37	1	35[a]	1	20[c]	4	16[a]	41[a]			-	PRT and contingent imitation	-	98%	-	47 (0.3)	87 (0.3)
	Ingersoll et al. (2006b)	5	100	41	1	38[a]	1	24[c]	4	23[c]				-	PRT and contingent imitation	-	99.5%	-	11 (1)	28 (1)
	Ingersoll et al. (2006c)	3	66.6	37	1	32[a]	1	17[c]	4	11[c]	9.7[c]	12[c]		-	PRT and contingent imitation	Parents	>80%	40 min. per session	10 (0.2)	23 (1)
	Hwang et al. (2000)	3	100	37		-	2	15[c]	6		9.7[c]	11.7[c]		-	PRT	Special Education Teachers	90%	20 hours of training	19 (-)	38 (-)
Eye Contact	Ninci et al. (2013)	1	100	48		-		-	7	38[a]				-	Prompting and reinforcement	-	92%	-	7.7 (0.1)	9 (-)
	Vernon et al. (2012)	3	100	38		-		-		-			1	68[b]	PRT	Parents	97.7%	-	8.3 (1)	16.7 (1)
	Benson et al. (2014)	2	50	43		-		-		-			1	69[b]	Physical and verbal cues	-	88%	-	6 (0.5)	12 (1.5)

	Hwang et al. (2000)	3	100	37		-	2	15[c]	6		9.7[c]	11.7[c]		-	PRT	Special Education Teachers	90%	20 hours of training	19 (-)	38 (-)
	Rollins et al. (2016)	4	100	30	2	22[a]		-	7	12[a]				-	Contingent imitation	Speech therapist	94.7%	10 min. per session	5.5 (0.5)	10.5 (1.5)
	Chou et al. (2016)	2	100	72		-		-		-				-	Token economy and prompting	-		-	11 (1)	4.5 (1)
Joint Attention	Rocha et al. (2007)	3	66.6	32		-	1	14.7[c]	4	14.7[c]				-	Discrete trial teaching (DTT) and PRT	Parents	96.5%	20 min of training	36 (0.3)	58.7 (0.3)
	Matsuzaki et al. (2012)	1	100	40		-	3	5[c]		-				-	PRT	Psychologist	-	-	3 (1)	14 (1)
	Hwang et al. (2000)	3	100	37		-	2	15[c]	6		9.7[c]	11.7[c]		-	PRT	Special Education Teachers	90%	20 hours of training	19 (-)	38 (-)
	Whalen et al. (2003)	5	-	50	1	30,8[a]	1	17[c]	4	16.2[c]				-	DTT and PRT	-	96%	Generalized training	-	10 weeks
	MacDuff et al. (2007)	3	100	48		-		-	3	19.7[c]			1	20,3[c]	DTT and PRT	-	-	-	5.5 per week	5.5 per week
	Ingersoll et al. (2006a)	5	80	37	1	35[a]	1	20[c]	4	16[a]	41[a]		-		DTT and contingent imitation	-	98%	-	47 (0.3)	87 (0.3)
	Benson et al. (2014)	2	50	43		-		-		-			1	69[b]	Physical and verbal cues	-	88%	-	6 (0.5)	12 (1.5)
	Milne et al. (2017)	16	-	50.2		-	-	101[b]	8	-	107[b]	104[b]	1	82.4[b]	PRT	-		-	-	-
	Gunby et al. (2018)	3	66.6	80		-		-		-				-	DTT and PRT	-	90.9%	-	-	-
	Weisberg et al. (2018)	3	100	46.6		-		-	3	12.67[c]	11[c]	15.3[2]	1	-	DTT and PRT	-	99%	-	10 (0.25)	80 (0.25)
Gestures	Ingersoll et al. (2006b)	5	100	41	1	38[a]	1	24[c]	4	23[c]			-		PRT and contingent imitation	-	99.5%	-	11 (1)	28 (1)
Play	Nelson et al. (2007)	4	100	49.5	1			-		-				-	PRT plus visual support	Teachers and Classmates	96%	Training classmates	1 day per week	0.5 per week
	Gillett et al. (2007)	3	100	52	3	101.3[a]		-	8			59[c]		-	DTT and PRT	Parents	96%	Training parents	5.3 (-)	10.7 (-)
	Lang et al. (2014)	3	66.6	43.7	1	35.3[a]		-		-				-	PRT	Speech language pathologist	91%	-	27.3 (0.1)	13.3 (0.1)
	Jurgens et al. (2009)	1	100	43	1	33[a]		-		-				-	Picture exchange system	-	>80%	-	5 (0.3)	21 (0.3)
	Murdock et al. (2013)	4	100	53.3			4	79.75[a]	3	77.7[a]	79[a]	79.2[a]		-	Video modelling	-	97%	-	6.5 (-)	23.7 (-)
	Ingersoll et al. (2006a)	5	-	37	1	35[a]	1	20[c]	4	16[a]	41[a]			-	PRT and contingent imitation	-	98%	-	47 (0.3)	87 (0.3)
	Morrison et al. (2002)	4	50	58.2	1		5	33.5[c]	10		26.7[c]	23[c]	2	21.2[c]	Photographic schedules	-	97%	Voluntaries	12.2 (-)	37.5 (-)
	Lee et al. (2017)	1	100	60		-	-	19[c]	-	17[c]				-	Video modelling	Behaviour specialist	100%	-	35 (-)	15 -)

Tx: Treatment. Exp: Expressive. Rec: Receptive. [a] Raw score. [b] Composite score. [c] Months. (-): Not reported

Test symptomatology: 1- Childhood Autism Rating Scale (CARS) (Schopler, Reichler & Rochen, 1988); 2- Autism Diagnostic Observation Schedule (ADOS) (Lord et al., 2000); 3- Gilliam Autism Rating Scale (GARS) (Swift, 1999)

Test cognitive development: 1- Bayley Scales of Infant Development (Bayley, 1993); 2- Uzgiris-Hunt Scales (Uzgiris & Hunt, 1987); 3- Kyoto Scale of Psychological developmental (Society for the Kyoto Scale of Psychological Development, 2002); 4- Kaufman Brief Intelligence Test-2 (Kaufman & Kaufman, 1999); 5- Battelle Developmental Inventory (Newborg, Stock & Wnek, 1984)

Test language: 1- Reynell III (Edwars et al.,1997); 2- Mullen Scales of Early Learning (Mullen, 1995); 3- Preschool Language Scales (Zimmerman, Steiner & Pond, 2002); 4- MacArthur-Bates Communicative Development Inventories (Fenson et al., (2007); 5- Griffiths Mental Development Scale (Griffiths, 1996); 6- Sequenced Inventory of Communication Development (Lichtenberger, 2008); 7- VB-MAPP: Verbal Behaviour Milestone Assessment and Placement Program (Sundberg, 2008); 8- Peabody Picture Vocabulary Test (Dunn, Dunn & Pearson Assessment, 2007); 9- Vineland Adaptive Behaviour Scales (Sparrow, Balla & Cicchetti, 1984; Sparrow, Cicchetti, Balla, 2005); 10- Battelle Developmental Inventory (Newborg, Stock & Wnek, 1984).

Social-adaptive assessment: 1- Vineland Adaptive Behaviour Scales (Sparrow, Balla & Cicchetti, 1984; Sparrow, Cicchetti, Balla, 2005); 2- Battelle Developmental Inventory (Newborg, Stock & Wnek, 1984).

Appendix D. Techniques found in the studies

Table 1. Techniques found in the studies included in the articles

Skill	Study	Design	Treatment Model	
			Approach	Technique
Imitation	Landa et al. (2011)	Groups	Behavioural	Discrete trial teaching (DTT) plus pivotal response training (PRT) plus routines based interactions
	Ingersoll (2010)		Behavioural, Naturalist and development	Reciprocal Imitation Training (RIT) - Contingent imitation. Therapist models an action, with either an object or a gesture, once a minute on average.
	Van der Paelt et al. (2016)			Improving Parents As Communication Teachers (IMPACT) - Milieu teaching plus contingent imitation. Focussed on spontaneous behaviour elicited by the specific situation.
	Salt et al. (2002)		Developmental	Contingent imitation. Child centred approach prescribed by the child's developmental level.
	Matsusaki et al. (2012)	Single Case	Behavioural	PRT
	Cardon (2012)		Video modelling	The relative is videotaped doing something, the child is taught to perform the same behaviour
	Cardon et al. (2011)		Behavioural, Naturalist and development	Contingent imitation plus video modelling
	Ingersoll et al. (2006a)			
	Ingersoll et al. (2006b)			Reciprocal Imitation Training (RIT) - PRT to elicit target behaviours plus contingent imitation
	Ingersoll et al. (2006c)			
	Hwang et al. (2000)			Contingent imitation with naturally occurring reinforcement
Eye Contact	Ninci et al. (2013)	Single Case	Behavioural	Systematic prompting (verbal and gestural) and reinforcement
	Benson et al. (2014)			Follow the child's preferences. Responding immediately to any behaviour. Use physical and verbal cues and making a gaze shift.
	Rollins et al. (2016)			Follow child's lead. Limit distractions. Reinforcement. Use of animation, gesture, facial expression, and vocal quality. Contingent imitation.
	Chou et al. (2016)			Art program: manipulation of motivating operations. Implementation of a token economy. Prompting procedures.
	Vernon et al. (2012)		Behavioural, Naturalist and development	PRT. Attracting the child's attention to the stimulus. Reinforcement related to the child's verbal attempt at responding.
	Hwang et al. (2000)			Contingent imitation with naturally occurring reinforcement
Joint Attention	Wong (2013) *	Groups	Behavioural, Naturalist and development	JASPER - Milieu teaching plus contingent imitation. DTT. Contingent imitation. Follow child's lead. Systematic modelling and prompting. Active coaching of caregivers. Talking about what the child is doing, repeating back what the child said, expanding on what the child said. Giving corrective feedback. Encouraging eye contact. Making environmental adjustments to engage the child
	Kaale et al. (2012) *			
	Kasari et al. (2014) *			
	Kasari et al. (2006) *			
	Kasari et al. (2010) *			
	Lawton et al. (2012) *			
	Shire et al. (2017) *			
	Chang et al. (2016) *			
	Schertz et al. (2013)			Mediated learning with active engagement. Focusing attention with self-regulation expanding skills.
	Schertz et al. (2018)			
	Lerna et al. (2012)		Developmental	PECS - Visual support based on picture exchange and its discrimination for learning sentences
	Salt et al. (2002)			Reciprocal Imitation Training (RIT) - Contingent imitation. Child centred approach prescribed by the child's developmental level.
	Van der Paelt et al. (2016)			Milieu teaching plus contingent imitation. Focussed on spontaneous behaviour elicited by the specific situation.
	Landa et al. (2011)			DTT plus PRT plus routines-based interactions
				Focusing, organizing and planning, giving meaning, encouraging, and expanding

	Rocha et al. (2007)	Single Case	Behavioural	DTT (provide instruction and feedback) and PRT (Follow child's leads and motivations)
	Matsuzaki et al. (2012)			PRT
	MacDuff et al. (2007)			DTT and PRT. Clear and systematic prompts. Follow child leads. Taking turns. Contingent reinforcement.
	Benson et al. (2014)			Follow child preferences. Responding immediately to any behaviour. Use physical and verbal cues. gaze shift.
	Milne et al. (2017)			Teaching the ability in small steps with demonstration, reinforcement and feedback
	Gunby et al. (2018)			Applied Behaviour Analysis (ABA)
	Weisberg et al. (2018)			Applied Behaviour Analysis (ABA)
	Ingersoll et al. (2006a)		Behavioural, Naturalist and development	Reciprocal Imitation Training (RIT) - PRT to elicit target behaviours plus contingent imitation
	Hwang et al. (2000)			Contingent imitation with naturally occurring reinforcement
	Whalen et al. (2003)			DTT (providing instruction and feedback) and PRT (Follow child's leads and motivations)
Gestures	Goods et al. (2013)	Groups	Behavioural, Naturalist and development	Milieu teaching plus contingent imitation. DTT
	Ingersoll et al. (2006b)	S. Case	Behavioural, Naturalist and development	Reciprocal Imitation Training (RIT) - PRT to elicit target behaviours plus contingent imitation
Play	Kasari et al. (2006) *	Groups	Behavioural, Naturalist and development	JASPER - Milieu teaching plus contingent imitation. DTT. Contingent imitation. Follow child's lead. Systematic modelling and prompting. Active coaching of caregivers. Talking about what the child is doing, repeating back what the child said, expanding on what the child said. Giving corrective feedback. Encouraging eye contact. Making environmental adjustments to engage the child
	Goods et al. (2013) *			
	Kasari et al. (2015) *			
	Chang et al. (2016) *			
	Van der Paelt et al. (2016)			IMPACT - Milieu teaching plus contingent imitation. Focussed on spontaneous behaviour elicited by the specific situation.
	Lerna et al. (2012)		Augmentative and Alternative Communication System	PECS - Visual support based on picture exchange and its discrimination for learning sentences
	Nelson et al. (2007)	Single Case	Behavioural	Visual support. Incidental teaching. Peer mediated teaching. Follow child`s play and motivations. Systematic prompts. reinforcement
	Lang et al. (2014)			Rotation in play locations. Systematic prompting to play appropriately. Reinforcement
	Gillett et al. (2007)		Behavioural, Naturalist and development	Presenting choices. Modelling. Prompting. Imitation. Reinforcement. Taking turns.
	Ingersoll et al. (2006a)			Reciprocal Imitation Training (RIT) - PRT to elicit target behaviours plus contingent imitation
	Morrison et al. (2002)			Photographic activity schedules on children's on-task and play correspondence behaviour during playtime
	Jurgens et al. (2009)		Augmentative and Alternative Communication System	PECS - Visual support based on picture exchange and its discrimination for learning sentences
	Murdock et al. (2013)		Video modelling	Imitate play from play stories trough videos from an iPad. Prompts to participate
	Lee et al. (2017)			Video self-modelling. Record the behavior. Watch the video, and do the behavior without prompting or reinforcement.

* All these studies use the same technique

Appendix E. Funnel plots for examining publication bias

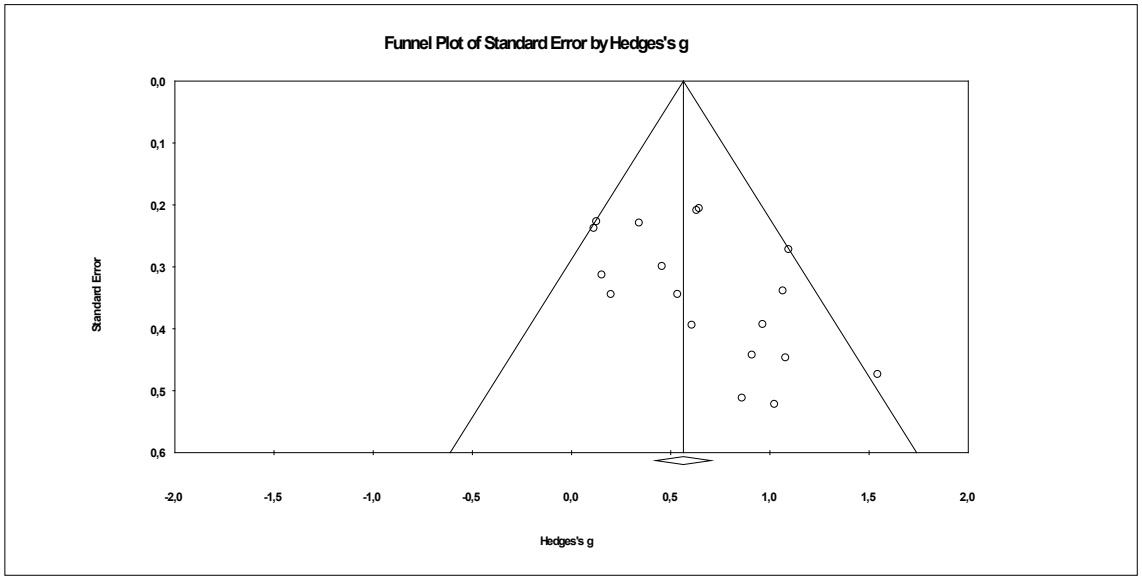


Fig 1. Funnel plot for examining publication bias for overall analysis. Here, Y-axis stands for the standard error of the Hedges' g and X-axis stands for the Hedges' g. Each dot stands for an individual study.

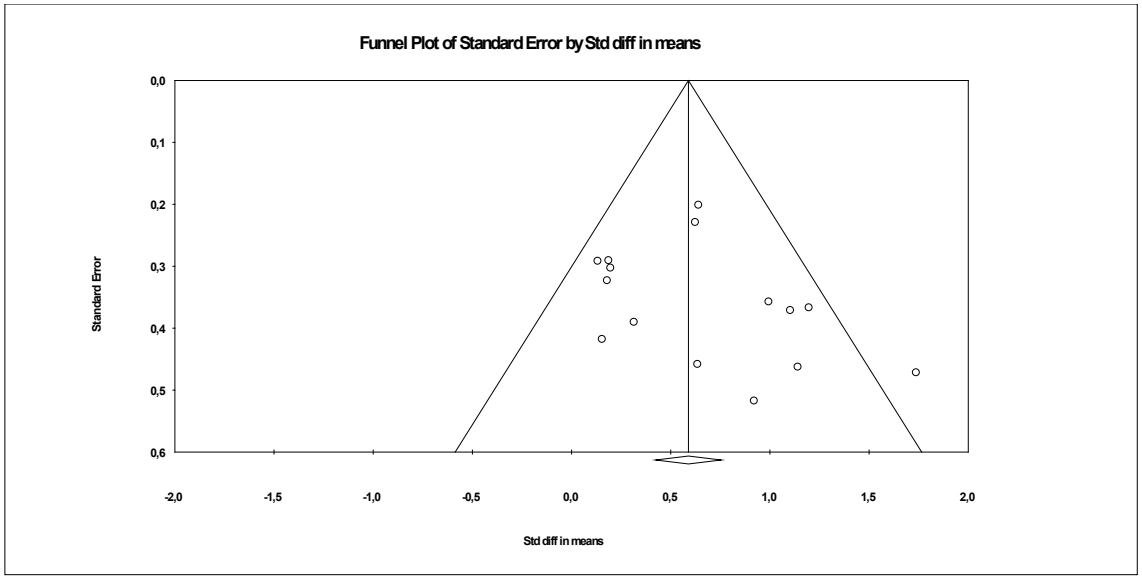


Fig 2. Funnel plot for examining publication bias for Treatment technique analysis. Here, Y-axis stands for the standard error of the Hedges' g and X-axis stands for the Hedges' g. Each dot stands for an individual study.

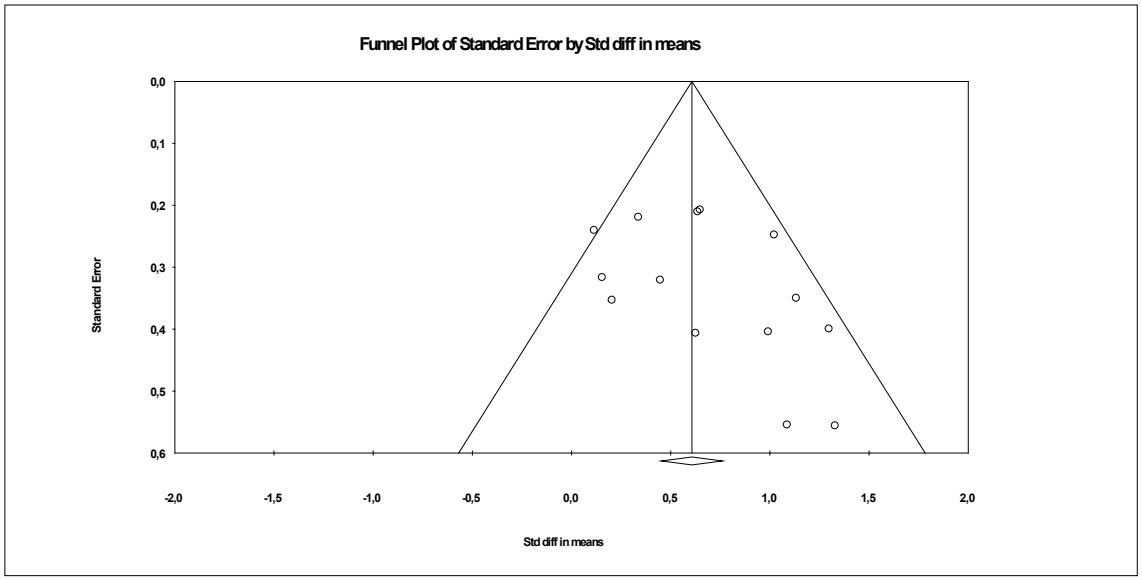
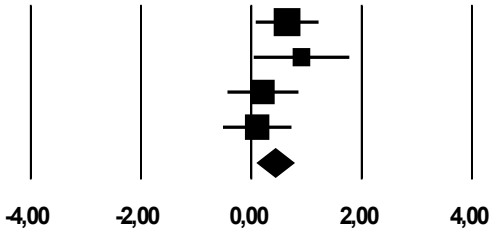


Fig 3. Funnel plot for examining publication bias for joint attention analysis. Here, Y-axis stands for the standard error of the Hedges' g and X-axis stands for the Hedges' g. Each dot stands for an individual study.

Appendix F. Effect sizes for imitation, joint attention and play with group design

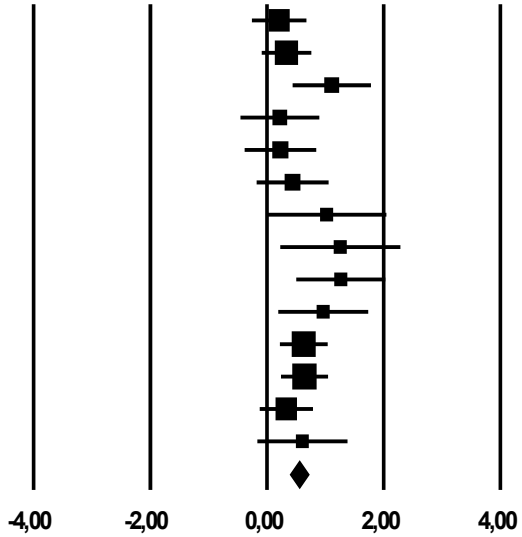
a

Study name	Hedges's g	Standard error	Variance	Lower limit	Upper limit	Z-Value	p-Value
Landa et al. (2012)	0,649	0,292	0,085	0,077	1,220	2,226	0,026
Ingersoll (2010)	0,910	0,442	0,196	0,043	1,777	2,058	0,040
Salt et al. (2002)	0,211	0,330	0,109	-0,435	0,858	0,642	0,521
Van der Paelt et al. (2014)	0,107	0,318	0,101	-0,516	0,730	0,336	0,737
	0,425	0,167	0,028	0,098	0,752	2,550	0,011



b

Study name	Hedges's g	Standard error	Variance	Lower limit	Upper limit	Z-Value	p-Value
Kaale et al. (2012)	0,209	0,238	0,057	-0,258	0,676	0,878	0,380
Chang et al. (2016)	0,335	0,217	0,047	-0,091	0,760	1,540	0,124
Kasari et al. (2006)	1,109	0,343	0,117	0,438	1,781	3,238	0,001
Kasari et al. (2010)	0,221	0,345	0,119	-0,454	0,897	0,642	0,521
Kasari et al. 2014	0,231	0,313	0,098	-0,384	0,845	0,736	0,462
Landa et al. (2012)	0,439	0,315	0,099	-0,178	1,056	1,396	0,163
Lawton et al. (2012)	1,024	0,522	0,272	0,001	2,047	1,961	0,050
Lerna et al. (2012)	1,257	0,526	0,276	0,226	2,287	2,391	0,017
Salt et al. (2002)	1,265	0,390	0,152	0,502	2,029	3,249	0,001
Schertz et al. (2012)	0,964	0,393	0,154	0,194	1,735	2,454	0,014
Schertz et al. (2018)	0,631	0,209	0,044	0,222	1,041	3,023	0,003
Shire et al. 2016)	0,644	0,206	0,042	0,241	1,047	3,129	0,002
Van der Paelt et al. (2014)	0,332	0,233	0,054	-0,125	0,788	1,424	0,154
Wong (2013) comp	0,607	0,394	0,155	-0,165	1,380	1,541	0,123
	0,545	0,077	0,006	0,394	0,696	7,061	0,000



c

Study name	Hedges's g	Standard error	Variance	Lower limit	Upper limit	Z-Value	p-Value
Chang et al. (2016)	0,403	0,249	0,062	-0,084	0,890	1,620	0,105
Goods et al. (2013)	0,860	0,512	0,262	-0,143	1,863	1,681	0,093
Kasari et al. (2006) comp	0,231	0,278	0,077	-0,314	0,777	0,832	0,406
Kasari et al. (2015)	0,147	0,263	0,069	-0,370	0,663	0,557	0,578
Lerna et al. (2012)	2,212	0,421	0,177	1,387	3,038	5,252	0,000
Stickles et al. (2013)	1,430	0,440	0,194	0,567	2,293	3,247	0,001
Van der Paelt et al. (2014)	0,006	0,255	0,065	-0,494	0,505	0,023	0,981
	0,474	0,117	0,014	0,245	0,702	4,063	0,000

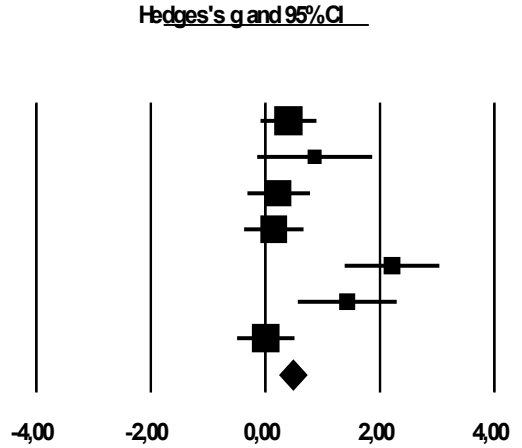
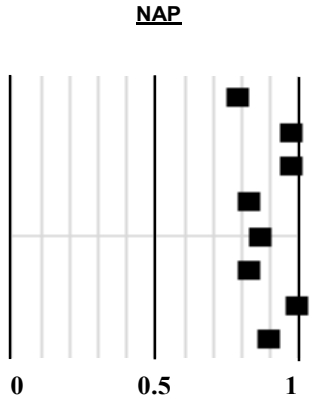


Fig 1. 1a. Effect sizes for imitation from group design studies. 1b. Effect sizes for joint attention from group design studies. 1c. Effect sizes for play from group design studies. All effect sizes are Hedges' g.
The size of the icons represented by the Hedge's g values represent the weight of the study within the meta-analysis

Appendix G. Effect sizes for imitation, joint attention and play with single case design

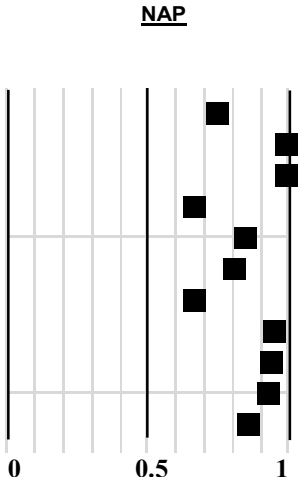
a

Study name	NAP	Lower limit	Upper limit	Z-Value	p Value
Matsuzaki et al. (2012)	0,792	-0,192	1,000	12,374	0,216
Cardon et al. (2011)	0,971	0,682	1,000	66,676	0,000
Cardon (2012)	0,980	0,738	1,000	55,319	0,000
Ingersoll et al. (2006a)	0,832	0,712	0,933	127,331	0,000
Ingersoll et al. (2006b)	0,869	0,712	0,751	94,800	0,000
Ingersoll et al. (2006c)	0,833	0,613	1,000	62,220	0,000
Hwang et al. (2000)	0,997	0,832	1,000	99,387	0,000
	0,896	0,585	0,955	74,015	0,031



b

Study name	NAP	Lower limit	Upper limit	Z-Value	p Value
Rocha et al. (2007)	0,755	0,511	0,999	50,862	0,000
Matsuzaki et al. (2012)	1,000	0,395	1,000	21,213	0,034
Hwang et al. (2000)	0,994	0,826	1,000	97,150	0,000
Whalen et al. (2003)	0,673	0,359	0,987	35,251	0,000
MacDuff et al. (2007)	0,854	0,632	1,000	55,430	0,000
Ingersoll et al (2006a)	0,809	0,693	0,926	113,960	0,000
Benson et al. (2014)	0,667	0,263	1,000	23,212	0,020
Milne et al. (2017)	0,950	0,855	1,000	143,255	0,000
Gunby et al. (2018)	0,949	0,725	1,000	69,644	0,000
Weisberg et al. (2018)	0,930	0,685	1,000	59,567	0,000
	0,858	0,594	0,991	66,954	0,005



c

Study name	NAP	Lower limit	Upper limit	Z-Value	p Value
Ingersoll et al. (2006b)	0,7704	0,61	0,9305	81,08	0.000
Nelson et al. (2007)	0,8114	0,59	1	60,22	0.000
Gillett et al. (2007)	0,9121	0,605	1	48,89	0.000
Lang et al. (2014)	0,7824	0,527	1	49,99	0.000
Jurgens et al. (2009)	0,5781	0,235	0,921	27,75	0,005
Murdock et al. (2013)	0,7231	0,5	0,946	53,29	0.000
Ingersoll et al. (2006a)	0,7479	0,629	0,867	103,59	0.000
Morrison et al. (2002)	0,9364	0,775	1	95,25	0.000
Lee et al. (2017)	1	0,744	1	64,24	0.000
	0,807	0,579	0,963	64,923	0,001

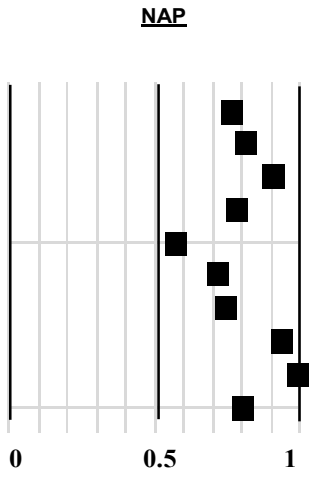


Fig 2. 2a. Effect sizes for imitation from single case design studies. 2b. Effect sizes for joint attention from single case design studies. 2c. Effect sizes for play from single case design studies. All effect sizes are NAP.

Appendix H. Metaregressions for group design studies

Figure 1. Regression of Hedges' g on Age (months)

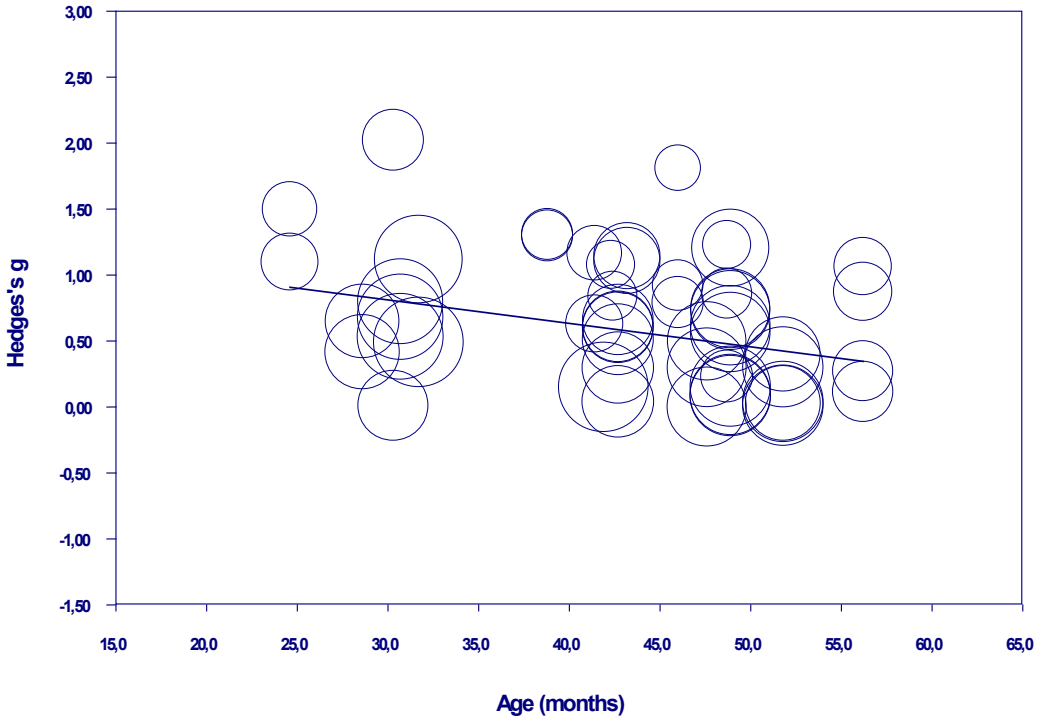


Figure 2. Regression of Hedges' g on IQ

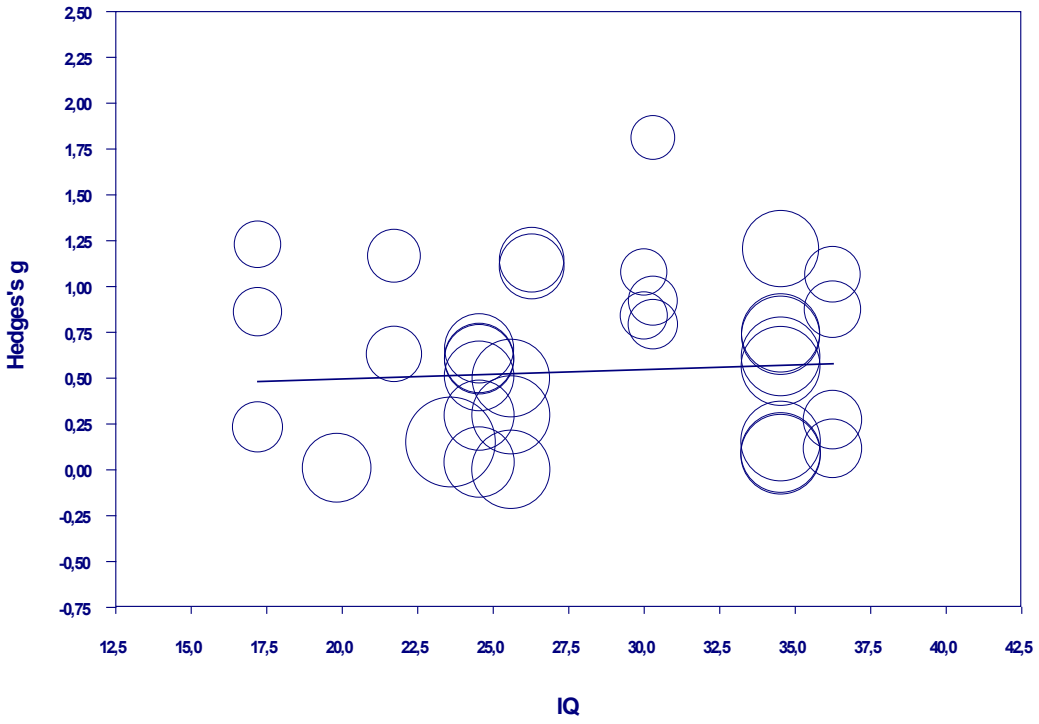


Figure 3. Regression of Hedges' g on Receptive Language

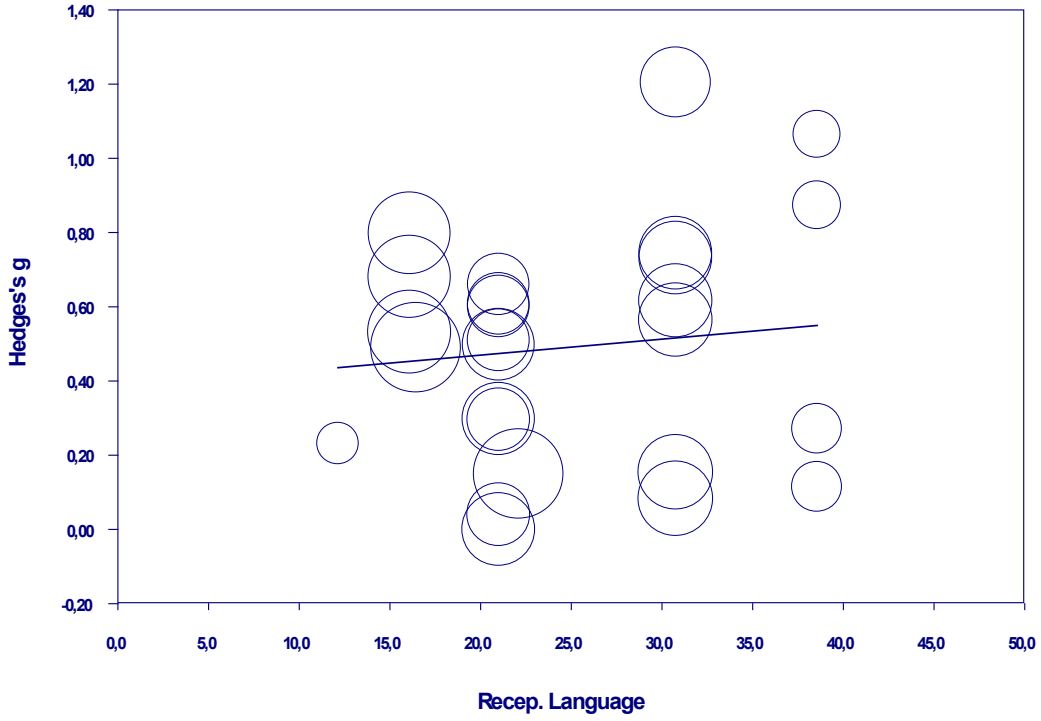


Figure 4. Regression of Hedges' g on Expressive Language

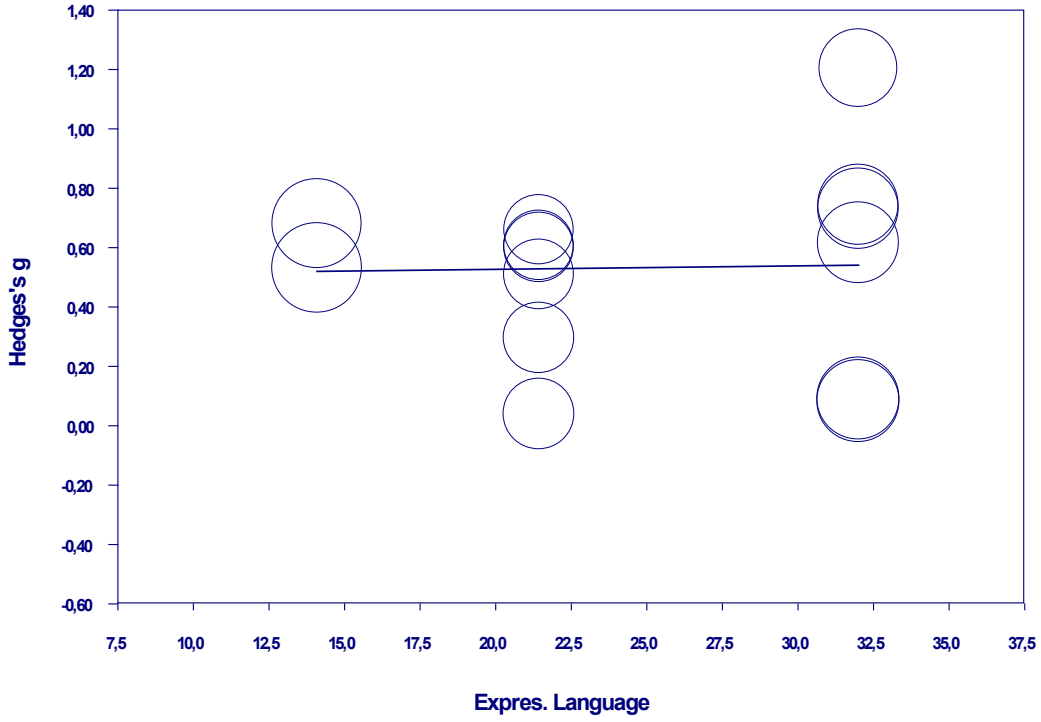


Figure 5. Regression of Hedges' g on Treatment Dosage (hours)

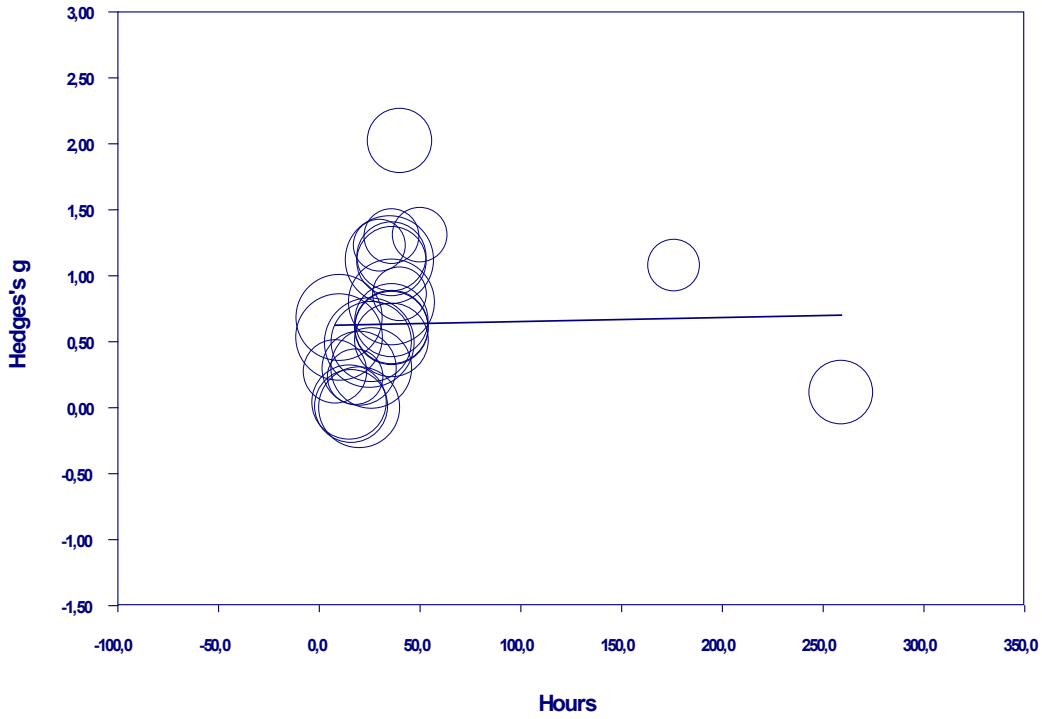
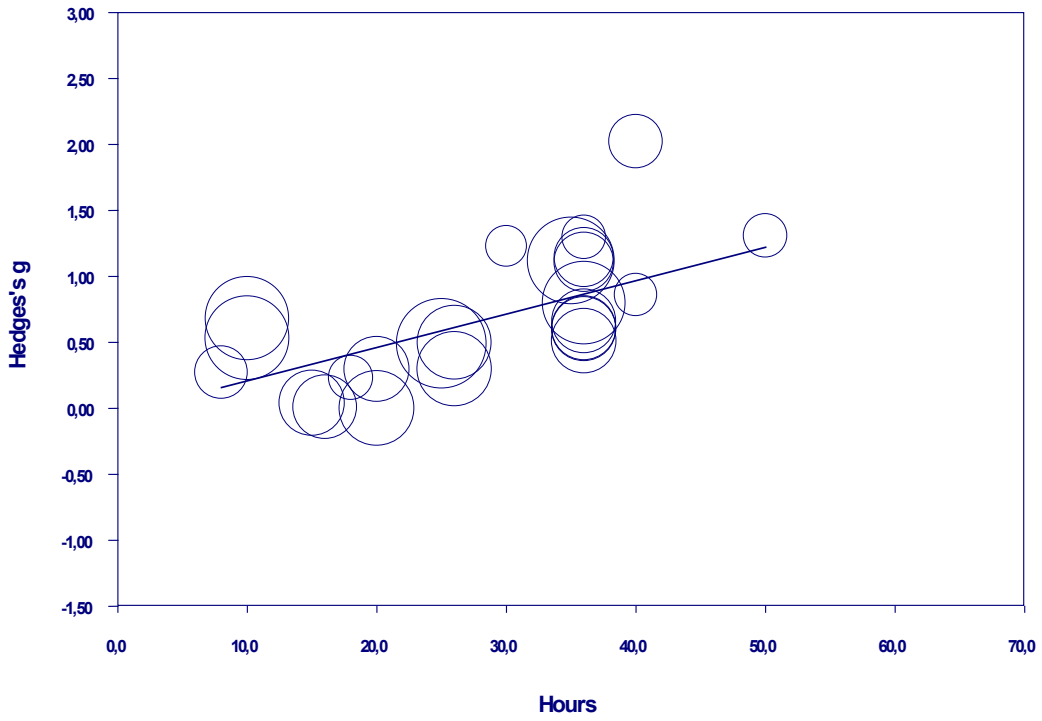


Figure 6. Regression of Hedges' g on Treatment Dosage (hours) without extreme values



Appendix H. Regressions for single case studies

Figure 1. Regression of Non Overlap of All Pairs (NAP) on Age (months)

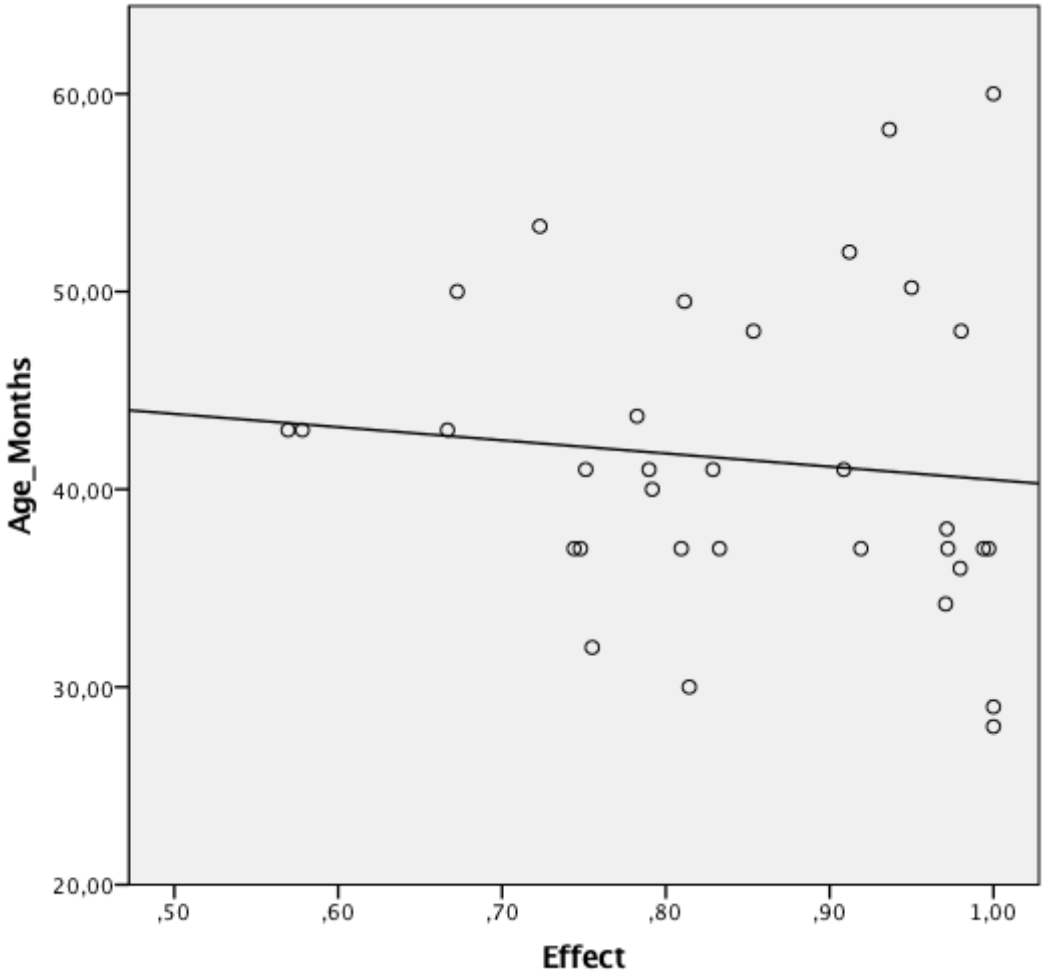


Figure 2. Regression of Non Overlap of All Pairs (NAP) on IQ

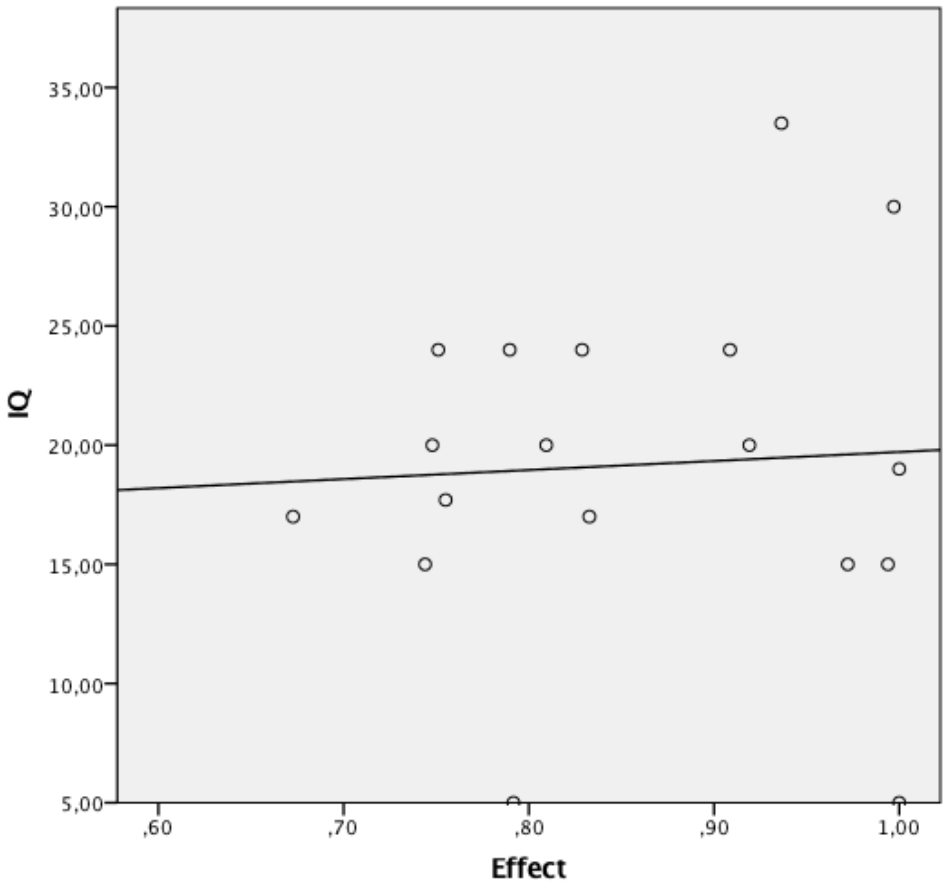


Figure 3. Regression of Non Overlap of All Pairs (NAP) on Expressive Language

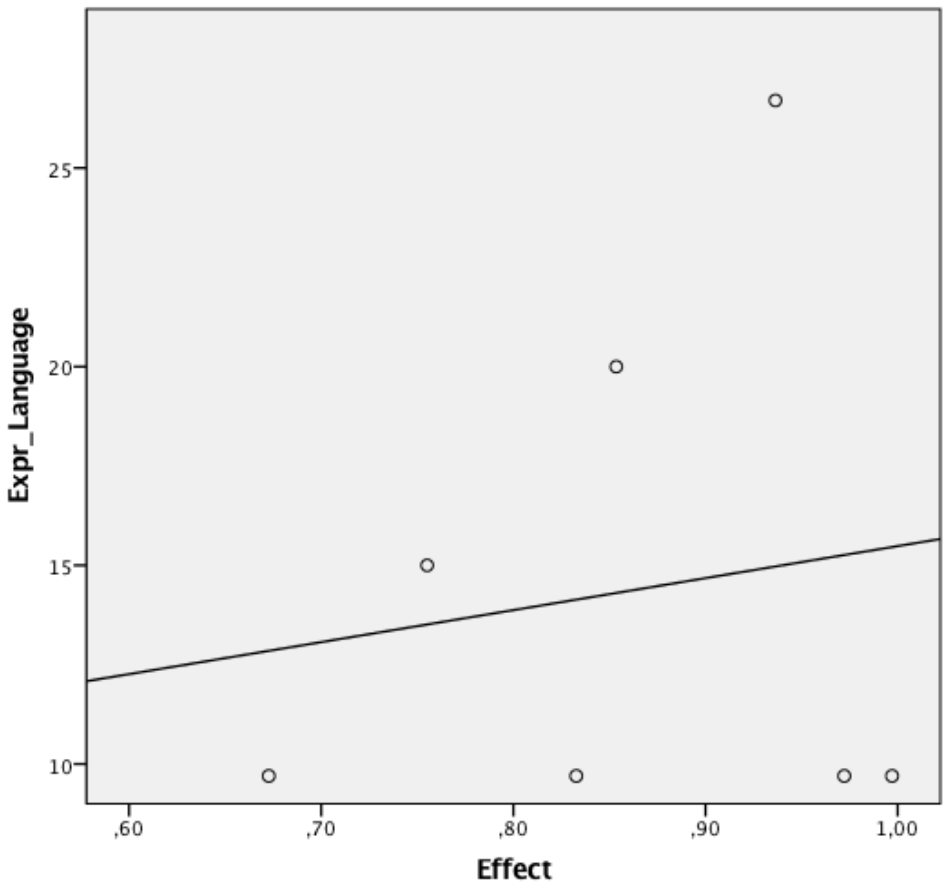


Figure 4. Regression of Non Overlap of All Pairs (NAP) on Receptive Language

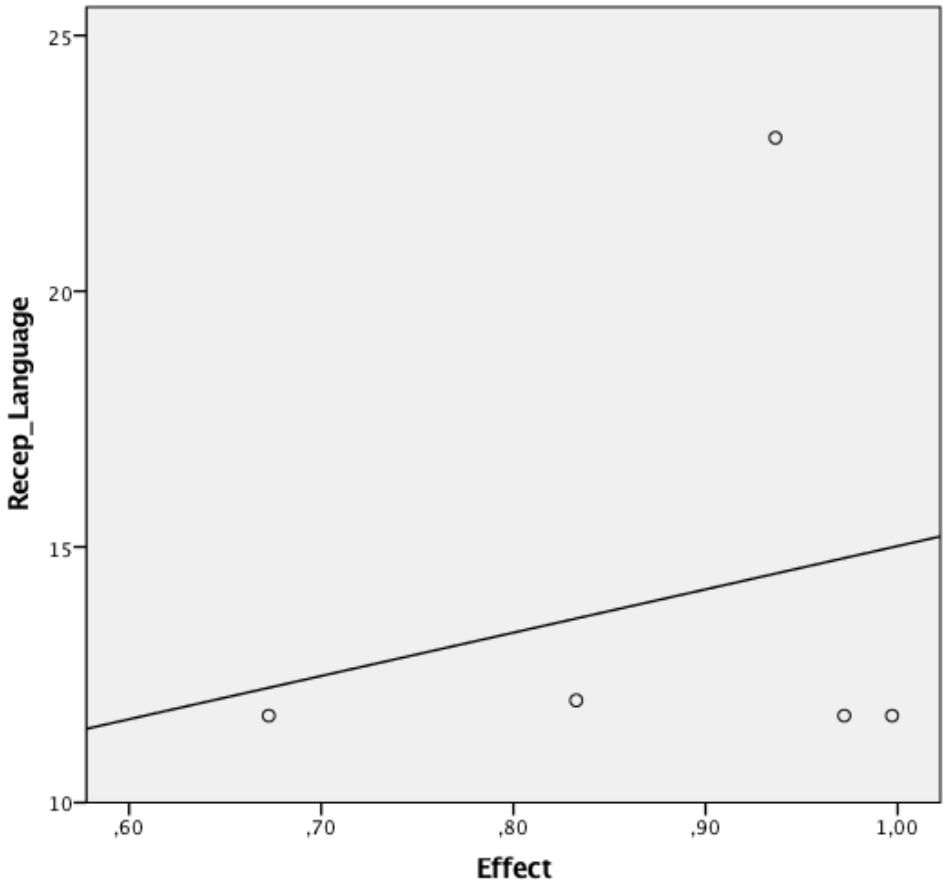
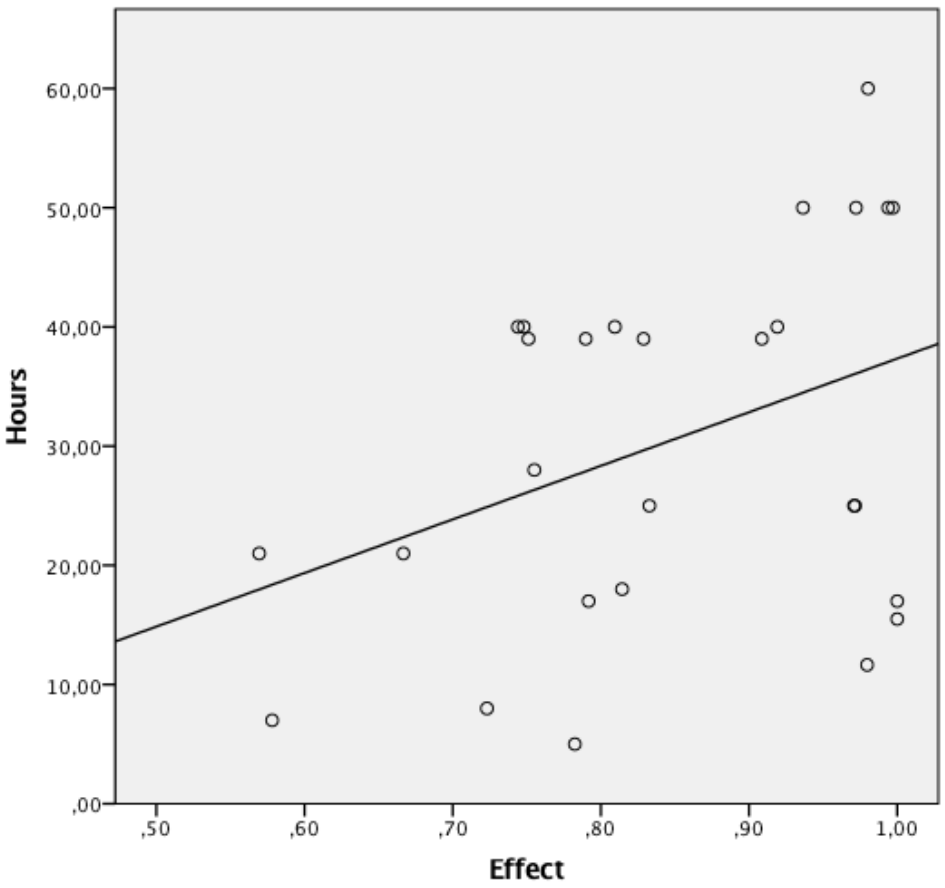


Figure 5. Regression of Non Overlap of All Pairs (NAP) on Treatment Dosage (hours)

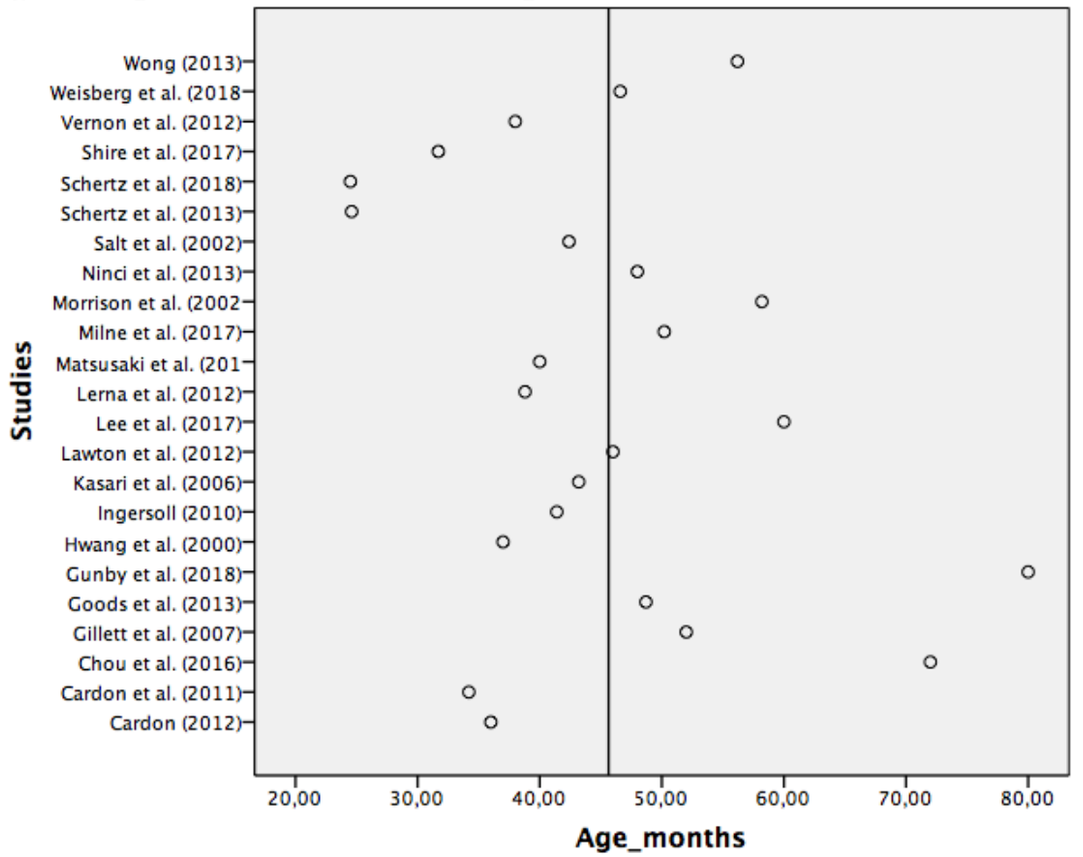


Appendix J. Descriptive analyses of the moderators in studies with larger effect size.

Table 1. Descriptive statistics of the moderators in studies with larger effect size

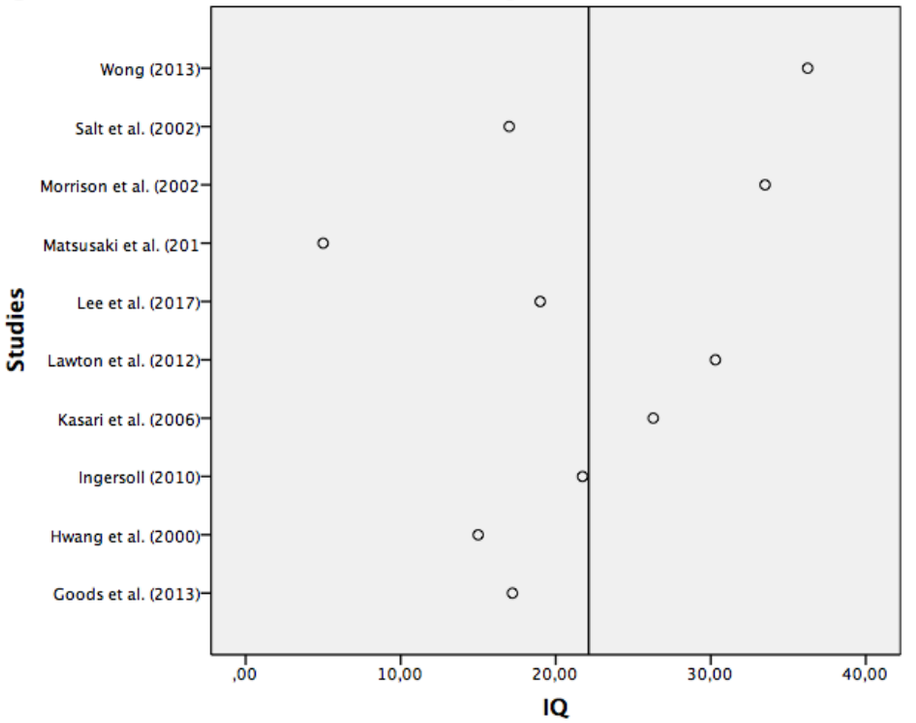
	N	Min.	Max.	Mean	Stand. Deviation	Coefficient of Variation of the mean	Variance	Asimetry
Age_months	23	24,50	80,00	45,6391	13,45348	29,48%	180,996	,814
IQ	10	5,00	36,25	22,1280	9,53042	43,07%	90,829	-,123
Expre_Language	10	9,70	29,73	19,6980	7,06250	35,85%	49,879	-,039
Recep_Language	8	11,70	38,55	19,8375	8,61427	43,42%	74,206	1,627
Dosage	13	8,00	176,00	33,2308	44,03437	132,51%	1939,026	3,286

Figure 1. Age in months in studies with larger effects



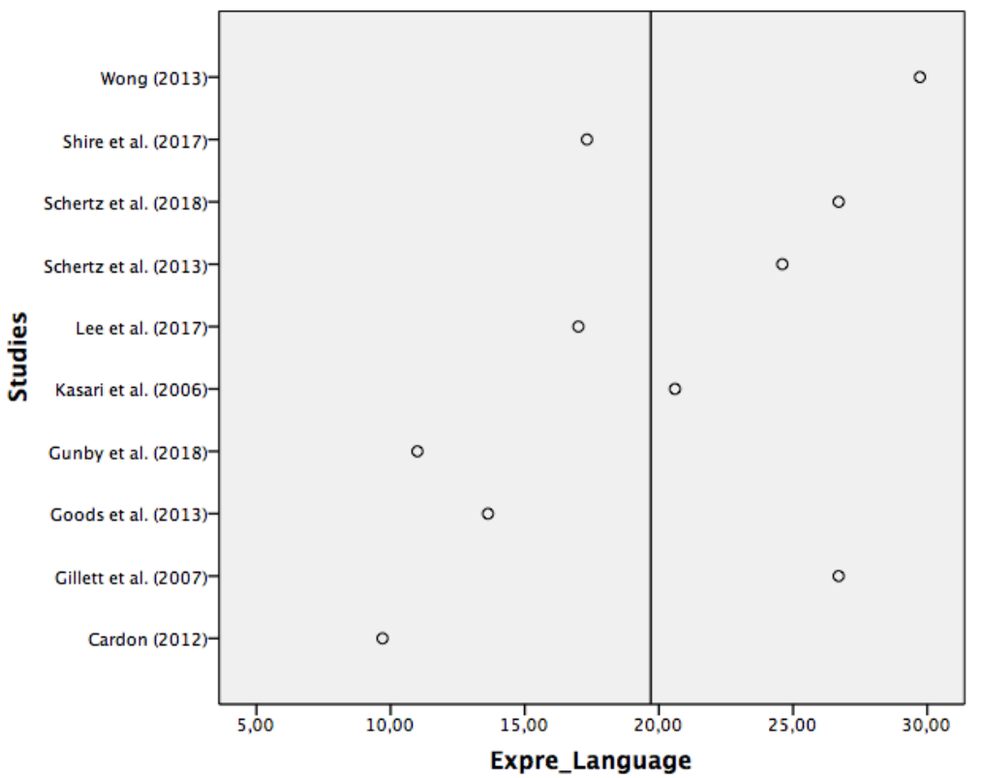
* Line represents the mean (Table 1)

Figure 2. IQ in months in studies with larger effects



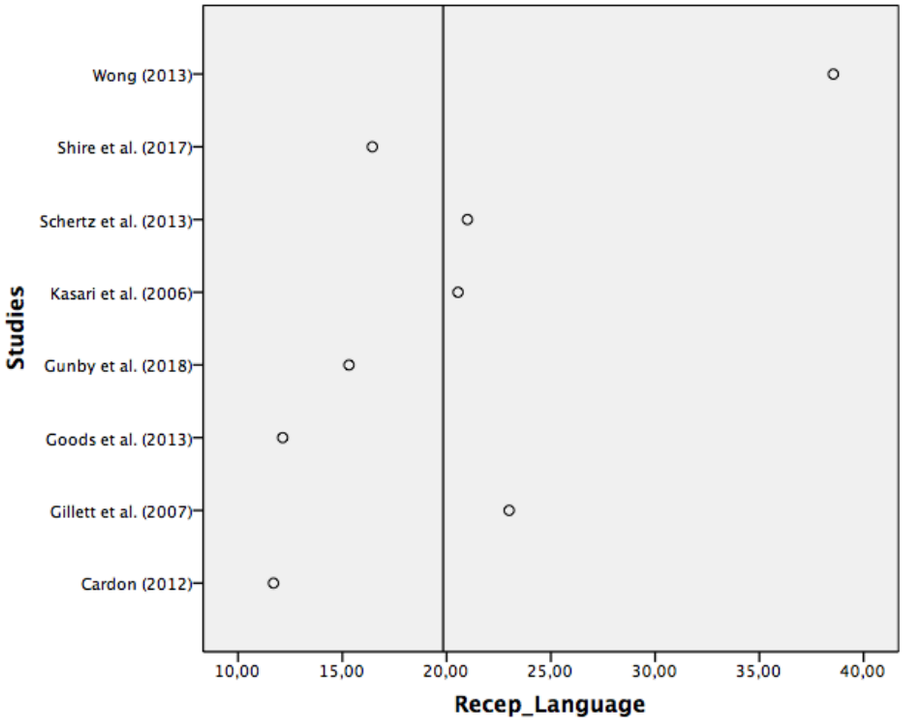
* Line represents the mean (Table 1)

Figure 3. Expressive Language in months in studies with larger effects



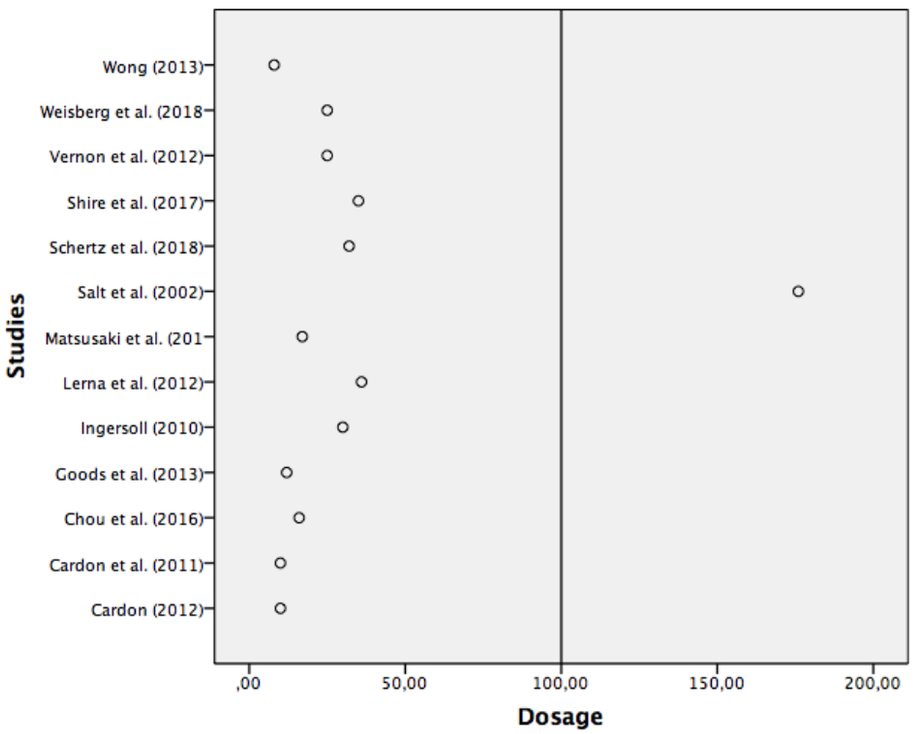
* Line represents the mean (Table 1)

Figure 4. Receptive Language in months in studies with larger effects



* Line represents the mean (Table 1)

Figure 5. Treatment Dosage (hours) in studies with larger effects



* Line represents the mean (Table 1)

Appendix K. Data extraction protocol variables for group and single case design.

GROUP DESIGN

1. **PARTICIPANTS PROFILE**
    1. Number of participants (Experimental and control group)
    2. Age (Experimental and control group)
    3. Gender (Experimental and control group)
    4. Race (Experimental and control group)
    5. Sample source (Experimental and control group)
    6. Previous intervention (Experimental and control group)
    7. Diagnosis:
        i. Principal diagnosis (Experimental and control group)
        ii. Comorbidity (Experimental and control group)
    8. Level of symptomatology:
        i. Test administered
        ii. Score (Experimental and control group)
        iii. Level of severity (Experimental and control group)
    9. Intellectual functioning:
        i. Test administered
        ii. Score (Experimental and control group)
        iii. Type of score (months, raw score, percentage…)
        iv. Level of severity (Experimental and control group)
    10. Language functioning
        i. Test administered
        ii. Score (Experimental and control group)
        iii. Type of score (months, raw score, percentage…)
        iv. Level of severity (Experimental and control group)
    11. Social-Adaptative functioning
        i. Test administered
        ii. Score (Experimental and control group)
        iii. Type of score (months, raw score, percentage…)
        iv. Level of severity (Experimental and control group)
    12. Focused skill (imitation/eye contact/joint attention/gestures/pointing/play): We will extract all types of focused skills, for example, in joint attention we could find response to joint attention and bids to joint attention.
        i. Test administered
        ii. Type of skill (for example spontaneous imitation)
        iii. Score (Experimental and control group)
        iv. Type of score (months, raw score, percentage…)

2. **CHARACTERISTICS OF THE INTERVENTION**
    1. Duration and intensity
        i. Months

ii. Weeks
iii. Days per week
iv. Hours per day
v. Total (hours)

2. Parents participation
    i. Type of time (hours in a month/week/day)
    ii. Time (in hours)
3. Technique used (Behavioural treatment (e.g. Applied Behaviour Analysis (ABA); Pivotal Response Training (PRT); Lovaas; Discrete Trial Training (DTT) ...) ; Developmental treatment (e.g. Relationship Development Intervention (RDI); Early Start Denver Model (ESDM) ...) ; Relationship-based treatment (e.g. Developmental Individual Difference Relationship based (DIR Model) Floortime; Thérapie d'Echange et de Développement (TED) ...) …
    i. Principal
    ii. Secondary
    iii. Third
    iv. Specify
4. Professional profile (Psychologist, teacher ...)
    i. Principal
    ii. Secondary
5. Materials
    i. Specify
6. Cost of the intervention
    i. Specify
    ii. Time (hours)
    iii. Cost

3. CHARACTERISTICS OF THE STUDY

1. Limitations
2. Experimental design (RCT, Quasi-experimental…)
3. Internal validity

4. OUTCOMES

1. Post-intervention
    i. Test administered
    ii. Type of skill
    iii. Score (Experimental and control group)
    iv. Type of score
2. Follow-up
    i. Test administered
    ii. Type of skill
    iii. Score (Experimental and control group)
    iv. Type of score
3. Effect of the intervention (p)
    i. Pre-post intervention (Experimental and control group)

ii. Post-follow up (Experimental and control group)
iii. Pre-post intervention (Experimental vs control group)
iv. Post-follow up (Experimental vs control group)

4. Collateral effects (post intervention)
    i. Language
        1. Test
        2. Score (Experimental and control group)
        3. Effect (p)
    ii. Symptomatology
        1. Test
        2. Score (Experimental and control group)
        3. Effect (p)
    iii. Intellectual functioning
        1. Test
        2. Score (Experimental and control group)
        3. Effect (p)
    iv. Social-Adaptative functioning
        1. Test
        2. Score (Experimental and control group)
        3. Effect (p)

SINGLE CASE DESIGN

In these studies, we will extract the data of each individual participant. In each variable, the data of each participant will be collected.

1. **PARTICIPANTS PROFILE**
    1. Number of participants
    2. Age (months)
    3. Gender
    4. Race
    5. Sample source
    6. Previous intervention
    7. Diagnosis:
        i. Principal diagnosis
        ii. Comorbidity
    8. Level of symptomatology:
        i. Test administered
        ii. Score
        iii. Level of severity
    9. Intellectual functioning:
        i. Test administered
        ii. Score
        iii. Type of score
        iv. Level of severity
    10. Language functioning
        i. Test administered
        ii. Score
        iii. Type of score (months, raw score, percentage…)
        iv. Level of severity
    11. Social-Adaptative functioning
        i. Test administered
        ii. Score
        iii. Type of score (months, raw score, percentage…)
        iv. Level of severity
    12. Focused skill (imitation/eye contact/joint attention/gestures/pointing/play): We will extract all types of focused skills, for example, in joint attention we could find response to joint attention and bids to joint attention.
        i. Test administered
        ii. Type of skill (for example spontaneous imitation)
        iii. Score (Experimental and control group)
        iv. Type of score (months, raw score, percentage…)

2. **CHARACTERISTICS OF THE INTERVENTION**
    1. Duration and intensity
        i. Months

ii. Weeks
iii. Days per week
iv. Hours per day
v. Total (hours)

2. Parents participation
    i. Type of time (hours in a month/week/day)
    ii. Time (in hours)
3. Technique used (Behavioural treatment (e.g. Applied Behaviour Analysis (ABA); Pivotal Response Training (PRT); Lovaas; Discrete Trial Training (DTT) ...) ; Developmental treatment (e.g. Relationship Development Intervention (RDI); Early Start Denver Model (ESDM) ...) ; Relationship-based treatment (e.g. Developmental Individual Difference Relationship based (DIR Model) Floortime; Thérapie d'Echange et de Développement (TED) ...) …
    i. Principal
    ii. Secondary
    iii. Third
    iv. Specify
4. Professional profile (Psychologist, teacher ...)
    i. Principal
    ii. Secondary
5. Materials
    i. Specify
6. Cost of the intervention
    i. Specify
    ii. Time (hours)
    iii. Cost

3. CHARACTERISTICS OF THE STUDY

1. Limitations
2. Experimental design (RCT, Quasi-experimental…)
3. Internal validity

4. OUTCOMES

1. Post-intervention
    i. Test administered
    ii. Type of skill
    iii. Score (Experimental and control group)
    iv. Type of score
2. Follow-up
    i. Test administered
    ii. Type of skill
    iii. Score (Experimental and control group)
    iv. Type of score
3. Effect of the intervention (p)
    i. Pre-post intervention (Experimental and control group)

ii. Post-follow up (Experimental and control group)
iii. Pre-post intervention (Experimental vs control group)
iv. Post-follow up (Experimental vs control group)

4. Collateral effects (post intervention)
    i. Language
        1. Test
        2. Score (Experimental and control group)
        3. Effect (p)
    ii. Symptomatology
        1. Test
        2. Score (Experimental and control group)
        3. Effect (p)
    iii. Intellectual functioning
        1. Test
        2. Score (Experimental and control group)
        3. Effect (p)
    iv. Social-Adaptative functioning
        1. Test
        2. Score (Experimental and control group)
        3. Effect (p)

ARTÍCULO II: PERSPECTIVAS DE FAMILIARES Y PROFESIONALES CON LOS SERVICIOS

Bejarano-Martín, Á., Canal-Bedia, R., Magán-Maganto, M., Fernández-Álvarez, C., Cilleros-Martín, M. V., Sánchez-Gómez, M. C., García-Primo, P., Rose-Sweeney, M., Boilson, A., Linertová, R., Roeyers, H., Van der Paelt, S., Schendel, D., Warberg, C., Cramer, S., Narzisi, A., Muratori, F., Scattoni, M. L., Moilanen, I., … Posada de la Paz, M. (2020b). Early Detection, Diagnosis and Intervention Services for Young Children with Autism Spectrum Disorder in the European Union (ASDEU): Family and Professional Perspectives. *Journal of Autism and Developmental Disorders*, *50*(9), 3380-3394. https://doi.org/10.1007/s10803-019-04253-0

Resumen

Introducción. Los servicios tempranos para el TEA deben recoger las opiniones de los padres y de los profesionales. Estas opiniones rara vez se comparan en un mismo estudio de investigación. ***Objetivos.*** Este estudio pretende conocer las opiniones de las familias y los profesionales sobre la detección temprana, el diagnóstico y los servicios de intervención para niños pequeños con TEA. ***Método.*** Una encuesta en línea recopiló y analizó los datos de 2032 encuestados de 14 países europeos (el 60,9% eran padres y el 39,1% profesionales). ***Resultados.*** Utilizando una escala ordinal de 1 a 7, las opiniones de los padres de los padres fueron más negativas (media = 4,6; SD 2,2) que las de los

profesionales (media = 4,9; SD 1,5) sobre la satisfacción con los servicios. ***Discusión.*** Los resultados sugieren que los servicios deberían tener en cuenta la edad del menor, los retrasos en el acceso a los servicios y la participación activa de las partes interesadas a la hora de mejorar los servicios.

Palabras clave. Trastorno del espectro autista, Detección precoz, Diagnóstico, Satisfacción del paciente, Servicios de salud mental, Encuesta

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ORIGINAL PAPER

Early Detection, Diagnosis and Intervention Services for Young Children with Autism Spectrum Disorder in the European Union (ASDEU): Family and Professional Perspectives

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Abstract

Early services for ASD need to canvas the opinions of both parents and professionals. These opinions are seldom compared in the same research study. This study aims to ascertain the views of families and professionals on early detection, diagnosis and intervention services for young children with ASD. An online survey compiled and analysed data from 2032 respondents across 14 European countries (60.9% were parents; 39.1% professionals). Using an ordinal scale from 1 to 7, parents' opinions were more negative (mean = 4.6; SD 2.2) compared to those of professionals (mean = 4.9; SD 1.5) when reporting satisfaction with services. The results suggest services should take into account child's age, delays in accessing services, and active stakeholders' participation when looking to improve services.

Keywords Autism spectrum disorder · Early detection · Diagnosis · Patient satisfaction · Mental health services · Survey

Introduction

Autism spectrum disorder (ASD) is a neurodevelopmental disorder of early onset, characterized by deficits in social communication, along with restricted and repetitive patterns of behavior, interests or activities which have significant consequences in daily life (American Psychological Association, APA 2013; World Health Organization 2018). When parents first begin to worry about their child's developmental difficulties, they must make a considerable effort to seek answers to their questions and obtain an accurate diagnosis. Families face the challenge of adapting to the new and unexpected reality of having a child with autism in the family, reorganising family roles, finding appropriate treatment/s, and in many cases paying for specialist input (DePape and Lindsay 2015; Hock et al. 2012; Keenan et al. 2010). Several studies indicate that parents of children with ASD report higher stress levels and lower service satisfaction than do parents of children with other disabilities (Baker-Ericzén et al. 2005; Gray 2006; Griffith et al. 2010; Hayes and Watson 2013). Families with a young child with ASD report greater difficulties in accessing services, higher associated costs, and a lack of information and support during the diagnostic process (Hodgetts et al. 2015; Kogan et al. 2008; Thomas, Parish et al. 2012a; Wang et al. 2013).

These families' challenges have been associated with factors linked, not only to the child's characteristics, but also to family characteristics, sociodemographic factors and the characteristics of service delivery. With regard to sociodemographic aspects, observation has shown that individuals

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with ASD belonging to families with a high parental socio-economic status (SES) and high parental educational level are diagnosed earlier, and that their families report greater satisfaction with the diagnostic process (Durkin et al. 2010; Goin-Kochel et al. 2006; Irvin et al. 2012; Moh and Magiati 2012; Thomas, Zahorodny et al. 2012b).

In relation to the services provided to young children with ASD, the sources of distress and dissatisfaction mentioned by parents are professionals' tardiness in addressing their initial concerns, delay in getting a diagnosis, and the lack of professional support (Altiere and von Kluge 2009; Bishop et al. 2007; Bluth et al. 2013; Crane et al. 2016; Divan et al. 2012; Moh and Magiati 2012; Osborne et al. 2008). It has also been suggested that some families' low level of satisfaction with the care they receive is related to communication difficulties between families and professionals (Liptak et al. 2006), inadequate organisation of care programmes (Chiri and Warfield 2012), and the absence or scarcity of skilled professionals specialised in ASD (Krauss et al. 2003). A very recent study (Crane et al. 2018) on the views of families, professionals and adults with autism about the diagnostic process found that delays to diagnosis of ASD and the lack of rapport between parents and professionals affected satisfaction with services. In addition, families wanted more guidance, counselling and emotional support to help them to understand the meaning and the implications of the diagnosis received, in order to be able to avoid crisis in the family and manage stress adequately (Crane et al. 2018).

Despite the difficulties expressed by families and professionals alike, it is generally accepted that, over the years, progress has been made in improving care for children with ASD (Austin et al. 2016), even though further improvements are still clearly required. In recent years, efforts to improve detection, diagnosis and early intervention services for children with ASD have paid more attention to the views of families and professionals, reflecting the belief that improvement strategies should focus on the child and his or her family (McConachie et al. 2015; Pellicano et al. 2014). The purpose is to ensure that families are more actively involved in assessment of the child's and the family's needs, and that professionals take a proactive approach to identify such needs. Families that report being actively involved in decisions and have good communication with professionals also report greater satisfaction with services, fewer gaps in services, fewer delays in accessing treatment and services, lower stress, and lower general ASD-related costs (Kuo et al. 2011; Moh and Magiati 2012; Burke and Goldman 2015). Likewise, recent studies have shown that, when professionals respond promptly to parents' concerns, delays in access to diagnostic services are reduced and overall satisfaction is increased (Zablotsky et al. 2017; Zuckerman et al. 2015).

An increasing number of researchers are taking advantage of social networks and institutional websites to distribute surveys aimed at exploring the state of art about unmet needs and services (Gotham et al. 2015; Zhao et al. 2019). These surveys are based on large proportion of responses and they constitute a cost-effectiveness procedure for the analysis of the situation and hypothesis generation, even though they fail providing inferences free of bias. However, they are excellent tools when analysing a large and diverse population.

Although the opinions and satisfaction of families and professionals with early detection, diagnosis, and early intervention services seem to have played a fundamental role in changing policies and improving services for the ASD community, the perspectives of these two different groups have rarely been considered together. Hence, it is important to obtain detailed information about the type of services which young children with ASD receive and the views held by various European stakeholders on such services, in order to inform the decisions of policy makers -at both a national and European level- affecting the financing of services and training of families and professionals.

To this end, we used the Autism Spectrum Disorder in the European Union (ASDEU 2015–2018) network to conduct a multinational study aimed at assessing and collecting the opinions and attitudes of the autism community (families and professionals) concerning early detection, diagnosis and intervention services for children with ASD under 9 years of age in 14 European countries. More specifically, our objectives with regard to early detection, diagnosis and intervention services were: (a) to identify the types of services received by children with ASD in Europe; (b) to examine families' and professionals' degree of satisfaction with services across Europe; (c) to explore variations in age at detection, diagnosis and intervention and delays in accessing services, as reported by parents and professionals; and lastly (d) to identify the variables that predict service satisfaction in both groups.

Methods

Survey Development

The development of the surveys was carried out in three steps. The first was a focus group activity aimed at obtaining initial direct information about the perceptions and ideas of people normally involved in the processes of detection, diagnosis and early intervention of children with ASD. This information helped us to delimit the content and topics of interest that we were going to include in the surveys. The second step focused on the development of the items and the structure of the questionnaires. The last step consisted of a controlled distribution of the survey (pilot study) to a group of families and professionals with the purpose of identifying

difficulties in understanding the items and evaluating the functioning of the survey.

Focus Groups

In this first step, twenty focus-group sessions were conducted across the ASDEU network. Taking into consideration the purpose of the study, we distributed the focus groups in relation to two thematic areas: (a) early detection and diagnosis; and (b) early intervention. Each of the 10 participating European countries conducted two focus group sessions addressing each of these two topics. The countries involved were Bulgaria; Denmark; Finland; France; Iceland; Italy; Poland; Portugal; Romania; and Spain. The size of the groups ranged from 5 to 11 participants, with a total of 225 participants in all (146 (64%) professionals and 79 (36%) familiars). Each focus group was led by a facilitator and one other researcher who was present as an assistant. The topics discussed were the age of access to services, delays in receiving necessary services and/or treatments and their causes, satisfaction with the care and treatments received, knowledge about autism that participants attribute to professionals, the limitations of the services (economic, in material resources, trained personnel), the participation of the family in the diagnosis process and during the treatment activities, the best practices known to the participants, the level of training in diagnosis and/or treatment that professionals have and that provided to families to meet the needs of children, coordination between services and general procedures that participants know for early detection, diagnosis and early intervention in each country.

Survey Content and Structure

Participants of focus groups were not directly involved in the creation of the surveys. The authors of this article analysed the transcripts obtained from the focus group discussions, extracting and grouping into categories and ordering the ideas, perceptions, concerns, and interests expressed by the participants. This set of categories served to elaborate the items of the questionnaires and to organize the surveys differentiating questions directed to families and questions directed to professionals. Following this procedure, two different surveys were drawn up to facilitate collection of data from the two respondent groups, namely, parents or families and professionals who were directly related to a child with ASD, (Appendixes 1 and 2 for Final Survey English Version).

Section one collected basic information about respondents' gender, age, country and city of residence. In addition, family members were asked about their relationship to the child, academic attainment, number of people living permanently in the household, and the gender, age, diagnosis and verbal ability of the child. Diagnostic categories were defined according to the Diagnostic and Statistical Manual of Mental Disorders (DSM) and International Classification of Diseases for Mortality and Morbidity Statistics (ICD) classifications (APA 2013; WHO 2018). Professionals, on the other hand, were asked about their main job and their experience in working with children with autism.

Sections two to four contained questions on early detection, diagnosis and early intervention respectively. Both the family member and professional surveys included a brief introductory explanation of the type of questions that the respondent would have to answer. Professionals were asked whether they were directly involved in any programme dealing with early detection, diagnosis or early intervention. Those who responded negatively to these questions were asked to provide contact details of someone involved in such programs, and then directed to the end of the survey. These respondents were not included in the final sample. Of the 35 participants nominated in this way, 28 participated in the survey. The questions were intended to elicit specific data on the processes of detection, diagnosis and beginning treatment, from the moment when families or professionals first began to worry until the time when the specialist treatment began. Respondents were thus asked about the age of the child when concerns first arose (detection), age at diagnosis and age when treatment or intervention started, as well as any delays in access to services, types of professional involved in the different processes, type of diagnosis, degree of family involvement at each stage, type of intervention, and the like. Therefore, the survey included specific questions about satisfaction with detection, diagnosis and intervention (See Appendixes 1, 2).

The principal response categories were: (i) age of the child at the time of accessing to detection, diagnosis and intervention services (families: list from 0 to 9 years; professionals: based on different ranges). The answers were stratified as 0–18, 18–24, 24–36, and > 36 months at detection; 0–18, 18–32, 32–46, 46–60, and > 60 months at diagnosis and intervention. (ii) Delays in access to services ranging between 0 and 3 months, 3–6 months, > 6 months. (iii) Assessment the satisfaction with services on a scale of 1 to 7 (1: extremely inadequate; 2: moderately inadequate; 3: slightly inadequate; 4: neither adequate nor inadequate; 5: slightly adequate; 6: moderately adequate; 7: extremely adequate). The different answer choices for all aforementioned questions were then stratified and recoded into three new categories, namely, negative (from 1 to 3), neutral (4) and positive (from 5 to 7). (iv) The level of participation in the intervention sessions. Parent participation were stratified into two categories, namely, active participation (very actively and actively participation responses) and occasional/no participation (occasional participation and I don't participate responses).

Survey Testing

After translation and adaption by researchers from the respective project countries, 14 country-specific versions of the survey were produced. This process included the use of official translations of some questions (e.g., intervention programs, manuals, etc.) where available in each country. The translations were uploaded to the Qualtrics web platform (https://www.qualtrics.com), altrics web platform (https://www.qualtrics.com).

Before the surveys were publicly launched, they were piloted in three countries (Spain, Denmark and Iceland) with the support from twelve parents from six family' organisations, five professionals from the ASDEU project network and three professionals not directly related to the project. Parents 12 (60%) and professionals 8 (40%) were asked to give their opinion on the content, format and accessibility of the surveys. All pilot respondents reported that the survey was accessible and that the questions were clear and comprehensible, indicating no need to further adapting wording or length of questions. Participants completed the survey in 15–20 min. The Flesch reading Ease was 60.8 and the Flesch-Kincaid Grade Level was 8 (word office tool). These scores were within the standards for a document to be accessible and easy to read for the population.

Recruitment Procedure

The survey was made available online and distributed by researchers affiliated to the ASDEU project website in the 14 participating countries. The main goal was to secure the largest possible sample from the countries that participated in the project, so as to obtain a global analysis that would be useful for the management of new hypotheses. Clinical practitioners as well as parents' and professionals' organisations promoted the survey through their own networks. Professionals were asked to distribute the survey to family members of children under 9 years old receiving a treatment for ASD and give them guidance on how to respond. Invitations to participate in the survey were also sent to websites visited frequently by the ASD community, i.e., service providers, private and public associations, Facebook groups, Twitter, bulletins, etc. In addition, links to the survey were provided in the online newsletter of the ASDEU project (http://asdeu.eu/newsletter/). Special education Schools, rehabilitation centres working with children with neurodevelopmental problems, psychiatry services for child and adolescent, home guidance centres and residential centres for children with ASD participated in the surveys and disseminated them through the families. Finally, a global sample was obtained from a total of 24 countries. The surveys were available online, so that any professional or family member of a child with autism could answer them, regardless of the country from which they came.

Ethical Approval

Ethical approval was given by the Ethics Committee of the University of Salamanca, Spain (201700008785). Respondents accessed the survey from this server. The same survey was conducted in all countries. There was a global survey in several languages, which participants accessed and consented to answer: prior to starting, all respondents were required to read the information about the survey and give their informed consent electronically.

Data Analysis

As the survey was administered electronically, the data were downloaded for further analysis, which was conducted in four distinct phases.

Comprehensive descriptive analyses of the two respondent groups (families and professionals) were performed.

Multinomial regression analyses were conducted to compare parents' and professionals' reports with respect to the following four different dependent variables: (i) age of access to services, to examine the likelihood of the child being detected, diagnosed and beginning the intervention earlier; (ii) delay in access to services, to ascertain who, parents or professionals, reported the longest waiting times; (iii) satisfaction with services, to examine the likelihood of positive versus negative satisfaction with detection, diagnosis and intervention; and (iv) lastly, whether parent participation was associated with intervention satisfaction.

Finally, to investigate the different items that predict (independent variables) the positive satisfaction of the early detection, diagnosis, and intervention services (dependent variables), multinomial logistic regressions models were made with the age of access to services and delays in accessing these services.

Results

Sample Characteristics

Although a total of 3693 people initiated the survey, only 2032 respondents met the inclusion criteria (Tables 1 and 2). The reasons for exclusion of the remaining 1661 respondents were: (1) failure to complete an adequate percentage (70%) of survey sections two and three; (2) not having a child with ASD in the family; (3) not working for institutions with ASD among their services, respectively; (4) not a European resident (European area). Countries with fewer than

Table 1 Sample size by respondents' countries

	Family members $n=1237$	Professionals $n=795$	Total $N=2032$ (%)	
Austria	23	12	35	1.7
Belgium	159	40	199	9.7
Denmark	94	96	190	9.4
Finland	52	200	252	12.4
France	105	140	245	12.0
Great Britain	19	1	20	1.0
Iceland	50	45	95	4.7
Ireland	79	15	94	4.6
Italy	86	30	117	5.7
Poland	222	79	301	14.8
Portugal	25	10	35	1.8
Romania	28	–	28	1.4
Spain	278	116	393	19.4
The Netherlands	6	4	10	0.5
Other	11	7	18	0.8

Countries in the "Other" category: Norway, Switzerland, Germany, Malta, Cyprus, Slovenia, Hungary, Croatia, Russia, Macedonia

five respondents were included in the sample but country-specific statistics were not reported.

The family group was the larger of the two respondent groups, with 1237 respondents (60.9% of all respondents). The majority of respondents in the family group were parents (81.3% mothers), with the most frequent educational level being a first degree or higher (64%). The average age of children with ASD in such families at the date of completing the survey was 76.7 (*SD* 31.0) months, and most of these children were male (82.7%). A total of 795 professionals answered the survey (39.1% of the whole sample), most of whom (90.3%) were women. The largest group were those working in mental health services (psychologists, psychiatrists or mental health therapists), followed by those working in other health services (general practitioners, paediatricians or nurses) and teachers working in the educational system. About two-thirds of professionals (64.2%) reported that they had more than 5 years' experience in that job.

Table 2 Sample characteristics and information about services by respondent type

	Family members, n = 1237	Professionals, n = 795
Age of respondents in years, mean (SD)	50.8 (7.1)	45.8 (11.5)
Gender (% male)	14.6%	9.7
Relationship to child (father: mother: other[a])	14.2%: 81.3%: 4.5%	NA
Educational level—first degree or higher	64.0%	NA
Child's age at time of survey, mean (SD)	76.7 (31.0)	NA
Child's gender (% male)	82.7%	NA
Profession—health: mental health: education	NA	34%: 39%: 17%
Professional experience—1–3 years: 3–5 years: > 5 years	NA	20%: 15%: 64%
Person who first raised concerns—caregiver: professional	70%: 30%	NA
Source of concern about services—professional's concern: survey	96%: 3%	NA
Professional involved in detection—health: mental health: education	21%: 28%: 11%	22%: 25%: 13%
Professional involved in diagnosis—health: mental health	48%: 77%	63%: 90%
Diagnostic classification used: DSM: ICD	–	23%: 26%
Information received in diagnosis—medical: educational: social: none	31%: 45%: 29%:20%	69%: 84%: 81%: 4%
Intervention information—results: programme type: cost: participation: none	NA	71%: 66%: 19%: 13%
Time of sessions, mean (SD)	0.81 (0.22)	1.90 (1.22)
Session format—group: individual	31%: 89%	40%: 75%
Parental participation—active: occasional: none	40%: 29%: 30%	73%: 22%: 4%
Distance to early intervention service (km), mean (SD)	12.6 (14.8)	NA
Travel time to early intervention service (minutes), mean (SD)	21.8 (15.1)	NA
Intervention programme: specific to ASD (e.g., Applied Behaviour Analysis): health: parental training	24%: 49%: 47%	NA
Age of access to detection services in months, mean (SD)	18.3 (13.4)	NA
Age of access to diagnostic services in months, mean (SD)	36.4 (17.7)	NA
Age of access to intervention services in months, mean (SD)	42.2 (15.4)	NA

[a]Grandparents, siblings

All percentages exclude missing values

Early Detection, Dianosis and Intervention Services

The majority of family respondents (70%) indicated that the first person to suggest that something was wrong with the child's development was a family member (Table 2). In general, family respondents said that they relied on the professionals' experience of typical development to recognise warning signs; only 3.1% said that they had noticed problems after responding to a specific ASD screening survey [e.g., M-CHAT-R (Robins et al. 2014) or Q-CHAT (Allison et al. 2008)]. Both respondent groups reported that the professionals most frequently involved in the detection and diagnostic processes were those working in mental health services. Professional reported that they informed families about the child's specific needs, highlighting the educational needs. Caregivers also reported this, however, noteworthy 20% of the families reported that they did not receive any information (e.g., medical or educational needs) at the time of the child's diagnosis. The majority of professionals reported using the DSM (23%) or ICD (26%) diagnostic classification at their centers.

Most family respondents indicated that they were not involved (40%) or only occasionally involved (30%) in the intervention process, whereas 70% of professional respondents reported that parents participated actively in interventions. Only 13.1% of professionals reported that they had not provided parents with information about intervention programs when their child started treatment. Family respondents indicated that the most commonly recommended interventions were speech therapy, physiotherapy and parental training sessions. Specific intervention programs to ASD people were available for 24% of the families respondents. The number of sessions that children with ASD receive according to their families was lower than the number of sessions reported by professionals. Both groups reported that the majority of sessions that children with ASD received were individual sessions. The distance and time to reach the intervention services varied greatly, ranged from 1 to 100 kilometres and 1 to 60 min.

Age at Detection, Diagnosis and Intervention, and Delay in Access to Services

The main objective of this comparison was to ascertain which type of participant reported lower access ages and whether these differences were statistically significant ($p < 0.05$). Additionally, the analysis was conducted to examine the likelihood of the child having fewer delays in access to detection, diagnostic and intervention services. According to family respondents, the average age at which concerns were first raised about the child who was later diagnosed with ASD, was 18.3 (*SD* 13.4) months. The average age at diagnosis was 36.4 (*SD* 17.7) months, with most diagnoses occurring between 32 and 46 months according to both families and professionals. Professionals reported that the age of most detected cases ranged from 24 to 36 months. Average age of starting an early-intervention programme reported by families was 42.2 (*SD* 15.4) months (Table 2).

Detection of symptoms appearing before the age of 18 months were more likely reported by family respondents compared to professionals. Also, families were more likely than professionals to report a delay detection over 6 months (Table 3). Again, most families reported a delay in the access to diagnostic services over 6 months, compared

Table 3 Age at detection, diagnosis and intervention and delays in access to services: group comparisons

	% Family members ($n = 1223$)	% Professionals ($n = 786$)	Family members vs. professionals OR (95% CI)
Child's age at detection			
0–18 months*	41.7	13.1	
18–24 months	7.4	30.8	**0.11 (0.07–0.17)**
24–36 months	31	42.1	**0.31 (0.21–0.45)**
> 36 months	19.9	14	0.66 (0.41–1.09)
Delay in access to detection services			
0–3 months*	21.6	23.3	
3–6 months	29.4	41.1	0.89 (0.62–1.28)
> 6 months	50	35.6	**2.90 (2.11–3.98)**
Child's age at diagnosis			
0–18 months*	3.1	2.1	
18–32 months	26.8	30.5	**0.44 (0.27–0.71)**
32–46 months	31.9	42.4	**0.39 (0.25–0.62)**
46–60 months	17.6	14	0.66 (0.38–1.14)
> 60 months	20.6	11	**1.48 (0.46–3.54)**
Delay in access to diagnostic services			
0–3 months*	9.8	29.4	
3–6 months	22.2	41.2	**0.23 (0.17–0.33)**
>6 months	68	29.4	**6.93 (4.75–10.12)**
Child's age at start of intervention			
0–18 months*	5.1	7.7	
18–32 months	42.0	52.0	0.98 (0.96–1.02)
32–46 months	34.5	31.7	1.00 (0.98–1.02)
46–60 months	14.2	6.1	1.03 (1.01–1.05)
> 60 months	4.2	2.5	1.01 (0.99–1.03)
Delay in access to intervention			
0–3 months*	62.6**	56.5	
3–6 months	22.9	18.7	1.11 (0.73–0.81)
>6 months	14.8	24.7	**0.54 (0.36–0.81)**

The table reports odds ratios (ORs) and the corresponding 95% confidence intervals (CIs), χ^2 and Nagelkerke's R^2. Predictors significant at $p < 0.05$ are indicated in bold

*Reference category

**14.8% of the 0- to 3-month group received an intervention before diagnosis

to 3–6 months reported by professionals. However, it was professionals more often than families who reported a longer delay -of over 6 months- in access to intervention services.

Satisfaction with Services in Relation to Respondent Characteristics

Interaction terms in the models were used to explore differences between families and professionals, taking into account respondents' sex where appropriate. To this end, separate analyses were performed for professionals and families (familial male vs. familial female; professional male vs. professional female). The sample size for these analyses was 2032. Figure 1 illustrates the mean rankings (from 1: extremely inadequate, to 7: extremely adequate) provided by each male and female respondent in each respondent group. Rankings indicate significant differences between respondents by group (families/professionals) and sex. Families were more likely to express less positive satisfaction (scale from 4 to 7, see Fig. 1) than professionals for all items evaluated (Table 4). Regarding detection, we found greater differences between families and professionals in the evaluation of the general process, as well as in the degree to which the professionals took into account the family's concerns. Regarding diagnostics, the greatest differences were found in the general evaluation of the process, in addition to the professional level of the team involved. Differences between families and professionals about the information and support received, as well as the number of sessions, in the evaluation of the intervention were also found, which previously noticed in the description of the services, reported by families and professionals (See Table 2).

No sex-related differences were observed except in the case of females in the professional group, who had a more positive opinion than did their male counterparts about specific factors in the diagnosis and intervention programmes. Female professionals were more likely to express more positive satisfaction than male professionals for the items "The staff's qualifications" (Detection) and "The number of sessions" (Intervention) (Fig. 1; Table 4).

Comparisons across participants regarding to their participation in the intervention sessions and the assessment of the service were conducted. The main goal of this analysis was to ascertain whether active participation by parents resulted in more positive satisfaction with services. The different answer choices for these questions were collapsed to examine the likelihood of positive versus negative satisfaction with parent participation (active participation vs. occasional/no participation). Families who were actively involved were more likely to express more positive satisfaction with the intervention process than were those families who did not participate or only participated occasionally (Table 4). This

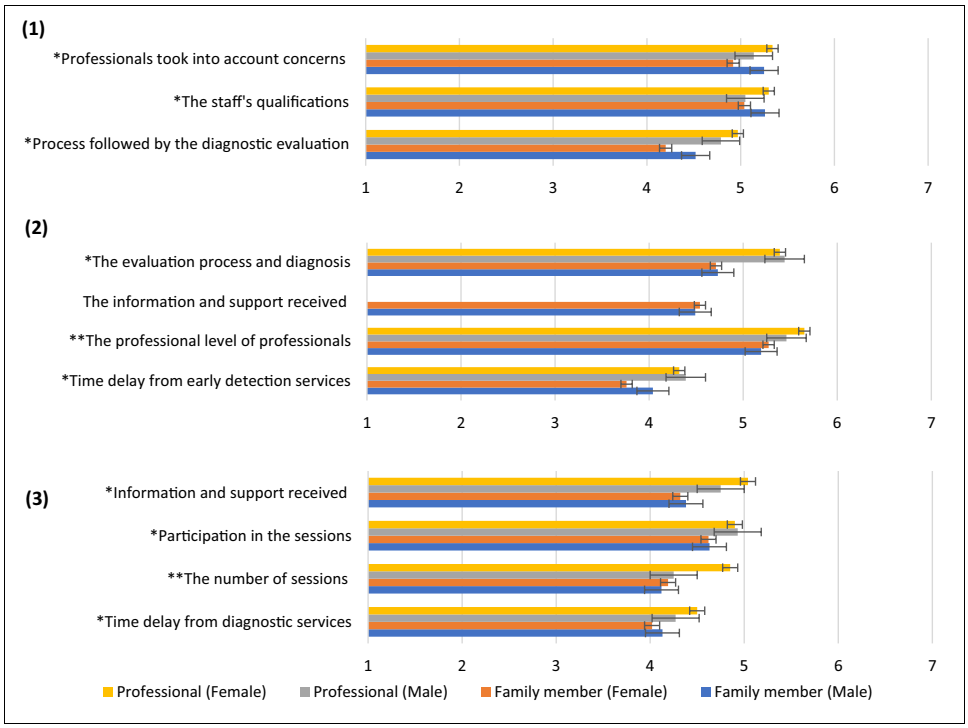

Fig. 1 Average opinion of services by family and professional respondents by sex. (1) Early detection process, (2) early diagnostic process, (3) early intervention process. Scale: from 1 (extremely inadequate) to 7 (extremely adequate), collapsed and transformed into the following categories: negative (from 1 to 3), neutral (4) and positive (from 5 to 7).*Difference between family and professional respondents; $p < 0.05$. **Sex difference within a respondent group; $p < 0.05$

(1) early detection process, (2) early diagnostic process, (3) early intervention process. Scale: from 1 (extremely inadequate) to 7 (extremely adequate), collapsed and transformed into the following categories: negative (from 1 to 3), neutral (4) and positive (from 5 to 7)
* Difference between family and professional respondents; $p<0.05$
** Sex difference within a respondent group; $p<0.05$

Table 4 Comparison of satisfaction among respondents according to sex and parents' participation in the intervention

	Family members vs. Professionals[b]	Male family member vs. Female family member[b]	Male professional vs. Female professional[b]	Active participation vs. occasional/no participation[b]
	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)
Detection				
Process followed by diagnostic evaluation[a]	**0.40 (0.32–0.49)**	1.41 (0.99–2.00)	0.81 (0.45–1.45)	–
Staff qualifications[a]	**0.62 (0.49–0.79)**	1.17 (0.79–1.74)	0.66 (0.36–1.20)	–
Professionals took into account family's concerns[a]	**0.45 (0.35–0.57)**	1.44 (0.96–2.16)	0.69 (0.36–1.32)	–
Diagnosis				
Delay between detection and diagnostic services[a]	**0.60 (0.50–0.74)**	1.23 (0.89–1.71)	1.18 (0.69–2.01)	–
The professional level of professionals[a]	**0.45 (0.34–0.60)**	0.98 (0.65–1.46)	**0.43 (0.22–0.82)**	–
Information and support received[a]	–	0.89 (0.64–1.26)	–	–
The evaluation process and diagnosis[a]	**0.37 (0.28–0.47)**	0.85 (0.60–1.20)	0.72 (0.37–1.41)	–
Intervention				
Information and support received[a]	**0.41 (0.31–0.53)**	1.01 (0.69–1.49)	0.58 (0.18–1.37)	**1.85 (1.28–2.66)**
Participation in sessions[a]	**0.69 (0.53–0.91)**	1.14 (0.73–1.76)	0.81 (0.42–1.56)	**1.60 (1.06–2.43)**
Number of sessions[a]	**0.47 (0.37–0.60)**	0.91 (0.63–1.33)	**0.46 (0.25–0.83)**	**1.52 (1.06–2.17)**
Delay between diagnosis and start of intervention[a]	**0.71 (0.51–0.95)**	0.96 (0.66–1.39)	0.61 (0.20–1.12)	**1.46 (1.03–2.09)**

The table reports odds ratios (ORs) and the corresponding 95% confidence intervals (CIs). Comparisons significant at $p < 0.05$ are indicated in bold

(–) Not applicable. Not asked or not possible to calculate

[a]Satisfaction ratings were classified into 3 groups: 0 = negative (reference category), 1 = neutral, and 2 = positive

[b]Reference group in the multinomial logistic regression

effect applied to all the aspects of intervention that were evaluated, highlighting the information and support received over the rest of the items evaluated.

Relationships Between Age at Detection, Diagnosis and Intervention, Delay in Access to Services and Satisfaction with Services

Multinomial logistic regressions models were fitted with the following independent variables: (1) age of detection (0–18 months), diagnosis (0–24 months), and intervention (0–36 months); and, (2) delays in accessing such services (> 6 months). Separate analyses of the total sample were performed for each group of participants. Families of children who reported to have been detected at an early age and have had less delay in access to this service were more likely to express higher family positive satisfaction with detection services (scale from 4 to 7, see also Fig. 1). Table 5 shows the odds ratios (ORs) for each predictor vis-à-vis each outcome measure of satisfaction (See Appendix 3 supplementary material all items evaluated separately). By reducing the age of detection, the perception of detection services would be more positive in all items for families. In addition, families who reported delays in access an early detection service of more than 6 months score the process worse. On the other hand, professionals who reported early child's age of detection were more likely to express higher positive satisfaction in any of the items related to the assessment of detection services (Table 5).

The results for diagnostic services follow a similar pattern to screening services for families only. Families who reported less delay in access to detection and diagnostic services were more likely to express higher satisfaction. On the contrary, professionals who reported early child's age of diagnosis and less delays in access to this service were not more likely to express higher satisfaction with diagnostic services (Table 5).

Finally, the results indicated that the same families of children who reported shorter delays in access to detection, diagnosis and early-intervention programmes were more likely to express higher family positive satisfaction of intervention services. Therefore, by reducing the delay in access to detection, diagnosis and intervention, the assessment of intervention services would be more positive. On the other hand, professionals who reported early child's age of intervention and less delays in access to this service were not more likely to express higher positive satisfaction in any of the related items. (See Table 5 and Appendix 3).

Table 5 Predictors of positive satisfaction

	Detection OR (95% CI)	Diagnosis OR (95% CI)	Intervention OR (95% CI)
Family members			
Child's age at detection (0–18 months)	**2.05 (1.46–2.90)**	**1.50 (1.04–2.17)**	**1.40 (0.97–2.01)**
Delay in access to detection (> 6 months)	**0.22 (0.15–0.33)**	**0.18 (0.11–0.29)**	**0.42 (0.29–0.61)**
Child's age at diagnosis (0–24 months)	–	**2.41 (1.18–4.93)**	**2.14 (1.18–3.88)**
Delay in access to diagnosis (> 6 months)	–	**0.29 (0.16–0.56)**	**0.56 (0.34–0.92)**
Child's age at intervention (0–36 months)	–	**–**	**2.08 (1.23–3.86)**
Delay in access to intervention (> 6 months)	–	**–**	**0.56 (0.36–0.89)**
Professionals			
Child's age at detection (0–18 months)	0.55 (0.12–2.58)	0.80 (0.09–8.02)	2.23 (0.35–16.7)
Delay in access to detection (> 6 months)	**0.25 (0.07–0.68)**	0.68 (0.14–5.79)	0.44 (0.11–1.12)
Child's age at diagnosis (0–24 months)	–	0.55 (0.07–5.60)	0.71 (0.21–3.29)
Delay in access to diagnosis (> 6 months)	–	0.74 (0.11–5.98)	0.89 (0.35–2.42)
Child's age at intervention (0–36 months)	–	–	2.32 (0.65–5.59)
Delay in access to intervention (> 6 months)	–	–	0.53 (0.19–0.86)

Discussion

The aim of this study was to analyse the characteristics of detection, diagnosis and intervention services received by children with ASD, and to compare and contrast the overall satisfaction reported by 1223 families and 760 professionals, in order to provide an evidence based framework for clinicians, decision-makers and researchers to consider, and so enable them to incorporate the views of these groups into their activities. Rather than seeking to be representative of the entire EU, this study sought instead to obtain a representative sample of most of the countries that participated in the ASDEU project (14), so as to make a global analysis that would be useful for the management of new hypotheses and changes. Since it was a free-access survey, it was not possible to control for the fact that in some countries the response of the participants was lower than expected. Although the unequal size of the number of participants in the respective countries renders it impossible to draw conclusions for a particular country, this is not so for all of them. Similarly, the fact that it was an open-access survey meant that some participants who were in the European region but outside the EU, responded to the survey. These participants were taken into account in the global analyses.

Overall satisfaction of participants was positive (> 4 on the scale) for all early detection, diagnosis and intervention items evaluated (Fig. 1). Survey participants tend to be more engaged in the process that non-respondents, and more likely to have had positive experiences with services, as well as more positive attitudes of the participants (Keusch 2015). Although overall satisfaction was positive, professionals were more satisfied than family members (Table 3). These differences could be due to the fact that families have to deal with the process not only of gaining recognition and acceptance of the fact that there is something wrong with their child's development, but also of waiting for services, as well as the sheer amount of services and medical visits that children with ASD need, all of which results in higher levels of stress (Burke and Goldman 2015; Summers et al. 2005). In addition, differences could be due to the fact that families respond based on their personal experiences, while the professionals respond based on the experiences across all of the parents they attend. Whereas providers might recognize that delays in the diagnosis or the onset of services is not optimal, they do not experience the frustration experienced by families, accumulated each month. As a consequence, these differences should be interpreted carefully in light of the great disparity between responders' respective experience of the process. For instance, significant differences were found in satisfaction with the number of intervention sessions (Table 4). Based on their experience, parents reported receiving less than half the time in intervention sessions that those reported by professionals, which would show how the personal experiences lived in the services could affect to satisfaction. Dissatisfaction with the information provided by practitioners, the support received and the delays in access to services observed in this study is consistent with the findings of previous studies (Dymond et al. 2007; Hodgetts et al. 2015; Liptak et al. 2006; Ngui and Flores 2006; Rogers et al. 2016). However, no previous studies have shown differences between family members and professionals in terms of satisfaction with detection, diagnosis and intervention services. Future studies should therefore focus on the reasons for these differences.

The ages of detection, diagnosis and access to intervention reported by family members are markedly lower than those reported in some previous studies (Baio 2018; Oswald et al. 2017) but similar to those reported in others (Adelman

and Kubiszyn 2017; Daniels et al. 2017; McIntyre and Zemantic 2017; Moh and Magiati 2012). It is possible that the variation in families' reports of age at first access to services for children with ASD simply reflects differences in socio-economic status, since it has been observed that families with greater socio-economic resources enjoy better access to services and specialists. Families with low socio-economic resources tend to report higher ages of access to services (Kalkbrenner et al. 2011; Liptak et al. 2008). Another possible explanation lies in differences in parent awareness of their child's early difficulties (Daniels and Mandell 2014; Sacrey et al. 2015; Zablotsky et al. 2017; Zuckerman et al. 2015). In this study, 70% of families reported having had some concerns about the development of the child who was subsequently diagnosed with ASD, something that may have reduced the age of detection and diagnosis, and thus speeded up access to an intervention programme. Families reported that the average delay between detection and diagnosis (18.1 months) was much longer than between diagnosis and treatment (5.8 months), and 14.8% of families reported that their child had started an intervention programme (private or public) before receiving a formal diagnosis. Another possible explanation could be the fact that in this type of surveys the participants were more aware and had greater resources, both personal and material. Because the recruitment process for the survey was carried out in parent associations, as well as in other ASD specific services, participants may have had access to resources such as diagnosis or intervention, which would significantly reduce delays to these services. Parents who are more engaged are more likely to be concerned earlier and to have experienced relatively longer delays in accessing services such as diagnosis. Future studies should investigate whether satisfaction with services is more closely linked to the length of delays in access or to the age at which the child obtains access to services.

Differences between families and professionals could be related to their differing experiences. In their recent experience, professionals may have conceivably dealt with cases where diagnosis was made quite early and delays in access to services were short, with the result that these recent positive experiences may have influenced their estimation of the promptness with which services respond to parents' concerns. However, the fact that families reported tardier and slower responses than did professionals would suggest that service lags exist and there is a need to provide professional staff with technical and human resources (training programmes and tools) which will speed up the detection and diagnostic processes and reduce delays in access to such services.

Families who had early access to services and experienced fewer delays tended to rate services more positively. These results are consistent with studies such as those by McKenzie et al. (2015) and Kuo et al. (2011), where parents who reported the greatest satisfaction with the information and support received were those whose child had been younger at the time of diagnosis. Most of the families that participated in the study reported that, after becoming concerned about their child and communicating their concerns to a paediatrician, they had to wait, first for a diagnosis of ASD from a specialist service and then for an intervention programme. In contrast, professionals' evaluations were more positive and more uniform than those of families, and they reported that waiting times were shorter and children younger when they gained access to services. This could be explained due to the fact that family members must complete the entire process, from detection to intervention, while professionals may belong to one of these services. Therefore, the experiences of family members, who have to go from one service to another will tend to be more negative.

An important finding related to detection is that very few families reported participation in ASD-specific screening programs (3.1%). These results are in line with previous studies on the use of ASD-specific programs in the USA (Adelman and Kubiszyn 2017) and Europe (García-Primo et al. 2014). However, 70% of families reported having expressed concerns to different professionals, which would imply the start of a development screening program. Therefore, detection was primarily based on the experience and knowledge of the professional. Use of an effective and efficient screening tool would allow professionals to detect potential ASD cases at an earlier age and refer them to diagnostic services earlier, thus reducing the delay between detection and diagnosis, which can be as long as 18 months, according to families. Reducing the delay in diagnosis would enable children to begin intervention programmes earlier. If intervention occurs early, when neuronal plasticity is much greater, long-term positive results can be achieved (Crais and Watson 2014a). It has been widely reported by paediatricians that there are many barriers to detection of ASD and the use of population screening programmes (Crais et al. 2014b), and there have been warnings about the lack of training to enable early detection of a disorder which is diagnosed frequently every year.

This study shows that active parental involvement increases family satisfaction with services, a finding consistent with other studies which show that parental involvement is fundamental to satisfaction with intervention programmes (McIntyre and Zemantic 2017; Stadnick et al. 2013). In recent years, active involvement has also been shown, not only to increase service satisfaction, but also to improve intervention outcomes by, for instance, increasing progress in skill acquisition (Ingersoll and Wainer 2013; Kasari et al. 2015; Pickles et al. 2016). In addition, involving parents reduces the costs of intervention programmes by decreasing the number of hours with professionals and increasing skill development in natural contexts (Ingersoll et al. 2017;

Pickles et al. 2016). All these factors mean that parental involvement in interventions reduces the economic burden on the family, health-care system and society, along with the stress associated with having a child with ASD (Kasari et al. 2015).

Limitations

One of the study limitations is that surveys targeted at EU-sized populations can only estimate the thrust of the analysis and establish hypotheses, but such hypotheses can never be considered definitive. One cannot ascertain the total number of family members with ASD children or professionals who work with this group in Europe. In terms of their external validity (generalization), these results should therefore be interpreted with caution. This study represents the first approach to comparing parents' and professionals' perceptions of the services made available to children with ASD. Accordingly, it falls to future studies to compare these same groups using larger sample sizes.

Another limitation is that our family sample was more highly educated (most respondents had a university degree or higher) and not as diverse as that of other studies (Mandell and Salzer 2007; Thomas, Zahorodny et al. 2012b). Even so, our study sample group was similar to many other studies based on surveys of families of children with ASD and professionals (Casagrande and Ingersoll 2017; Liptak et al. 2008; Weiss et al. 2012). In addition, participation in the study was limited to people with Internet access, a factor that may have excluded some potential low-income respondents without good access to the Internet. These potential sources of selection bias may have rendered the sample unrepresentative of the general community (Salomone et al. 2015). Although online surveys are commonly used and the limitations associated with them are well known, it is possible that our results cannot be generalised to populations with lower socio-economic levels. Sample size differed from country to country, and consequently countries with large samples may not be representative of all the countries that were included within a given category. An additional limitation of our sample was that we did not have parental and professional ratings for the same individual. Future research should therefore compare the views of families and professionals about the *same* children with ASD, in order to have a more accurate picture of the differences found in this study.

Although survey dissemination was the same in all countries, parents' organizations, special education schools, centers specializing in ASD, etc., were not identical and, in addition, have different policies. The surveys were distributed to national associations and centers in each country, which then disseminated them to local associations and centers. It is impossible to ascertain the number of associations or centers that participated in the study, or the number of participants belonging to each association or centre. It follows, therefore, that the number of participants invited by their associations or centers to complete the survey may not be the same in all countries, thereby increasing the variability of the results. Another source of variability was the type of participants that participated in the surveys. Because they were distributed mainly in services for ASD people, it is likely that highly engaged families with knowledge of ASD, as well as professionals with a high degree of experience have been the largest group of surveys participants (Table 2). Satisfaction with processes and services is usually assessed through surveys. These are so-called "self-selection" surveys (Bethlehem 2010) which are not based on probabilistic sampling. The survey is simply uploaded to the website. The respondents are those who have access to the Internet and visit the website. In our case is because they have some interest in relation to autism and decide to participate in the survey. Therefore, the participants are usually parents and professionals who are committed in some way to autism, but also with a higher level of education, and with more economic resources relative to the general population (Bethlehem 2010; Infante-Rivard and Cusson 2018). Assuming this reality, the results of self-selection web surveys can be considered representative when there are a large number of respondents, or as a result of using advanced adjustment weighting procedures in the methods of analysis (Bethlehem 2010). Future studies should compare these results with those that can be obtained by surveying parents (or family members) less involved in services and professionals with less specific ASD experience.

An additional limitation as regards respondents' characteristics is the role of the professional. This respondent characteristic was not taken into account in analyzing responses. For instance, someone in education may have a poorly informed opinion of the importance of using an ASD-detection screening tool. Accordingly, future studies should analyze the point of view of different professional profiles to ascertain whether there are differences in service satisfaction.

Another potential sample limitation is that, since the recruitment system was online and anonymous, we were unable to ascertain why some potential respondents decided not to participate. A total of 1661 people started but did not complete the survey, without it being possible to establish why they failed to complete it once they had begun (e.g., due to connection or computer problems, lack of time, distractions, etc.). It is however reasonable to assume that those who decided to complete the survey were the most committed and competent respondents, and that, by extension, may thus not be representative of the autism community as a whole (Fletcher-Watson et al. 2017).

Moreover, our data were mostly derived from responses to closed questions, which compel the respondent to select

from a fixed, restricted set of answers. Use of this question format was necessary for several reasons, such as the international nature of the survey, and the accompanying lack of translation resources to translate respondents' answers to open-ended questions. Ultimately it was a compromise, whereby the restriction on response options enabled us to collect data from a larger sample of the autism community.

Lastly, another factor affecting the range and access to services for children with ASD is location (rural, urban etc.). The location of the nuclear family has a significant impact on the number of services and professionals available (Murphy and Ruble 2012). Family and professional survey participants reported residing in urban areas. Future research should study these relationships in a more representative sample, so as to be able to provide the best recommendations, taking into account the particular characteristics of each family and the points of view of the professionals concerned.

Conclusions

Our results indicate that, though families and professionals in the autism community are broadly satisfied with services and that children's ages were lower and delays in access to services were shorter than in other studies, differences were nevertheless found between these two groups. In particular, families of children with ASD reported lower overall satisfaction with and higher child ages and longer delays in access to services than did professionals who routinely work with children with ASD. Notwithstanding this, the results suggest that, in both families and professionals, greater satisfaction with services is associated with low ages of detection and diagnosis, as this enables intervention to begin sooner. The clearest message from this study is that it is parents who are still crucial for the detection of the first ASD signs. Families are telling us that there is a need of collaborative, inclusive and self-critical professionals, and that they should be involved in every aspect of care for their child. Service policies and future research should focus on reducing delays in access to services, through, say, the implementation of early ASD-specific detection programmes, in order to increase families' satisfaction with services and thereby possibly reduce their stress and improve their wellbeing.

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Author Contributions ABM, RCB, MMM and MP designed the study and wrote the manuscript; ABM, MP and RCB carried out the statistical analyses and interpreted the results; CFA and PGP collaborated in writing the manuscript. ABM, RCB, MMM, CJF and MP designed the surveys and MRS, AB, RL, HB, SVP, DS, CW, SC, AN, FM, MLS, IM, AY, ES, SLJ, MEB, AV, CR, BR, QG, SB, LP, JXK ODK, RK, EP and TS translated the surveys into their native languages and disseminated them in their respective countries. All authors have read and approved the final manuscript.

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Data Availability A copy of the surveys can be seen at: http://asdeu.eu/wp2-activities/.

Compliance with Ethical Standards

Conflict of interest The authors have no conflict of interest to declare.

Ethical Approval Ethical approval was given by the Research Ethics Committee of the University of Salamanca, Spain (201700008785).

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Family Survey - final

Identification, diagnosis and early intervention for young children with autism spectrum disorder (ASD)

A survey study to improve support for young children with ASD

Before you respond to the survey, we want to explain the reason for this research and what it will involve for you. Please take the time to read this information carefully. For more information, do not hesitate to contact Ricardo Canal (rcanal@usal.es).

The survey is designed to collect information from people who have or have recently had any direct involvement with screening, diagnosis or treatment services for young children under 6 years of age with ASD. Direct involvement means you are a close family member of a child under 6 years of age who has received any of those services and knows firsthand this kind of services for children under 6 years of age that are provided where the child lives.

The objective of the survey is to gather the opinion of each of the respondents about the clinical services for early detection, diagnosis and treatment for children with ASD under 6 years of age. Specifically, the aim is to gather information about the personal experiences that the respondents have had in relation to these issues, including age of identification and diagnosis; demographic characteristics of families; opinions on the organization of services for young children with ASD; and suggestions for ways to improve this type of service.

Ethics approval has been given by the University of Salamanca. Copies of the ethics approval letter are available if you wish to see them.

We will not ask for any identifying information – e.g. your name, your full address
Participation will last approximately 15 minutes.

Thank you for taking the time to read this Information. If you wish to complete this survey, please check all the following items

Please read the consent form below.
I have read and understood the survey information sheet
I understand that all data collected from this survey will be anonymously coded and stored confidentially and securely
I am willing to take part in this research survey

If you wish to complete this survey, please click "I agree to participate" to continue

I agree to participate
- ❍ Yes
- ❍ No

BACKGROUND INFORMATION The questions in this initial section will ask you about your gender, where you live, your age, your education, the current monthly household income of the family the child with autism live in (optional), the number of people living in your home, and three more questions about age, gender and verbal ability of the child with autism.

1. Please select your country and city of residence
   Country
   City

2. Gender
- ❍ Male
- ❍ Female

3. How old are you?

4. What is your relationship with the child with autism?
- ❍ Mother or father
- ❍ Grandparent
- ❍ Sibling
- ❍ Other, please specify: _____

5. How many people live in the municipality where you reside?
- ❍ Less than 10.000.
- ❍ Between 10.000 to 50.000.
- ❍ Between 50.000 to 150.000.
- ❍ Between 150.000 to 1.000.000.
- ❍ More than 1.000.000.

6. Please indicate the highest level of education reached
- ❍ No formal education completed
- ❍ Primary education (or similar: elementary, middle school …)
- ❍ Secondary school, High school
- ❍ University degree
- ❍ Professional training
- ❍ College Education (Bachelor degree or higher)

7. How many people live permanently in the household of the child with ASD?

	1	2	3	4	5	6	7	8	9	10
Number of people										

8. This question is optional: Please state the current monthly household income (currency in the relevant country)

9. What is the child's with ASD age?

10. Child's gender
- ❍ Male
- ❍ Female

11. What is the child's verbal ability?
- ❍ Does not talk
- ❍ Uses single words only (e.g. "daddy", "mommy")
- ❍ Uses two- or three- word phrases (e.g. "want cookie")
- ❍ Uses sentences with four or more words (e.g. "I want a biscuit")
- ❍ Uses complex sentences (e.g. "When we get home, can I have a biscuit?")

DETECTION In this section you are going to answer questions about the process of detecting the child's difficulties with ASD before knowing the diagnosis. After this section, there will be another specific for the diagnostic process.

12. How old was the child when you or someone else first have concerns about he/she had developmental problems?
(Table Truncated to 63 Columns)

13. What was that first concern? Apply more than one, if any
- ❑ Does not direct large smiles or expressions of joy to the adult at 6 months
- ❑ Does not exchange sounds, smiles or facial expressions since 9 months
- ❑ Do not babble at 12 months
- ❑ Does not make gestures (pointing, saying goodbye by hand, etc.) at 12 months
- ❑ Does not say simple words at 16 months
- ❑ Does not say spontaneous phrases of 2 words (not simply echoics) at 24 months
- ❑ ANY loss in ANY area (language or social skill) at ANY age
- ❑ Other. please, specify _____

14. Who was the first person who suspected that something was wrong with the child's development?
- ❍ You detected the problem
- ❍ A family member. Please, specify _____
- ❍ The pediatrician or nurse from public health service
- ❍ The pediatrician or nurse from private health services
- ❍ A teacher or school staff from (nursery, kindergarten, school, etc.)
- ❍ Other. Please specify _____

15. The person who raised the first concerns about the child's development was based on (check all that apply)

- ❑ His / her knowledge about this child with ASD
- ❑ His / her own experience and knowledge on child development in general
- ❑ A questionnaire we filled in the doctor's office/ school (name of the questionnaire if you remember it) _____
- ❑ A program specifically aimed at identifying problems on communicative and social development available in health / school / social services in my city.
- ❑ Other. Please, specify _____

16. Do you consider it was easy to have access to information about programs and early detection services where you are residing?

- ❍ Yes
- ❍ No

Please, explain why

17. What was the next step in the detection process?

- ❍ We ourselves had to look for a diagnostic service
- ❍ Someone gave us a phone call to refer us to a diagnostic service
- ❍ We received a letter with a medical appointment from the hospital
- ❍ The professional who had the first concern refer us directly to a specialized service
- ❍ Other. Please, specify _____

18. How much time (in months) passed from the confirmation of the first concerns until the child was attended by an autism specialist?

- ❍ Less than 1 month
- ❍ From 1 to 2 months
- ❍ From 3 to 4 months
- ❍ From 5 to 6 months
- ❍ More than 6 months
- ❍ Other. Please, specify _____

19. Did you have any professional guidance and support to address your first concerns?

- ❍ Yes
- ❍ No

Please check all that apply if it was more than one.

- ❑ Paediatrician
- ❑ Psychologist
- ❑ Psychiatrist
- ❑ Nurse
- ❑ Neuropediatrician
- ❑ Kindergarten/school teacher
- ❑ Other. Please specify: _____

20. How adequate do you consider the detection process?

	Extremely adequate	Moderately adequate	Slightly adequate	Neither adequate nor inadequate	Slightly inadequate	Moderately inadequate	Extremely inadequate
Detection process followed by the diagnostic evaluation	❍	❍	❍	❍	❍	❍	❍
The staff's qualifications who attend the child during the detection process	❍	❍	❍	❍	❍	❍	❍
The degree to which the professionals involved in the process listened and took into account your concerns	❍	❍	❍	❍	❍	❍	❍

21. If you have any suggestions about the detection process of early ASD signs, please specify

DIAGNOSIS In this section you will be asked about the diagnosis process of ASD. We are interested in your perception of the direct care you have been receiving at the different centers or institutions that have cared for your child during the diagnostic process. The questions incorporated in this section refer to all professionals and institutions that have been directly involved in the diagnostic process of your child, and with whom you have been in direct contact. They can be, for example, family doctors, pediatricians, nurses, physiotherapists, speech therapists, psychologists, teachers, etc.

22. Has the child received any of the following diagnoses?

❍ Autism Spectrum Disorder (ASD)
❍ Pervasive Developmental Disorder
❍ Autistic Disorder / Childhood autism
❍ Asperger´s syndrome /Asperger´s disorder
❍ Atypical autism
❍ Pervasive developmental disorder not otherwise specified
❍ Other. Please, specify _____

23. At what age was your child given an autism spectrum disorder diagnosis?

24. Do you recall how passed (approximately), from the suspicion of the child's developmental problems until the diagnosis confirmation?

- ❍ Less than 1 month
- ❍ From 1 to 2 months
- ❍ From 3 to 4 months
- ❍ From 5 to 6 months
- ❍ More than 6 months
- ❍ Other. Please, specify _____

25. To what extent did the professionals who attend your child...

	A lot	Quite	Regular	Little bit	Very little	Never
Took enough time to talk to you about your child's problems?	❍	❍	❍	❍	❍	❍
... Helped you feel more secure about yourself as a parent?	❍	❍	❍	❍	❍	❍
... Offered support, as well as information?	❍	❍	❍	❍	❍	❍
Gave you the opportunity to make decisions about using different tests?	❍	❍	❍	❍	❍	❍
... Informed you about the results of your child's assessment?	❍	❍	❍	❍	❍	❍
... Gave you information about the services offered by institutions or other services in your community?	❍	❍	❍	❍	❍	❍
... Gave you advise on how to access more information or how to contact other parents in the same situation (e.g. contact with parent organizations	❍	❍	❍	❍	❍	❍

or specialized centers)? ... were they coordinated so that all the services involved provided information to arrive at a coherent diagnosis?	❍	❍	❍	❍	❍	❍

26. Which professionals assisted you in the diagnostic process? Apply more than one, if any

- ❑ Psychologist
- ❑ Pediatrician
- ❑ Nurse
- ❑ Neuropediatrician
- ❑ Psychiatrist
- ❑ Other. Please, specify _____

27. Did you receive advice or information from the professionals who gave you the ASD diagnosis report? (Check the box of the aspects on which you received appropriate or sufficient information) Apply more than one, if any

- ❑ Medical needs (specialists, medicine, genetic counselling…)
- ❑ Educational needs (centres, support…)
- ❑ Social needs (organizations, family support…)
- ❑ Materials (bibliography, agencies, web pages…)
- ❑ Other. Please, specify _____
- ❑ No

28. Did you receive written information about the diagnosis?

- ❍ Yes
- ❍ No

29. How adequate do you consider the diagnostic process?

	Extremely adequate	Moderately adequate	Slightly adequate	Neither adequate nor inadequate	Slightly inadequate	Moderately inadequate	Extremely inadequate
The time passed since the first suspicion of developmental problems until the confirming diagnosis	❍	❍	❍	❍	❍	❍	❍
The professional level of the personnel who attended the child in the diagnostic process	❍	❍	❍	❍	❍	❍	❍
The information and support you received by these professionals	❍	❍	❍	❍	❍	❍	❍
The evaluation process and diagnosis	❍	❍	❍	❍	❍	❍	❍

30. If you have any suggestions for the diagnostic programs, please specify

EARLY INTERVENTION In this section of the survey you will be asked about the type and quantity of Early Intervention services that your child has received in the last 12 months. You will be asked to say the time that passed since they received the diagnosis of their child until the treatment began, type of intervention your child receives, how many hours of treatment your child usually receives per week (For example, if your child receives 1 hour of therapy every 2 weeks, you should enter 0.5 hours per week), the degree of your involvement in the treatment, and your opinion about the treatment your child receives

31. Does the child currently receive early intervention?

❍ Yes
❍ No, but the child has received until less than 12 months
❍ No

31.1. Do you remember how much time passed since your child's diagnosis until the intervention program started?

❍ Yes. Please, specify (in months) _____
❍ No

31.2. Could you say what type of intervention, both private and public, the child currently receives?

❑ Public. Please, specify _____
❑ Private. Please, specify _____

31.2.1. How many public intervention sessions does the child receive on a weekly basis? Please indicate how long each session lasts on average

31.2.2. How many private intervention sessions does the child receive on a weekly basis? Please indicate how long each session lasts on average

31.3. How are the intervention sessions? (You can select more than one option)

❑ In group
❑ Individual
❑ Other. Please, specify _____

32. To what extent do you participate in the intervention sessions with your child?

❍ Very actively
❍ Actively
❍ Occasional participation
❍ I don't participate

Please, explain why

33. How far is the centre where your child with ASD receives regular interventions?

34. How long does it take you to get to that service?

31.1. How long the child does not receive early intervention?

❍ Less than a year
❍ Between 1 and 2 years
❍ Between 2 and 3 years
❍ More than 3 years
❍ The child has not received any intervention

31.2. Do you remember how much time passed since your child's diagnosis until the intervention program started?

❍ Yes. Please, specify (in months) _____
❍ No

31.3. Could you say what type of intervention, both private and public, the child received?

- ❑ Public. Please, specify _____
- ❑ Private. Please, specify _____

31.3.1. How many public intervention sessions did the child receive on a weekly basis? Please indicate how long each session lasted on average

31.3.2. How many private intervention sessions did the child receive on a weekly basis? Please indicate how long each session lasted on average

31.4. How were the intervention sessions? (You can select more than one option)

- ❑ In group
- ❑ Individual
- ❑ Other. Please, specify _____

32. To what extent did you participate in the intervention sessions with your child?

- ❍ Very actively
- ❍ Actively
- ❍ Occasional participation
- ❍ I don't participate

Please, explain why

33. How far was the centre where your child with ASD received regular interventions?

34. How long did it take you to get to that service?

35. Has your child received

	Yes	No
Behavioural treatment (e.g. Applied Behaviour Analysis (ABA); Pivotal Response Training (PRT); Lovaas; Discrete Trial Training (DTT) ...)	❍	❍
Developmental treatment (e.g. Relationship Development Intervention (RDI); Early Start Denver Model (ESDM) ...)	❍	❍
Relationship-based treatment (e.g. Developmental Individual Difference Relationship based (DIR Model) Floortime; Thérapie d'Echange et de Développement (TED) ...)	❍	❍
Portage intervention	❍	❍
Psychoanalytic treatment	❍	❍
Speech and language therapy	❍	❍
Occupational therapy / physiotherapy	❍	❍
Parent training / coaching /counselling to help you with your child	❍	❍
Another psychological / educational / behavioural treatment (not previously specified)	❍	❍

36. How adequate do you consider the intervention process?

	Extremely adequate	Moderately adequate	Slightly adequate	Neither adequate nor inadequate	Slightly inadequate	Moderately inadequate	Extremely inadequate
The waiting time to receive a public/private intervention program	❍	❍	❍	❍	❍	❍	❍
The number of sessions that the child receives	❍	❍	❍	❍	❍	❍	❍
Your level of participation in the intervention sessions	❍	❍	❍	❍	❍	❍	❍
The information that you received about the intervention programs	❍	❍	❍	❍	❍	❍	❍

37. If you have any suggestions for early intervention programs, please enter it below

asdeu
Autism Spectrum Disorders in the European Union

Professional survey - final

Identification, diagnosis and early intervention for young children with autism spectrum disorder (ASD)

A survey study to improve support for young children with ASD

Before you respond to the survey, we want to explain the reason for this research and what it will mean for you. Please take the time to read this information carefully. For more information, do not hesitate to contact Ricardo Canal (rcanal@usal.es).

This survey is designed to collect information from people who have or have recently had any direct involvement with screening, diagnosis or treatment services for young children under 6 years of age with ASD. Direct involvement means you are a professional who currently works, or has worked in recent years, in the field of autism and knows firsthand the kind of services provided to children under 6 years of age in their community.

The objective of the survey is to assess the opinion of each of the respondents about the clinical services for early detection, diagnosis and treatment for children with ASD under 6 years of age. More specifically, the aim is to gather information about the personal experiences that the respondents have had in relation to these issues, including age of identification and diagnosis; demographic characteristics of respondents; opinions on the organization of services for young children with ASD; and suggestions on ways to improve these type of services.

Ethics approval has been given by the University of Salamanca. Copies of the ethics approval letter are available if you wish to see them.

If you provide personal information this will only be used to contact you in case we need more detailed information or a clarification. Completing the survey will take approximately 15 minutes.

Thank you for taking the time to read this Information. If you wish to complete this survey, please check all the following items

Please check all the following items:
I have read and understood the survey information sheet
I understand that all data collected from this survey will be anonymously coded and stored confidentially and securely
I am willing to take part in this survey

If you wish to complete this survey, please click "I agree to participate" to continue

I agree to participate
- ❍ Yes
- ❍ No

BACKGROUND INFORMATION The questions in this initial section will ask you about your gender, where you work, your age, your job, and two more questions about your experience with children with autism.

1. How old are you?

2. Gender
- ❍ Male
- ❍ Female

3. What country and city do you work in?
   Country
   City

4. How many people live in the municipality where you reside?
- ❍ Less than 10.000.
- ❍ Between 10.001 to 50.000.
- ❍ Between 50.001 to 150.000.
- ❍ Between 150.001 to 1.000.000.
- ❍ More than 1.000.000.

5. What job or career title fits you best? (Select only one, whichever is your main job)
- ❍ Counsellor
- ❍ Department head or chief
- ❍ Director of an organization
- ❍ General practitioner
- ❍ Psychiatrist
- ❍ Nurse
- ❍ Other medical professional, please specify _____
- ❍ Psychologist
- ❍ Social worker
- ❍ Teacher
- ❍ Teaching assistant
- ❍ Mental health therapist
- ❍ Physical or occupational therapist
- ❍ Other, please specify _____

6. Do you work or have you worked in recent years with children with ASD under six years?
- ❍ Yes
- ❍ No

7. How many years in total have you been working in jobs that brought you into contact with young children with autism (younger than 6 years old)?

- ❍ < 1 year
- ❍ 1 - 3 years
- ❍ 3 - 5 years
- ❍ > 5 years

DETECTION In this section you are going to answer questions about the process of detecting the child's difficulties with ASD before knowing the diagnosis. After this segment, there will be another one specific for the diagnostic process.

8. Do you participate or have participated in the last 6 years in an ASD early detection program?

- ❍ Yes
- ❍ No

We would like to contact you directly so that you can describe the program in detail. Can you provide us with an email address?

We would like to contact someone directly involved in the ASD early detection program. Could you provide us with an email address to get more details on how the detection is carried out in your organization?

9. Does the centre / institution where you work provide any specific services for early detection of children with ASD?

- ❍ Yes
- ❍ No

Please specify (You can select more than one option):

- ❑ Training for awareness raising for families
- ❑ Awareness aimed at professionals
- ❑ Developmental surveillance
- ❑ Search for early signs during routine consultations
- ❑ Application of a standardized questionnaire. Name of the questionnaire _____
- ❑ Other, please specify _____

10. Does the centre/organization provide information about early ASD signs and/or early detection programs running in the area or region where your centre provides services?

- ❍ Yes
- ❍ No

Please specify (select at least one from each row)

Target audience	❑ Families	❑ Professionals	❑ Media	❑ Other
Promoting institutions	❑ Educational services	❑ Health services	❑ Social services	❑ Other
Resources	❑ Written material	❑ Meetings and seminars	❑ Posters / brochures	❑ Other
Most used media for disseminating information	❑ Email / website	❑ Post mail	❑ Phone	❑ Other
Frequency	❑ Only once	❑ Seasonal (campaigns)	❑ Permanently	❑ Other

If you have selected other target audience, please, specify

If you have selected other promoting institutions, please, specify

If you have selected other resources, please, specify

If you have selected other media for disseminating the information, please, specify

If you have selected other frequency, please, specify

11. Does your centre/organization collaborate with any other agencies/institutions in your region or country of residence to improve ASD early detection?

❍ Yes
❍ No

12. Do you know how much time passes, on average, from the parent's first concerns until they request an appointment at your centre/service?

❍ Less than 1 month
❍ 1-2 months
❍ 3-4 months
❍ 5-6 months
❍ 7-8 months
❍ 9-10 months
❍ 11 months or more
❍ I do not know

13. Do you have information on the average age at which you or other professionals in your institution usually identify ASD early signs in the population area where you are attending?

- ❍ Less than 12 months
- ❍ 13-18 months
- ❍ 19-24 months
- ❍ 25-32 months
- ❍ 33-39 months
- ❍ 40-46 months
- ❍ 47-53 months
- ❍ 54-60 months
- ❍ 61 months or more
- ❍ I do not know

14. Does your region or country have any practice guidelines to serve people with autism in the detection and identification phase?

- ❍ Yes
- ❍ No

15. Which professionals (most frequently) assist the children during the process of identifying the first signs in your service/institution? You can select more than one option

- ❑ Paediatrician
- ❑ Psychologist
- ❑ Psychiatrist
- ❑ Nurse
- ❑ Neuropediatrician
- ❑ Kindergarten/school teacher
- ❑ Other. Please specify: _____

16. In the absence of an ASD screening program in your region or country: please describe briefly how early ASD signs are detected in young children

17. How adequate do you consider the detection process?

	Extremely adequate	Moderately adequate	Slightly adequate	Neither adequate nor inadequate	Slightly inadequate	Moderately inadequate	Extremely inadequate
Detection process followed by the diagnostic evaluation	❍	❍	❍	❍	❍	❍	❍
The staff's qualifications who attend the child during the detection process	❍	❍	❍	❍	❍	❍	❍
The degree to which professionals involved in the process listen and take into account the concerns of parents	❍	❍	❍	❍	❍	❍	❍

18. If you have any suggestions about the detection process of early ASD signs, please specify

DIAGNOSIS In this section you will be asked questions about the diagnostic process of ASD. We are interested in your perception of the centers or institutions that have cared for the child during the diagnostic process. The questions incorporated in this section refer to all professionals and institutions that have been directly involved in the diagnostic process of children with ASD.

19. Are you directly involved or have you been involved in the last 6 years in conducting diagnostic processes for children with ASD?

❍ Yes
❍ No

We would like to contact you directly for more details on how the diagnostic process is carried out in your organization. Can you provide us with an email address?

We would like to contact someone directly involved in the diagnostic process. Could you provide us with an email address to get more details on how the diagnostic process is carried out in your organization?

20. What are the institutions/services that most frequently provide diagnoses of ASD in your country?

	Psychiatry	Pediatrics	Neurology	Psychology	Other
Public Hospital/Centre	❑	❑	❑	❑	❑
Private Hospital	❑	❑	❑	❑	❑
Local/regional parent organization	❑	❑	❑	❑	❑
Private consultation with a specialist	❑	❑	❑	❑	❑
Other. Please, specify	❑	❑	❑	❑	❑

21. Does your centre/service (You can select more than one option):
- ❑ Conduct diagnostic assessments
- ❑ Refer clients to other services to make a diagnosis
- ❑ Take the evaluations conducted by other services or centers

22. Who refers (most frequently) children under 6 years of age with autism or possible autism to your service/institution? You can select more than one option:
- ❑ The pediatricians or nurses from public health services
- ❑ The pediatricians or nurses from private health services
- ❑ Teachers or school staff from public education services (nursery, kindergarten, school, etc.)
- ❑ Teacher or school staff from private education services (nursery, kindergarten, school, etc.)
- ❑ Professionals from public social services (agencies or other)
- ❑ Professionals from private social services (agencies or other)
- ❑ Other. Please, specify _____

23. Which professionals (most frequently) assist the children during the process of diagnosis? You can select more than one option:
- ❑ Paediatrician
- ❑ Psychologist
- ❑ Psychiatrist
- ❑ Nurse
- ❑ Neuropediatrician
- ❑ Other. Please, specify _____

24. Does your centre have a specific requirement regarding the type of professional who must take part in the diagnostic process (e.g., a doctor, teacher or psychologist)?

❍ Yes
❍ No

Please, specify. You can select more than one option

❑ Paediatrician
❑ Psychlogist
❑ Psychiatrist
❑ Nurse
❑ Neuropediatrician
❑ Kindergarten/school teacher
❑ Other. Please, specify _____

25. Does your centre require that any specific instrument be used during the diagnostic process?

❍ Yes
❍ No

Please, specify. You can select more than one option

Autism Symptomatology	❑ Autism Diagnostic Observation Schedule (ADOS-2)	❑ Social Communication Questionnaire (SCQ)	❑ Autism Spectrum Quotient (AQ)	❑ Autism Diagnostic Interview Revised (ADI-R)	❑ Social Responsiveness Scale (SRS)	❑ Other
IQ measures	❑ Wechsler Preschool & Primary Scale of Intelligence (WPPSI-R)	❑ Mullen Scales of Early Learning (MSEL)	❑ Merrill-Palmer-Revised Scales of Development (MP-R)	❑ Leiter International Performance Scale- R (Leiter-R)	❑ Bayley Scales of Infant Development (BSID-II/III)	❑ Other
Behaviour & Comorbid traits	❑ Vineland Adaptive Behaviour Scales (VABS-II)	❑ Child Behaviour Checklist (CBCL)	❑ Strengths & Difficulties Questionnaire (SDQ)	❑ Conners' Rating Scale (CRS)	❑ Other	❑ Other

If you have selected other instrument for autism symptomatology, please specify

If you have selected other instrument for IQ measures, please specify

If you have selected other instrument for behaviour & comorbid traits, please specify

26. Do you know the average age of the children to whom a diagnosis of ASD occurs in your centre / service?

- ❍ Less than 12 months
- ❍ 13-18 months
- ❍ 19-24 months
- ❍ 25-32 months
- ❍ 33-39 months
- ❍ 40-46 months
- ❍ 47-53 months
- ❍ 54-60 months
- ❍ 61 months or more
- ❍ I do not know

27. Do you know how much time passes, on average, from the first meeting with the specialist on ASD detection until the child receives a diagnosis of ASD?

- ❍ Less than 1 month
- ❍ 1-2 months
- ❍ 3-4 months
- ❍ 5-6 months
- ❍ 7-8 months
- ❍ 9-10 months
- ❍ 11 months or more
- ❍ I do not know

28. Does your centre/service give information to the family after the ASD diagnosis?

- ❍ Yes
- ❍ No

Please, specify. Check the box of the aspects on which parents usually receive appropiate or sufficient information. You can select more than one option

- ❑ Medical needs (specialists, medicine, genetic counselling…)
- ❑ Educational needs (centres, support…)
- ❑ Social needs (organizations, family support…)
- ❑ Materials (bibliography, agencies, web pages…)
- ❑ Other. Please specify _____

29. Do you follow a classification of mental disorders manual for the diagnosis of children with ASD?

- ❍ Yes
- ❍ No

Please, specify. You can select more than one option

- ❑ DSM-IV-TR
- ❑ DSM-5
- ❑ ICD-10
- ❑ Other. Please specify _____

30. How adequate do you consider the diagnostic process?

	Extremely adequate	Moderately adequate	Slightly adequate	Neither adequate nor inadequate	Slightly inadequate	Moderately inadequate	Extremely inadequate
The time passed since the first suspicion of developmental problems until the confirming diagnosis	❍	❍	❍	❍	❍	❍	❍
The staff's qualifications who attend the children during the diagnostic process	❍	❍	❍	❍	❍	❍	❍
The evaluation process and diagnosis	❍	❍	❍	❍	❍	❍	❍

31. If you have any suggestions for the diagnostic programs, please specify

BIOMARKERS Biomarkers in the process of detection and diagnosis

32. Do you think it is feasible?

	Yes, we already do this	Yes, but this is currently not common practice	Yes, but I don't think this would be useful	No, but I believe this would be useful	No, and I don't think this would be useful
To use EEG in the early detection of ASD?	❍	❍	❍	❍	❍
To use MRI (under sedation) in the early detection of ASD?	❍	❍	❍	❍	❍
To use blood assessment (e.g. to test for immune system parameters, hormone levels…) in the early detection of ASD?	❍	❍	❍	❍	❍
To use urine samples (e.g. to assess metabolic parameters) in the early detection of ASD?	❍	❍	❍	❍	❍
To use a neonatal blood draw for bilirubin testing in the early detection of ASD?	❍	❍	❍	❍	❍
To use eye tracking (e.g. to assess attention) in the early detection of ASD?	❍	❍	❍	❍	❍
To use a motor assessment in the early	❍	❍	❍	❍	❍

detection of ASD? To use questionnaires to assess temperament characteristics in the early detection of ASD?	❍	❍	❍	❍	❍

Please provide your personal email if you want to contact us for further information

EARLY INTERVENTION In this section of the survey you will be asked about the type and quantity of Early Intervention services in your region / country

33. Are there any regional / national reference service or specialized centres for ASD early intervention in your region or country? You can select more than one option

❍ Yes. Please, specify _____
❍ No. This type of services are carried out by general early intervention teams for all types of children with developmental difficulties
❍ There are no early intervention services for ASD

34. Are you a member or have you been a member in the last 6 years of an early intervention team that serves children with ASD?

❍ Yes
❍ No

We would like to contact you directly so that you can describe the early intervention program you are conducting for children with ASD in detail. Can you provide us with an email address?

We would like to contact someone directly involved in the ASD early intervention program. Could you provide us with an email address to get more details on how the intervention is carried out in your organization?

35. Please indicate some of the fundamental principles your early intervention model for children with ASD is based on (e.g., parent involvement, based on natural contexts, using behavioral methods, individualized objetives, etc.)?

1
2
3
4
5
6
7
8

36. What is the average age at which your centre often stars early intervention for children with ASD?

- ❍ Less than 12 months
- ❍ 13-18 months
- ❍ 19-24 months
- ❍ 25-32 months
- ❍ 33-39 months
- ❍ 40-46 months
- ❍ 47-53 months
- ❍ 54-60 months
- ❍ 61 months or more
- ❍ I do not know

37. Do you know how much time passes from the children's diagnosis until the intervention program starts?

- ❍ Less than 1 month
- ❍ 1-2 months
- ❍ 3-4 months
- ❍ 5-6 months or more
- ❍ I do not know

38. Would you recommend any particular treatment methods for individuals with autism?

- ❍ Yes. Please specify _____
- ❍ No

Please, specify the reason (You can select more than one option)

- ❑ Because in our team we are trained in that model of intervention
- ❑ Because it is based on the evidence
- ❑ Because we consider it appropriate for children with ASD
- ❑ Because it is the most used in our region / country
- ❑ Other reasons (specify) _____

39. How many intervention sessions and time could the children receive weekly in your service / centre?

40. How are the intervention sessions in your service / centre? (You can select more than one option)

- ❍ In group
- ❍ Individual
- ❍ Other. Please, specify _____

41. What is the level of parent participation in the intervention programs in your service / centre?

- ❍ Very active
- ❍ Active
- ❍ Occasional participation
- ❍ Parents hardly participate

Please, explain why

42. Does your service / centre give information to the family about the ASD intervention program?

- ❍ Yes
- ❍ No

Please, specify the type of information (You can select more than one option)

- ❑ Information on evidence of program results
- ❑ Information on the suitability of the program to the characteristics of the child
- ❑ Information on the economic cost of the program
- ❑ Information about how parents will be involved
- ❑ Information about the child's progress
- ❑ Other. Please, specify _____

43. How adequate do you consider the intervention process?

	Extremely adequate	Moderately adequate	Slightly adequate	Neither adequate nor inadequate	Slightly inadequate	Moderately inadequate	Extremely inadequate
The number of sessions that the child receives	❍	❍	❍	❍	❍	❍	❍
The level of parents' participation in the intervention sessions	❍	❍	❍	❍	❍	❍	❍
The information that the parents received about the intervention programs	❍	❍	❍	❍	❍	❍	❍

44. If you have any suggestions for early intervention programs, please specify

Table 5. Predictors of positive satisfaction

	Detection			Diagnosis				Intervention			
	Process leading to diagnostic evaluation	Staff qualifications	Professionals took into account family's concerns	Delay between detection and diagnosis	Professionalism of practitioners	Information and support received	Evaluation process and diagnosis	Delay between diagnosis and start of intervention	Number of sessions	Parental participation	Information and support received
	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)
Family members											
Child's age at detection (0-18 months)	**1.97 (1.42-2.74)**	**1.85 (1.31-2.61)**	**1.80 (1.28-2.54)**	**1.57 (1.12-2.21)**	**1.55 (1.03-2.34)**	**1.65 (1.15-2.36)**	1.16 (0.80-1.71)	**1.54 (1.07-2.22)**	1.20 (0.84-1.72)	**1.50 (0.98-2.23)**	**1.65 (1.14-2.38)**
Delay in access to detection (>6 months)	**0.39 (0.28–0.55)**	**0.45 (0.30–0.66)**	**0.61 (0.43-0.88)**	**0.54 (0.39–0.74)**	0.86 (0.57–1.31)	0.72 (0.51–1.01)	0.75 (0.51–1.09)	0.84 (0.60–1.18)	0.88 (0.62–1.23)	0.77 (0.52–1.13)	1.09 (0.77–1.55)
Child's age at diagnosis (0-24 months)	–	–	–	**4.84 (2.73-8.57)**	**2.49 (1.12-5.54)**	**4.24 (2.18-8.23)**	**3.10 (1.51-6.38)**	**3.51 (2.00-6.16)**	**2.13 (1.22-3.68)**	**2.38 (1.24-4.55)**	**2.27 (1.56-4.90)**
Delay in access to diagnosis (>6 months)	–	–	–	**0.07 (0.03–0.16)**	**0.24 (0.09–0.62)**	**0.20 (0.10–0.42)**	**0.31 (0.15–0.65)**	1.04 (0.65–1.66)	0.99 (0.62–1.57)	0.95 (0.56–1.61)	0.92 (0.57–1.49)
Child's age at intervention (0-36 months)	–	–	–	–	–	–	–	**2.23 (1.06-4.59)**	**2.02 (1.20-3.78)**	**3.45 (2.08-5.94)**	**2.89 (1.46-5.21)**
Delay in access to intervention (>6 months)	–	–	–	–	–	–	–	**0.92 (0.88–0.95)**	**0.96 (0.93–0.98)**	0.99 (0.96–1.02)	**0.95 (0.92–0.98)**
Professionals											
Child's age at detection (0-18 months)	2.95 (0.75–11.6)	4.02 (0.96–16.8)	0.99 (0.21–4.50)	0.46 (0.09–2.30)	0.78 (0.05–12.6)	–	1.30 (0.14-11.7)	2.08 (0.32–12.7)	2.01 (0.36–11.1)	2.86 (0.47–17.3)	2.62 (0.22–30.8)
Delay in access to detection (>6 months)	**0.19 (0.07–0.55)**	**0.32 (0.09–0.91)**	**0.29 (0.07–0.95)**	1.02 (0.29-3.63)	0.32(0.01–7.90)	–	0.79 (0.11–5.84)	0.45 (0.15–0.12)	0.37 (0.14–0.97)	0.55 (0.19–1.58)	0.50 (0.16–1.54)
Child's age at diagnosis (0-24 months)	–	–	–	0.93 (0.15–5.59)	0.13(0.01–4.12)	–	0.58 (0.05–7.17)	0.85 (0.25–4.59)	0.25 (0.04–1.56)	0.68 (0.11–4.11)	1.08 (0.40–2.94)
Delay in access to diagnosis (>6 months)	–	–	–	0.66 (0.16–2.75)	0.41(0.02–6.73)	–	1.14 (0.14–9.05)	0.85 (0.32–2.15)	0.93 (0.38–2.29)	0.76 (0.28–2.08)	1.11 (0.38–3.27)
Child's age at intervention (0-36 months)	–	–	–	–	–	–	–	2.45 (0.89–5.37)	1.48 (0.39–5.65)	1.40 (0.25–7.87)	4.07 (0.39–42.3)
Delay in access to intervention (>6 months)	–	–	–	–	–	–	–	**0.40 (0.23–0.70)**	0.43 (0.19–1.01)	0.65 (0.25–1.65)	0.68 (0.24–1.91)

The table reports odds ratios (ORs) and the corresponding 95% confidence intervals (CIs). Predictors significant at $p < 0.05$ are indicated in bold.

(-): Not applicable. Not asked or not possible to calculate

ARTÍCULO III: PUESTA EN MARCHA DE UN PROGRAMA DE ATENCIÓN TEMPRANA

Bejarano-Martín, Á., Canal-Bedia, R., Magán-Maganto, M., Hernández Fabián, A., Calvarro Castañeda, A. L., Manso de Dios, S., Malmierca García, P., Díez Villoria, E., Jenaro Río, C., & Posada de la Paz, M. (2021). Effect of a Focused Social and Communication Intervention on Preterm Children with ASD: A Pilot Study. *Journal of Autism and Developmental Disorders*. https://doi.org/10.1007/s10803-021-05068-8

Resumen

Introducción. Aunque los avances en los cuidados intensivos neonatales han mejorado mucho las tasas de supervivencia de los niños prematuros, la incidencia de los trastornos del neurodesarrollo en este grupo sigue siendo elevada, siendo el trastorno del espectro autista (TEA) uno de los más frecuentes. ***Objetivos.*** Con este fin, llevamos a cabo una intervención socio-comunicativa destinada a investigar la eficacia en las habilidades socio-comunicativas. ***Método.*** Participaron en el estudio 18 niños (prematuros y nacidos a término con TEA y niños prematuros) de entre 18 y 20 meses de edad. ***Resultados.*** Nuestros resultados indican que la mayoría de los participantes en los grupos de intervención registraron mejoras significativas en términos de habilidades socio-comunicativas, desarrollo cognitivo y lenguaje. ***Discusión.*** En consecuencia, estos datos

piloto subrayan la necesidad de seguir investigando y aplicando intervenciones tempranas en niños prematuros con TEA.

Palabras clave. Trastorno del espectro autista, Prematuridad, Intervención, Social, Comunicación

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ORIGINAL PAPER

Effect of a Focused Social and Communication Intervention on Preterm Children with ASD: A Pilot Study

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Abstract

While advances in intensive neonatal care have greatly improved survival rates among preterm infants, incidence of neurodevelopmental disorders in this group is still high, with autism spectrum disorder (ASD) being one of the most frequent. To this end, we conducted a social-communication intervention aimed at investigating efficacy in social-communicative skills. Eighteen children (preterm and full-term with ASD and preterm children) aged 18 through 20 months participated in the study. Our findings indicate that most participants in the intervention groups registered significant improvements in terms of socio-communicative skills, cognitive development, and language. Accordingly, these pilot data underscore the need for further research and implementation of early interventions in young preterm children with ASD.

Keywords Autism spectrum disorder · Preterm · Intervention · Social · Communication

Introduction

Globally, 15 million babies are born prematurely (< 37 weeks' gestation) each year (Blencowe et al., 2013). While advances in neonatal intensive care have greatly improved survival rates among premature infants (Anderson et al., 2016), this increase has nonetheless been accompanied by a worrying rise in the incidence of neurodevelopmental disorders in this group (Cheong et al., 2017). Low gestational age may increase the vulnerability of the developing brain and, along with other exposures associated with preterm birth, might act as casual pathways for autism spectrum disorder (ASD; Joseph et al., 2017). ASD is a neurodevelopmental disorder of early onset, characterized by deficits in social communication, accompanied by restricted and repetitive patterns of behavior, interests or activities which have

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significant negative consequences for daily life (American Psychological Association, 2013). A recent meta-analysis reported an ASD prevalence of 7% (range 4% through 9%) in this population, based on a total sample of 3366 preterm infants (Agrawal et al., 2018). This difference is relevant when compared to the general population, in which the overall prevalence of ASD is 1% (Lord et al., 2020).

Preterm children have an increased likelihood of ASD, and those who eventually receive an ASD diagnosis display fewer early socio communicative behaviors than do typically developing children (Mateus et al., 2020). Furthermore, they register significantly higher scores in the Autism Diagnostic Observation Schedule (Lord et al., 2000) than do preterm peers who eventually do not receive a diagnosis of ASD, thus indicating less optimally developed social and communicative skills (De Groote et al., 2006; Matheis et al., 2018). Recent studies observing infant communication skills in mother-infant interaction at 12 months found that preterm infants displayed less frequent social and communicative interaction and produced fewer pointing, giving, and representational gestures than did full-term infants (Benassi et al., 2016; Sansavini et al., 2015). This population likewise shows different early cognitive and language development trajectories (Allotey et al., 2018; Chen et al., 2020).

Early intervention programs for the ASD population, such as social and communication interventions (SCIs), have been purpose-designed to reduce ASD signs. The aim of such early interventions is to prevent the consequences of these difficulties, by initiating proactive treatment even before the child receives a formal diagnosis. In addition to being shown to improve social and communicative skills, cognitive development, language, and adaptive behavior (Bejarano-Martín et al., 2020a; Gates et al., 2017; Zwaigenbaum et al., 2015), it has also been suggested that early interventions can reduce the burden of ASD and improve the quality of life of children and their families (Bejarano-Martín, 2020b; Salomone et al., 2016; Fletcher-Watson et al., 2017). SCIs target behaviors such as imitation, joint attention (JA) and play, which have a fundamental role in developing social skills, social-emotional communicative functions, and theory of the mind, as well as language and play skills (Adamson et al., 2019; Dohmen et al., 2016; Kasari et al., 2015; Pickard & Ingersoll, 2015).

A number of SCIs have been implemented with the aim of reducing initial symptoms of ASD in children with increased likelihood of ASD before a formal diagnosis is made (Landa, 2018; Rogers & Vismara, 2014). Similarly, several early intervention programs have sought to improve different social skills (e.g., imitation, joint attention, and play) of preterm infants (e.g., Bagner et al., 2010; Chernego et al., 2018). To date, however, no early intervention program has studied its efficacy on preterm children who eventually receive a diagnosis of ASD. On the other hand, there is currently no evidence to show that this group is comparable with full-term infants with ASD, or even that the impairments of preterm infants who show characteristics of ASD can be reduced through an early intervention program.

This paper reports the results of an early-intervention pilot study aimed at ascertaining whether a focused social-communication intervention can yield broader gains in social, cognitive, language, and adaptive functioning in young preterm children with autism and its feasibility and acceptability. More specifically, our study sought to: (a) examine the effect of the social-communication intervention program on young preterm and full-term infants with ASD; (b) explore whether there were differences in intervention gains as between preterm infants with ASD, full-term children with ASD (comparison group), and another preterm infant group without increased ASD (control group); (c) investigate the individual effect of the intervention; and lastly (d) assess feasibility and acceptability of the SCI in preterm and full-term children with ASD. Moreover, this is the first study to use individual change indices in a pre-post design with preterm infants with ASD, a comparison group, and a control group.

Methods

Ethical Approval

Ethical approval was given by the Research Ethics Committee of the University of Salamanca (201700031949) and the Salamanca Clinical Hospital, Spain (PI9910/2017). Prior to starting the program, all participant families were required to read the information about the study and give their informed consent.

Design

To determine the effects of the intervention on preterm children with ASD, a quasi-experimental design was used to compare the following three groups: (1) an experimental group of preterm children with ASD and receiving an SCI; (2) a comparison group of children with ASD and receiving an SCI; and lastly, (3) a control group of preterm children. To examine individual child growth and variables affecting these outcomes, data from all subjects in each group were analyzed jointly as well as individually. Figure 1 shows the participant flow across the study.

Participants

Children were recruited from a developmental disorder diagnostic and rehabilitation centre and from the Salamanca Clinical Hospital through a developmental surveillance

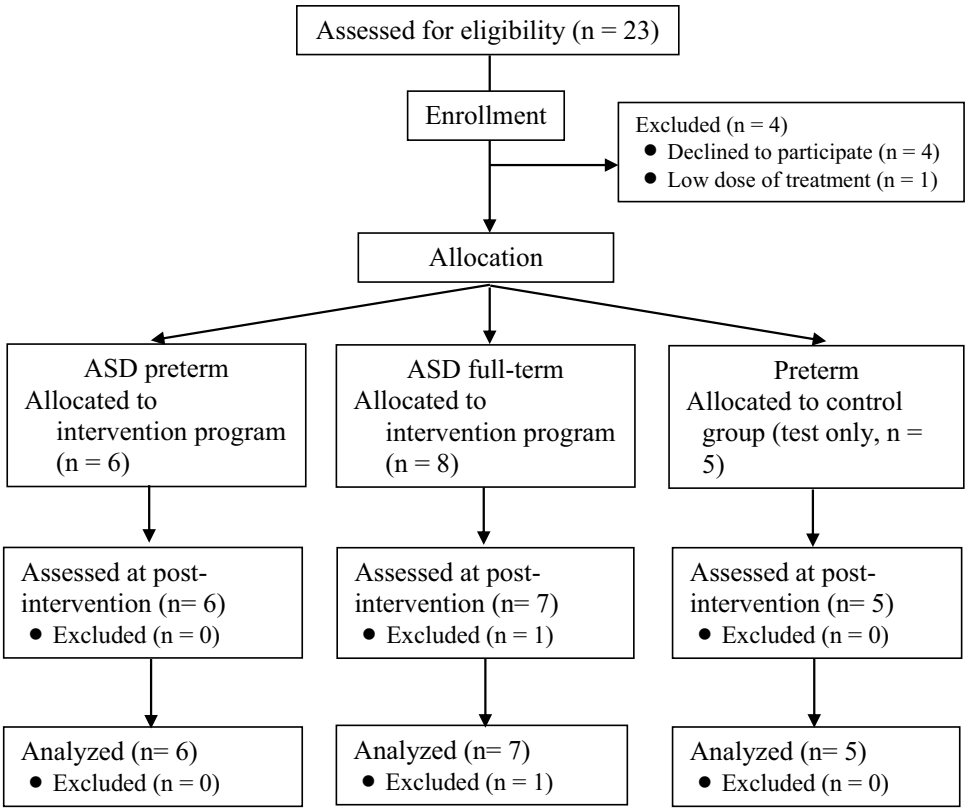

Fig. 1 Recruitment flow diagram

program (visits at 6, 12, 18 and 24 months) that was ongoing at the time of the study, and they were allocated to the three groups in accordance with the respective inclusion criteria, i.e., (1) Preterm children with ASD: (a) < 37 weeks of gestation at birth, (b) chronological age 18–20 months (corrected by subtracting gestational age at birth from 40), and (c) score above the designated cut-off level on the Autism Diagnostic Observation Scale – 2nd edition (Toddler module, Lord et al., 2012) and on the Modified Checklist for Autism in Toddlers – revised/follow-up (M-CHAT-R/F, Robins et al., 2014), deemed to show concern of ASD (ADOS ≥ 9; M-CHAT-R/F ≥ 3). (2) Full-term children with ASD: (a) chronological age 18–20 months, and (b) score above the designated cut-off level on the ADOS2 – T module and on the M-CHAT-R/F, deemed to show concern of ASD. (3) Preterm children: (a) < 37 weeks of gestation at birth, (b) chronological age 18–20 months (corrected by subtracting gestational age at birth from 40), and (c) score under the designated cut-off level on the ADOS2 – T module and on the M-CHAT-R/F, *not* deemed to show concern of ASD (ADOS < 9; M-CHAT-R/F < 3). The exclusion criteria were defined as moderate-severe hearing/visual deficits or motor disorders (e.g., cerebral palsy, epilepsy, spinal cord), or other co-morbid medical conditions. ASD diagnosis was confirmed at 24 months.

Recruitment took place over 24 months (December 2017 through November 2019). Due to this rolling enrollment process, children continued to enter the intervention throughout the treatment period. Control group (preterm children) were selected randomly from the Salamanca Clinical Hospital database. Four families declined to participate due to busy family schedules and associated medical conditions.

The final sample comprised 18 children assigned to the three groups. ASD characteristics was assessed with the ADOS2-T module by a multi-disciplinary team on the basis of a comprehensive clinical evaluation and the M-CHAT-R/F. There were no significant differences between preterm and full-term children with ASD. However, there were significant differences between the experimental and control groups in the ADOS-T module ($d = 73$), Cognitive ($d = 81$) and Language ($d = 85$) composite scores, and Communication ($d = 86$), Socialization ($d = 88$), Object Imitation ($d = 71$), Joint attention (Response; $d = 66$), and Play (Acts, $d = 67$; Frequency, $d = 70$) measures. The members of the control group did not have characteristics of ASD, and all their pre-treatment scores were higher. Table 1 below summarizes the descriptive sample information.

Measures

Coding and Reliability

For each coding system, 100% of the videos were double-coded to establish reliability statistics (intraclass correlation

Table 1 Child demographics

	Preterm ASD (n = 6)	Full-term ASD (n = 7)	Preterm (n = 5)	Preterm ASD vs. Full-term ASD	Preterm ASD vs. Preterm
	M (SD)/frequency (%)	M (SD)/frequency (%)	M (SD)/frequency (%)	$\chi 2$/F	$\chi 2$/F
Gender					
Male	5 (83%)	7 (100%)	4 (80%)	$\chi 2 = 0.48$	
Female	1 (17%)	0 (0%)	1 (20%)		
Chronological age[+]	19.17 (0.41)	20.00 (1.15)	18.60 (0.89)	0.279	0.219
Gestation at birth (weeks)	31.67 (4.88)	–	30.80 (2.77)	–	0.597
ADOS2-T	16.33 (3.45)	15.86 (3.49)	0.86 (0.71)	0.50	12.16*
Cognitive[a]	70.50 (9.53)	71.71 (7.61)	93.00 (5.70)	− 2.05	− 23.58*
Language[a]	63.00 (8.50)	59.71 (6.85)	88.00 (5.29)	0.50	− 28.17*
Communication[b]	70.17 (8.23)	66.86 (5.79)	94.60 (2.70)	2.75	− 17.58**
Socialization[b]	79.17 (7.58)	77.14 (5.04)	91.20 (6.46)	0.75	− 8.23
Object imitation	5.83 (4.17)	5.71 (3.55)	14.60 (1.14)	− 1.30	− 8.87*
JA initiation	0.33 (0.52)	1.29 (1.50)	4.80 (0.84)	− 0.75	− 4.41*
JA response	4.83 (1.94)	3.57 (2.64)	6.60 (0.89)	1.20	− 1.33
Play					
Acts	4.67 (2.58)	4.93 (2.45)	9.25 (2.02)	− 1.20	− 5.53*
Frequency	12.57 (1.75)	11.95 (2.96)	18.64 (1.53)	− 0.54	− 7.12*
Parents' highest level of education					
Secondary school or less	1 (17%)	1 (14%)	1 (20%)	$\chi 2 = 0.98$	
High school	1 (17%)	2 (29%)	1 (20%)		
Graduate	4 (66%)	4 (57%)	3 (60%)		
Treatment received (hour)	29.10 (1.53)	29.23 (1.76)	–	− 0.62	–
Treatment as usual received (hour)	8.00 (4.90)	9.14 (3.02)	8.60 (3.29)	− 1.17	− 1.02

JA Joint attention; $\chi 2$ Chi-square

[+]Corrected for preterm children (subtracting gestational age at birth from 40)

*p < 0.05, **p < 0.001

[a]Composite Score in Bayley-III

[b]Standard Score in Vineland-II

coefficients (ICCs) and Kappa coefficients), which are reported for all coding systems except parents' measures. The mean between pre- and post-treatment measures was calculated, as were the ICCs using the formula of a two-way mixed effects model, with absolute agreement and single rater/measurement (Koo & Li, 2016). Assessments were videotaped and all tests were scored from the video, except for the M-CHAT/R and Vineland-III due to the fact that these tools are based on questions that parents must answer. Research assistants were purpose-trained to code the videotaped measures reliably. Videos were distributed to independent coders. Research assistants were blind to the study hypothesis, treatment condition, and time points.

Feasibility and Acceptability

A mixed methods approach was adopted to establish feasibility and acceptability. As such, we collected data on feasibility and acceptability as outlined below.

Feasibility was assessed by collecting data on recruitment and retention rates, adherence rates, time required to recruit, and attrition. Lastly, feasibility of testing procedures and data collection methods, including completion rates.

Acceptability of the intervention and study procedures was assessed both quantitatively and qualitatively. Quantitative data was collected to investigate the number of sessions attended and length of intervention. A survey was designed to collect parents' views and experiences of the intervention, including what they perceive to be barriers and facilitators. Families were asked to report the degree of adequacy of the service on a scale of 1 to 6 (1: extremely inadequate; 2: moderately inadequate; 3: slightly inadequate;

4: neither adequate nor inadequate; 5: slightly adequate; 6: moderately adequate; 7: extremely adequate). The different answer choices for these questions were then collapsed and recoded into three categories, namely, Inadequate (from 1 to 3), Neutral (4) and Adequate (from 5 to 7), for the purpose of conducting a comparative analysis. Families were asked to rate the agreement in the following variables: information and support received, the evaluation process and recruitment, participation in sessions, the number of sessions, the length of intervention, the setting of the intervention, the delay between recruitment and start of intervention, staff qualifications, and the level in which professionals took into account family's concerns (see Table 2 in the Results section).

Primary Outcome Measures

The effect of the intervention on imitation, joint attention and play skills was examined using three assessments scored by trained research assistants blind to the participants' group assignment.

The Unstructured Imitation Assessment tool (UIA; Ingersoll, 2010) was used to measure children's ability to imitate in a spontaneous, social-interactive context. The UIA consists of 10 object- and 10 gesture-imitation tasks, but the gesture-imitation task was eliminated due to the age of the participants. The examiner engaged the child in free play with a set of toys, and then alternated between imitating of the child's nonverbal behavior and modeling actions for the child to imitate (e.g., roll a ball on a Table 3 times saying 'roll, roll, roll'). The child's response was scored '0' for no response or an incorrect response, '1' for an emerging response, and '2' for a full imitative response. Scores could range from 0–40, with a Kappa value of 0.92.

The Joint Attention Protocol (JAP, Nowell et al., 2018) was used to measure a child's response to initiation bids and his/her own initiation bids during a semi-structured interaction with the examiner. The protocol alternates between eight initiation trials and eight response trials, providing a total of 16 opportunities to observe joint attention. Response to JA tasks increases in level of support and prompting gradually across the assessment. All items were scored dichotomously as follows: '1' for pass or '0' for fail. Kappa scores were 0.95 for JA Response and 0.94 for JA Initiation measures.

The Structured Play Assessment instrument (SPA; Ungerer & Sigman, 1981) was used to measure the frequency and acts of spontaneous play behaviors. The child is presented with five sets of toys at a table. The examiner may respond to the child's communication but may not direct the child's play or show the child how to play with the toys. The play interaction lasted approximately 15–20 min. The variables used in the analysis were Simple object (e.g., separate objects, mouthing objects), Combination (e.g., nesting cups), Presymbolic (e.g., object used in a way that indicates a pretend quality), and Symbolic play (e.g., object used to represent another, doll used as a play agent). Simple object and Combination play, and Presymbolic and Symbolic play were collapsed into two categories (Manipulative – Pre/Symbolic), by summing the scores. The ICCs were as follows: for (1) Play Acts, they were 0.93 for Simple object, 0.91 for Combination, 0.89 for Functional, and 0.90 for Symbolic play; and for (2) Play frequency, they were 0.87 for Simple object, 0.86 for Combination, 0.88 for Functional, and 0.85 for Symbolic play.

Secondary Outcome Measures

The Ados2 T-Module and M-CHAT-R/F were used as screener tests to measure the risk for ASD. The Toddler Module algorithm contains separate domain categories of Social Affect (SA) and Restricted, Repetitive Behaviors (RRB) and a single total score to determine classification.

Table 2 Percentage of satisfaction among parents according to the intervention group

	Preterm children with ASD parents (n = 6)			Full-term children with ASD parents (n = 7)		
	Inadequate	Neutral	Adequate	Inadequate	Neutral	Adequate
Information and support received	0	0	100	0	14.3	85.7
The evaluation process and recruitment	0	0	100	0	0	100
Participation in sessions	0	0	100	0	0	100
Number of sessions	16.7	0	83.3	0	14.3	85.7
Length of intervention	0	0	100	0	0	100
Setting of intervention	0	16.7	83.3	0	0	100
Delay between recruitment and start of intervention	0	0	100	0	0	100
Staff qualifications	0	0	100	0	0	100
Professionals took into account family's concerns	0	0	100	14.3	0	85.7

Table 3 Means and standard deviations of pre- and post-intervention measures for all three groups of children

Test	Preterm with ASD						Full-term with ASD						Preterm					
	Pre-test Mean (SD)	Post-test Mean (SD)	t	CI (95%) Low	CI (95%) Up	Cohen's *d*	Pre-test Mean (SD)	Post-test Mean (SD)	t	CI (95%) Low	CI (95%) Up	Cohen's *d*	Pre-test Mean (SD)	Post-test Mean (SD)	t	CI (95%) Low	CI (95%) Up	Cohen's *d*
Object imitation	5.83 (4.17)	13.83 (3.49)	− 4.30*	− 16.09	− 2.40	0.95	5.71 (3.55)	13.57 (3.60)	− 7.72*	− 10.87	− 5.12	0.88	14.60 (1.14)	16.00 (0.71)	− 2.98	− 3.26	− 0.59	0.60
JA initiation	0.33 (0.52)	3.33 (0.82)	− 6.69*	− 4.79	− 1.70	0.86	1.29 (1.50)	2.86 (1.77)	− 2.32	− 4.39	0.39	0.54	4.80 (0.84)	5.80 (0.45)	− 2.98	− 3.25	0.59	0.60
JA response	4.83 (1.94)	6.83 (1.33)	− 2.65*	− 4.40	0.40	0.74	3.57 (2.64)	5.14 (2.54)	− 1.44	− 5.24	1.64	0.33	6.60 (0.89)	7.60 (0.55)	− 1.95	− 4.27	1.60	0.69
Play acts manipulative	3.00 (0.92)	2.92 (0.77)	0.51	− 2.59	3.59	0.25	3.00 (1.53)	1.43 (0.78)	1.83	− 0.72	3.52	0.52	2.00 (0.71)	1.73 (0.65)	1.49	− 1.26	2.59	0.58
Play Freq. manipulative	21.33 (3.83)	11.33 (3.27)	− 0.05	− 16.16	15.66	0.02	15.86 (7.15)	5.86 (2.80)	2.20	− 3.22	28.02	0.72	11.20 (0.84)	6.20 (1.30)	1.05	− 5.18	8.51	0.32
Play acts Pre/Symbolic	1.67 (1.63)	4.67 (2.34)	− 7.35*	− 4.30	− 1.70	0.70	3.14 (1.95)	6.14 (2.91)	− 4.49*	− 3.88	− 0.91	0.68	5.40 (1.57)	6.40 (1.14)	− 1.97	− 3.27	− 0.77	0.46
Play Freq. Pre/Symbolic	3.83 (3.71)	14.00 (6.66)	− 4.94*	− 17.68	− 3.82	0.84	8.71 (6.10)	18.43 (7.12)	− 6.39*	− 12.91	− 5.08	0.96	14.40 (2.07)	18.00 (1.87)	− 2.25	− 9.72	3.05	0.52
Cognitive[a]	70.50 (9.53)	85.83 (8.01)	− 2.58	− 31.22	3.22	0.64	71.71 (7.61)	83.57 (8.06)	− 2.47	− 27.98	1.58	0.64	93.00 (5.70)	96.00 (5.48)	− 0.74	− 11.28	7.95	0.31
Language[a]	63.00 (8.50)	71.50 (9.94)	− 6.55*	− 15.60	− 5.39	0.44	59.71 (6.85)	70.29 (9.53)	− 7.03*	− 13.11	− 5.69	0.60	88.00 (5.29)	90.00 (3.54)	− 2.13	− 7.04	2.38	0.41
Communication[b]	70.17 (2.71)	78.17 (2.64)	− 22.50**	− 8.56	− 6.43	0.96	66.86 (5.79)	75.00 (5.41)	− 28.69**	− 9.21	− 7.58	0.97	94.60 (2.70)	96.20 (3.12)	− 1.73	− 3.48	1.48	0.26
Socialization[b]	79.17 (3.82)	82.17 (5.15)	− 2.61	− 7.76	0.76	0.65	77.14 (5.04)	81.86 (6.04)	− 3.26*	− 7.40	− 0.60	0.48	93.00 (3.16)	91.20 (6.46)	− 0.19	− 7.61	6.95	0.08

JA Joint attention

*$p < 0.05$, **$p < 0.001$

[a]Composite Score in Bayley-III

[b]Standard Score in Vineland-III

Clinical cut-off scores are grouped within levels of concern for ASD (risk for ASD SA + RBB > 9). In the M-CHAT-R/F parents answer 20 yes/no questions; if children screen positive, parents are asked structured follow-up questions to obtain additional information and examples of at-risk behaviours (risk for ASD a score ≥ 3).

The Bayley Scales of Development – 3rd edition (Bayley, 2006) was used to measure cognitive development and language. The ICCs were as follows: 0.94 for Cognitive scalar score; 0.92 for Age development; 0.97 for Language scalar score; 0.95 for Age receptive; and 0.97 for Age expressive.

The Vineland Adaptive Behaviour Scales, 3rd edition (Sparrow et al., 2016) was used to measure the Communication and Socialization domains. The scales are organized by reference to the three domains that correspond to the three broad domains of adaptive functioning specified in the DSM-5 (APA, 2013). The Parent/Caregiver form was used.

Assessment Procedure

Pre- and post-tests were administered at the children's treatment centre by one of two independent testers. The age of preterm children was corrected in pre- and post-treatment (by subtracting gestational age at birth from 40). The assessment started with the ADOS, followed by the Bayley-III; after a 30-min break, the UIA, JAP and SPA were then administered, in that order. Parents completed the M-CHAT-R/F and Vineland-III. Parents were also asked about the background characteristics of the child and family members, and specifically about the child's health history, as well as any additional interventions received before and during the study. Parents were allowed in the testing room during the assessment and the assessment took approximately 180 min. The time between pre- and post-assessment was four months. ASD diagnosis was confirmed at post-treatment time.

Intervention Procedure

Intervention Groups

All therapy was conducted by a graduate-level research assistant coached in the treatment model by trained interventionists (established fidelity of > 0.80 prior to commencement of and during treatment). Children in treatment groups (preterm and full term with ASD) received an SIC which targeted object imitation, joint attention and play one h per day, two days per week for 15 weeks (30 h). All sessions were conducted in a small treatment room, with a small table and chairs placed in the centre. Parents were allowed to be present during the intervention sessions. Fifteen sets of play materials were used in all treatment sessions. The program followed the principles of the treatment program developed by Ingersoll and Dvortcsak (2009, 2019): (Improving Parents as Communication teachers, Project ImPACT). Challenging behavior chapter was eliminated due to the focus of the study (social and communication skills). This program was chosen because is recognized as one of the most effective programs for children with ASD and related social communication delays. This intervention teaches strategies to help children develop social, communication, imitation, and play skills during daily routines and activities from 18 months of age. Therefore, the program is supported by research and based on developmental science and applied behavioral analysis (ABA) principles. It was recently recognized as a Manualized Intervention Meeting Criteria by the National Clearinghouse on Autism Evidence and Practice (NCAEP).

The SIC used naturalistic and behavioral techniques to teach social-communication skills during play and daily life activities (e.g., snack time). The therapist focused on children by following their lead and imitating them, adjusting the communication, creating opportunities, and teaching new skills. To follow the child's lead, the therapist stayed face-to-face with children, letting them choose and lead the play game or activity. Thereafter, the therapist joined in the child's play. To promote reciprocity, the therapist imitated the child's verbal and nonverbal behavior, adjusting the communication to the child's level. Both verbal and non-verbal communication were exaggerated, and a simple and repetitive language was used around the child's play. Communication opportunities were created, based on turns and toys placed in different containers or out of the child's reach.

Lastly, prompts and rewards to teach new skills were used. To teach object imitation the therapist modelled an action for the child to imitate. If the child did not learn the action within three models, the therapist used physical guidance. The actions were modelled with the duplicate of the toy with which the child was currently playing. The therapist taught responses to joint attention by pointing to different child's favourite toys. The initiations of joint attention were taught throughout exaggeration each time the child show or give something to the therapist. To expand play, the therapist used a verbal instruction or modelled an action for the child to imitate. If the child did not learn the action (imitation, joint attention, play) within three models or prompts, the therapist used physical guidance. Modelled and prompted actions were varied across toys to help children generalize behaviours avoiding associate a specific skill with a specific toy. Therapist rewarded children after attempts that met criteria at social-communication skills. The rewards were natural, and therapist gave them only for positive behaviours, and immediately.

Families were not barred from receiving other community services alongside the SCI, and documented other community services received by the two groups. Community treatment consisted of early stimulation sessions aimed at developing motor and cognitive skills.

Control Group

The preterm children participated in all assessment activities, but no SCI was administered during the study. Instead they received treatment-as-usual within their community settings. The control group did not present with ASD symptoms and thus received no SCI. There were no differences between the groups in terms of the number of hours of community services received (Table 1).

Data-Analysis

Statistical analyses were performed, using the IBM SPSS Statistics 26 and R Development Core Team software packages in the following ways: (1) analyses of intervention, determining the feasibility and acceptability; (2) analyses by group, determining the significance and effect of the intervention in each group, and comparing groups; (3) analyses by participant, determining the significance and effect of the intervention on each participant.

Firstly, comprehensive descriptive analyses of the two intervention groups (preterm and full-term with ASD parents) were performed related to feasibility and acceptability of treatment and identifying barriers and enabling factors to completion of the program. The categories of the assessment of the acceptability with intervention was a scale of 1 to 7 (1: extremely inadequate; 2: moderately inadequate; 3: slightly inadequate; 4: neither adequate nor inadequate; 5: slightly adequate; 6: moderately adequate; 7: extremely adequate). The different answer choices for all questions were then stratified and recoded into three new categories, namely, inadequate (from 1 to 3), neutral (4) and adequate (from 5 to 7).

Secondly, robust statistical analyses were conducted to control the range of probability distributions, especially in the case of small samples or distributions that were not normal (Daszykowski et al., 2007). To ascertain whether the groups differed in background variables, chi-square tests and robust analyses of variance (ANOVA) were performed. Robust paired-sample t-tests were used to determine the significance and effect of the intervention on pre- and post-treatment variables in each group. Additionally, robust ANOVA were performed to determine whether the groups differed in terms of Change Score (CS = post – pre score). Post-hoc analyses were conducted where significant differences were found between groups. Cohen's *d* was used to calculate the effect size (small < 0.20; medium ≥ 0.50; large ≥ 0.80).

Lastly, the Reliable Change Index (RCI, Jacobson et al., 1984) was calculated to determine whether the individual change in the different measures was reliable. Of the different RCI versions available, we used one in which the equality of pre- and post-test variances was not assumed (Christensen & Mendoza, 1986; Jacobson & Truax, 1991; Maasen, 2004). Reliability was estimated from the pre-post correlation (Estrada et al., 2019; Ferrer & Pardo, 2014) as follows:

$$RCI = \frac{Di}{\sqrt{(Spre\sqrt{1 - Rpre - post})^2 + (Spost\sqrt{1 - Rpre - post})^2}}$$

where Di = individual pre-post difference, $Spre$ = standard deviation pre-test, $Spost$ = standard deviation post-test, and $Rpre\text{-}post$ = pre-post correlation. This formula was computed for each participant in all measures, with individual changes being deemed reliable in any case where the RCI was higher than 1.96 ($p < 0.05$). The individual pre-post difference (Di) was calculated on the basis of the measure used to obtain a comparable RCI (≥ 0), namely: (1) 'Pre – Post' in measures aimed at reducing the pre-treatment score (e.g., Ados2—T module); and, (2) 'Post – Pre' in measures aimed at increasing the pre-treatment score (e.g., Object Imitation, Cognitive development).

Results

Feasibility and Acceptability

Twenty-seven children were assessed for eligibility. The time to recruit children was 180 min, where the complete assessment was administrated. The 14.8% of children were excluded before starting the program. In one case the reason for exclusion was that the child had associated health complications. In the remaining cases the reason for exclusion was that parents refused to participate due to busy schedules or medical associations. Fourteen children in both groups (preterm and full-term children with ASD) were allocated to intervention program. Only one child (7.1%) was excluded due to the low dose of treatment. The family refused to continue with the program when the child had received only eight hours because of the distance with the service and the lack of transport. All children completed pre- and post-treatment assessment (primary and secondary measures).

Children in the intervention groups completed almost all the sessions. Preterm children with ASD received 29.1 h (97.1% of the program), and full-term children with ASD received 29.2 h (97.3% of the program). Children in both groups attended at 98% of the programmed sessions. Parents reported high level of satisfaction with the intervention program. Between 83 and 100% reported that the program was adequate throughout all the variables (See Table 2). Parents of preterm children with ASD reported that medical appointment was a barrier to attend to some intervention sessions.

Intervention Effects

As shown in Table 3, the SCI had a statistically significant effect on most measures of the preterm and full-term ASD risk groups. The effect size was large ($d > 0.80$) in the same measurements for both groups. In pre-post JA Initiation and JA Response, preterm participants with ASD obtained a higher and significant effect. Significance was higher in both groups in pre-post Communication scores. No significant pre-post effect was observed in the preterm group.

Figure 2 shows the mean pre-post treatment scores for each group. While pre-treatment differences were higher between the intervention and control groups, these differences diminished post-treatment; and although the intervention groups obtained lower scores before starting treatment, the differences were significantly reduced after treatment. Control group scores remained stable over time.

Change Score between Groups

Change Scores were used to control for pre-treatment differences. Despite the fact that the pre-post treatment effect was lower in the intervention groups for most of the measures assessed, statistically significant inter-group differences were solely found in the Change Scores for the Object Imitation, Play Acts Pre/Symbolic, and Language measures (Table 4). The differences were greater in Object Imitation ($F = 27.60$, $p < 0.001$) and Communication ($F = 72.50$, $p < 0.001$). Preterm and full-term ASD risk groups obtained a significantly higher CS. As can be seen in Fig. 2, these differences were

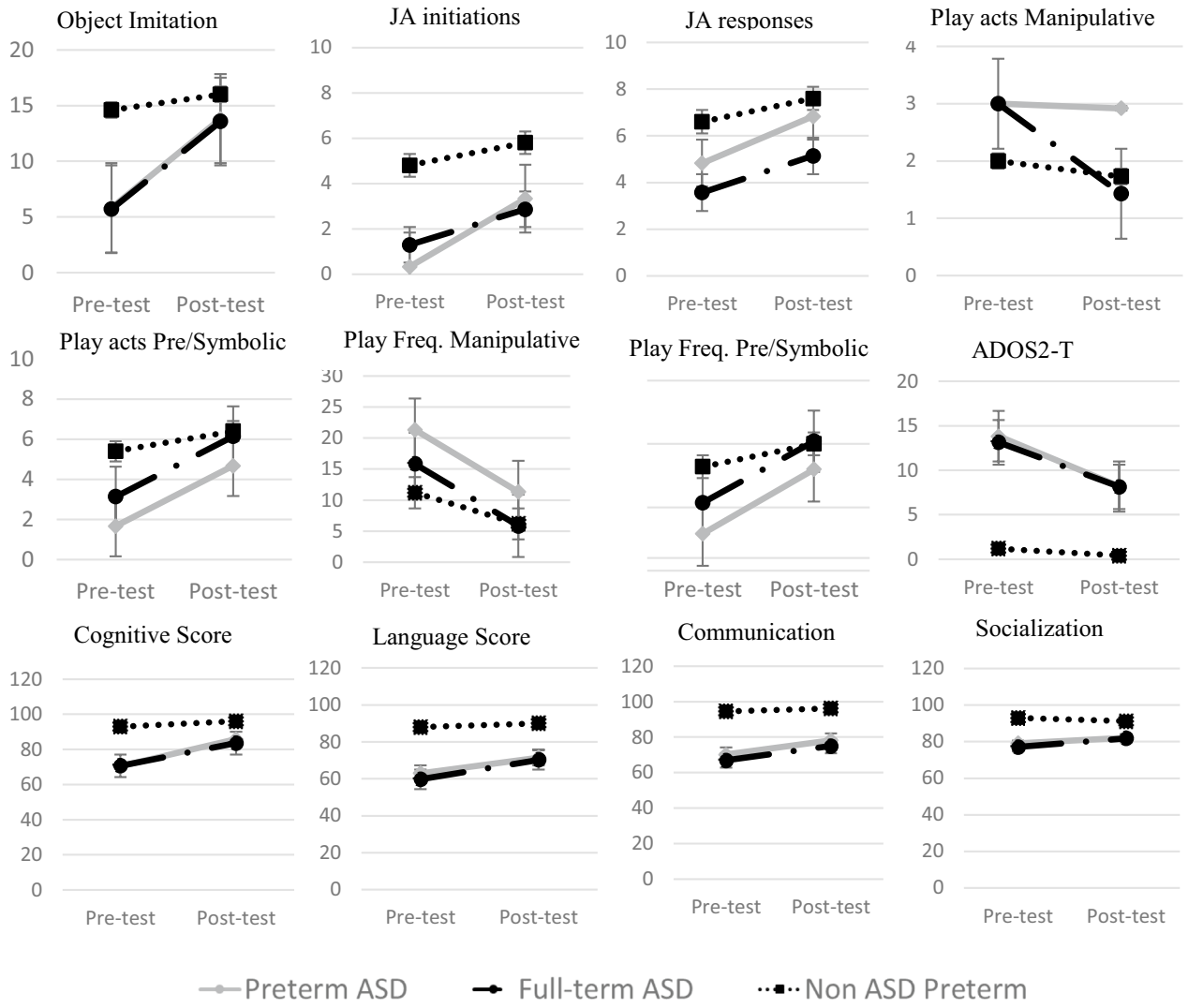

Fig. 2 Pre-post treatment means of each measure by group. Errors bars represent standard error

Table 4 Change score group differences

Test	Differences	Post-hoc analyses								
	F	Preterm with ASD vs. Full-term with ASD			Preterm with ASD vs. Preterm			Full-term with ASD vs. Preterm		
		psi-hat	CI (95%)		psi-hat	CI (95%)		psi-hat	CI (95%)	
			Low	Up		Low	Up		Low	Up
Object imitation	27.60**	− 0.30	− 6.75	6.15	5.83**	− 0.93	12.60	6.13**	3.51	8.75
JA initiation	4.17	–	–	–	–	–	–	–	–	–
JA response	0.89	–	–	–	–	–	–	–	–	–
Play acts manipulative	2.05	–	–	–	–	–	–	–	–	–
Play Freq. manipulative	3.08	–	–	–	–	–	–	–	–	–
Play acts Pre/Symbolic	11.3*	0.45	− 2.27	3.17	2.25*	0.83	3.67	1.80	− 0.91	4.51
Play Freq. Pre/Symbolic	3.21	–	–	–	–	–	–	–	–	–
Cognitive[a]	3.97	–	–	–	–	–	–	–	–	–
Language[a]	12.2*	− 1.10	− 6.27	4.07	5.50*	0.51	10.49	6.60*	2.04	11.15
Communication[b]	72.50**	− 0.40	− 2.69	1.89	6.33**	3.85	8.81	6.73**	4.66	8.81
Socialization[b]	2.69	–	–	–	–	–	–	–	–	–

JA Joint attention

*$p < 0.05$, **$p < 0.001$

[a]Composite Score in Bayley-III

[b]Standard Score in Vineland-III

larger pre-treatment and decreased post-treatment. No statistically significant differences were found between preterm and full-term ASD risk groups.

Reliable Change Index

The preterm and full-term ASD risk groups registered a significant, reliable mean change in the same measures (Table 5). In both groups, participants' mean RCI was significant (> 1.96) for the Object Imitation, Play Acts and Frequency Pre/Symbolic, Language, and Communication measures. Most participants obtained a significant RCI in these measures, with 4 to 6 (57% − 85%) participants in both groups achieving a reliable change (> 1.96). No significant RCIs were found in the preterm group. Individually, only one participant obtained a significant RCI in Play Frequency Pre/Symbolic (see Fig. 3 for fuller details of participants who obtained a significant RCI in each group).

Discussion

This pilot study examined the effect of an SCI on social and communicative functioning, cognitive development, and language in young preterm and full-term children with ASD. Children in the intervention groups made significantly greater pre-post-treatment gains in Object Imitation, Play, Language and Communication than did the control group. Furthermore, there were no differences between preterm and full-term children with ASD in any domain, with individual RCI analyses supporting the results obtained in group analyses. Although there was a significant change in specific social and communication skills, the results should be taken cautiously. The present study is a pilot study and was limited by its small sample size.

These findings are similar to those reported by other studies aimed at improving social and communication skills in young children with ASD (Landa, 2018; Rogers & Vismara, 2014). Although the developmental trajectory studied was short (from pre- to post-treatment, 5 months), the trajectory of preterm children with ASD was close to the trajectory of the control group, post-treatment. This finding may suggest that it was the early intervention rather than simple maturation which enhanced these skills (Marsden & Torgerson, 2012). Preterm children with ASD tend to show impairments in their social and communication, cognitive or language developmental trajectories, with these trajectories moving downwards over time and thereby increasing the difference with other preterm children (Chen et al., 2020; Nguyen et al., 2018). This is the first pilot study of an early intervention program in preterm children with ASD. Since this is a group with a high prevalence of ASD (7%; Agrawal et al., 2018), future research should focus on the study of the early intervention programs which produce better results.

Comparison of both groups with ASD (preterm and full-term) seemed to show no differences. Likewise, no

Table 5 Reliable change index with R correlation

	Preterm with ASD						*M*	Full-term with ASD							*M*	Preterm					*M*
Test	1	2	3	4	5	6		1	2	3	4	5	6	7		1	2	3	4	5	
Object imitation	2.03	4.74	2.37	1.35	3.72	2.03	2.71*	3.05	2.71	4.06	2.71	2.71	1.35	2.03	2.66*	1.02	1.35	1.35	0.68	1.35	1.15
JA initiation	1.90	1.90	1.27	2.54	2.54	1.27	1.90	0.00	− 0.63	3.17	0.00	1.27	1.90	1.27	0.99	0.63	1.27	0.00	0.63	0.63	0.63
JA response	1.93	2.57	0.00	0.64	1.29	1.29	1.29	3.86	0.00	1.29	0.64	0.00	0.64	0.64	1.01	0.00	1.29	1.29	0.00	0.64	0.64
Play acts manipulative	0.67	0.00	1.34	0.67	0.67	1.34	0.11	1.34	1.34	2.01	− 0.67	0.67	0.67	2.01	1.05	0.67	0.00	2.01	0.67	0.67	0.53
Play Freq. manipulative	− 1.16	0.25	− 0.25	0.31	0.12	0.31	− 0.07	− 0.49	2.33	1.23	0.12	0.74	0.80	1.04	0.82	0.37	0.00	− 0.06	0.06	0.12	0.07
Play acts Pre/Symbolic	3.52	3.52	2.64	2.64	2.64	0.88	2.64*	1.76	3.52	5.28	0.88	2.64	0.88	3.52	2.64*	0.88	0.88	0.00	1.76	0.88	0.88
Play Freq. Pre/Symbolic	4.29	4.29	1.65	3.30	2.97	1.98	3.08*	3.30	4.95	4.29	1.98	3.63	1.98	2.31	3.20*	1.98	0.66	0.66	1.98	0.66	1.18
Cognitive[a]	2.28	1.37	0.09	0.91	2.83	0.91	1.40	− 0.91	0.91	3.01	0.55	1.10	1.55	1.37	1.08	0.46	0.00	0.46	0.00	0.46	0.27
Language[a]	3.13	2.23	2.01	0.97	1.54	2.01	1.98*	1.79	1.79	4.25	1.56	2.68	1.79	2.68	2.36*	− 0.67	0.45	0.89	0.67	0.89	0.44
Communication[b]	5.55	4.31	4.93	4.93	5.55	4.31	4.93*	5.55	4.93	3.70	4.93	5.55	5.55	4.93	5.02*	1.85	1.23	0.00	0.62	1.23	0.98
Socialization[b]	0.76	0.00	0.57	0.38	1.51	0.19	0.57	1.51	0.57	1.51	0.94	0.57	0.94	0.19	0.89	− 1.04	0.19	0.38	0.38	0.57	0.09

If the value of the reliable change index (RCI) is greater than 1.96, the difference in scores between the two intervals is statistically significant at a 95% confidence interval

JA Joint attention

*RCI mean of participants statistically significant

[a]Bayley-III

[b]Vineland-III

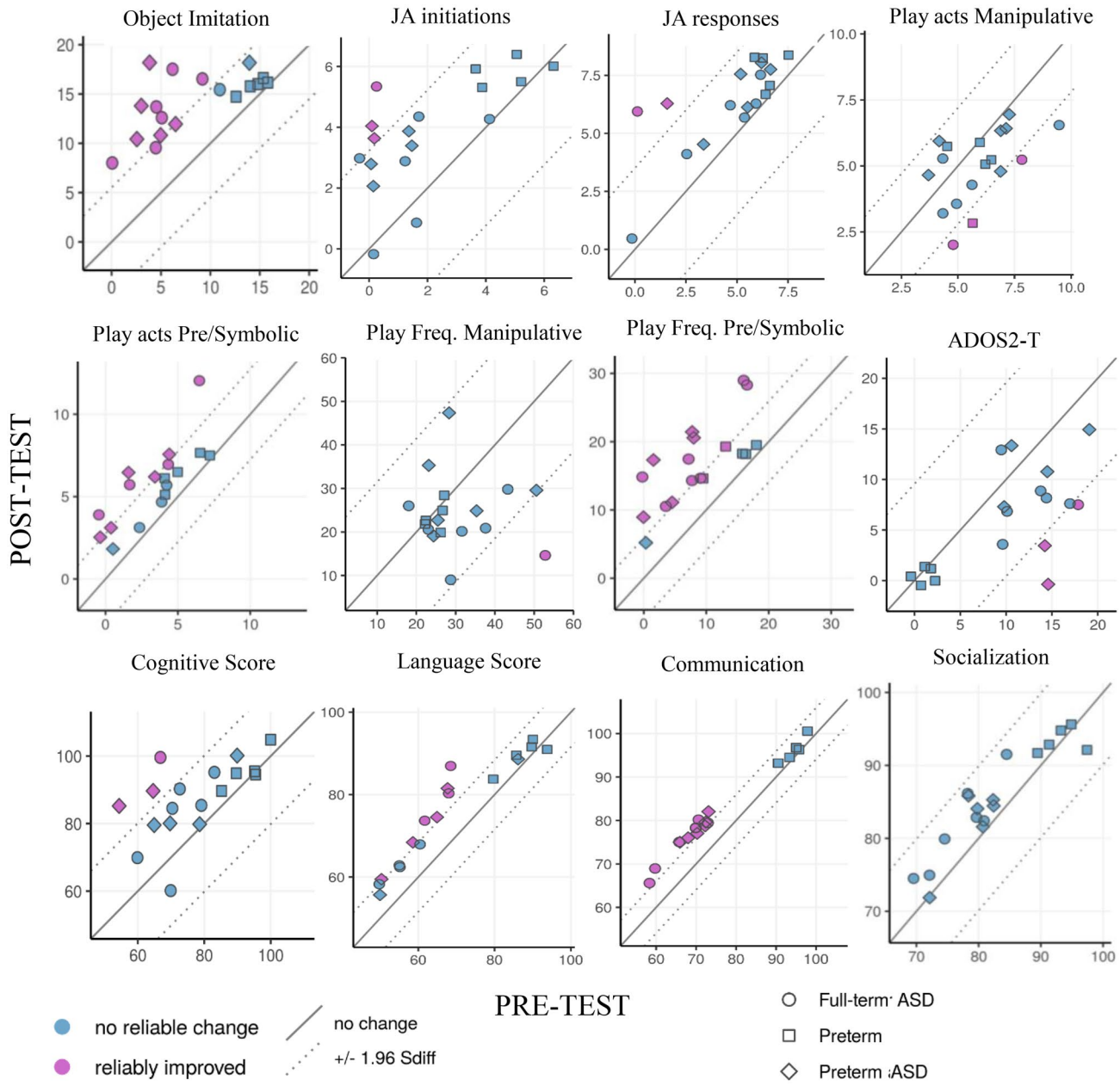

Fig. 3 Reliable Change Index by participant in each group. Participants out of the boundaries reliably improved

differences were found pre-treatment. This could be due to the fact that both groups display the same characteristics in terms of signs of ASD and its associated impairments. Even so, these results should be viewed cautiously, as this was a pilot study and there is little literature on the effect of early interventions on preterm and full-term children with ASD. When the three groups were compared, there were significant differences between the intervention groups and the control group in those instances where pre-treatment differences had been greater. Although the literature is limited, these results are comparable with other studies which compare difficulties in preterm children with ASD versus preterm children with no obvious ASD risk (Chen et al., 2020; Groote et al., 2006; Johnson et al., 2010). Future research should focus on intervention studies that compare preterm children with ASD with their non-risk peers and full-term children with ASD, in order to compare these with the results obtained by us and determine whether preterm children with ASD call for specific intervention procedures or actions.

Individual analyses showed that, with the exception of Joint Attention, the mean RCI for preterm children with ASD

was significant. These results could support group analysis of the effect size. The lack of reliable changes within the non-ASD preterm group suggests that this group's trajectory is stable over time. Although the RCI statistic is not commonly used in pre-post-treatment studies, it is nonetheless an extremely useful source of individual information. In cases where only pre- and post-treatment measures are available, a logical sequence of analytic steps would be (Estrada et al., 2019): (a) to assess individual changes via the RCI; and, (b) to perform a classical average-based effect size estimation, such as Cohen's *d*. This would not only help researchers ascertain whether an intervention has had a positive effect in each group, but would also enable decisions to be made about each particular case. Further research is needed to understand more about the respective intervention ingredients that would stimulate social development in preterm children with ASD who show limited social improvement.

Furthermore, the results of feasibility and acceptability study may provide valuable information useful in carrying out a full-scale trial early intervention with children with ASD. An economic evaluation should also be included to assess the cost effectiveness of the intervention. Parents of preterm children with ASD reported a barrier to assist to the program due to different medical appointments. To this, the intervention program was adapted to the agenda of each family. Services should consider families' schedules to increase the attendance ratios and reduce the attrition.

The results of this study are the first to show that preterm children with ASD acquire skills in areas considered to be core deficits of this disorder. The significance of these changes in specific skills is reinforced by some factors that were controlled for in the study. There is always the possibility that participants may engage in another intervention program during the course of a study. In our study, however, we controlled the intervention dosage. Although there was some variability in the number of treatment sessions which each child received, these differences were small and non-significant between groups. We also controlled how much and what type of treatment children received, by ensuring that all participants received 2 h per week of the same early intervention program. As reported by parents, no additional SCI programs were initiated while the children were under study.

Limitations and Future Research

Some limitations should be noted. First, the presence of pre-existing groups (preterm with ASD vs. full-term with ASD vs. preterm) meant that there were non-equivalent groups pre-treatment. However, these differences were offset by the Change Score analysis when it came to comparing groups. Using a repeated measures design, each subject was his/her own control when calculating gains, and the pre-test role acted as a covariate, thereby reducing the effect of pre-test differences. If groups are samples from two populations with different means, then regression of individual scores to the mean of their own population does not change the group means, and CS -rather than ANCOVA- may be unbiased (Van Breukelen, 2013). Hence, CS ANOVAs will outperform ANCOVAs in very small samples, and may produce far less biased effect estimates in a real-world situation (Kim, 2019). Randomized controlled trials (RCTs) are important in showing the effect of an intervention under ideal circumstances. However, it is important to show that, within a specific group (preterm children), a treatment is effective in a naturalistic setting where circumstances are not always ideal. RCTs also include a waitlist control group which provides stronger evidence. With a waitlist control group, the results can be compared directly with the experimental group. However, having a waitlist control group is not always the best option to start the program intervention as early as possible. Future research should study preterm children with ASD within the context of an RCT design.

Second, the present study is a pilot study, and it was based on a small sample size in each group. Despite the high prevalence of ASD in this group, approximately 143 preterm infants are born each year in the region where the study was conducted (data from Spain's National Institute of Statistics for the year 2018). With an ASD prevalence of 7% (Agrawal et al., 2018), there would be a maximum of 10 preterm infants with ASD per year. Due to the small sample of preterm children with ASD, it was not possible to have a control group with similar characteristics (with ASD). Though limited by the small sample size, findings from this study are nonetheless promising due to the positive outcomes found in early core characteristics of autism in preterm children with ASD. Robust statistics were therefore used, since these perform well for data drawn from a wide range of probability distributions, especially those that are not normal (Daszykowski et al., 2007). Also, there was not include a full-term control group in the study. However, the main goals were related to compare children attending the "ASD" and "Preterm" conditions. Data based a larger sample size and with a full-term control group will allow for a more in-depth analysis of factors affecting optimal treatment outcomes in this population.

Third, Children were recruited at 18 months old if they showed concern of ASD signs on the ADOS-2 and M-CHAT-R/F. After the intervention program, ASD diagnosis was confirmed at 24 months old. Although recent literature suggests that ASD diagnosis could become stable at this age, there is still controversy about the age at which a diagnosis of ASD is stable (from 24 to 36 months). Future studies should include a follow-up at 30–36 months to confirm the ASD diagnosis and support the results obtained in the field of ASD.

Finally, there were no follow-up data to assess the long-term impact of the treatment's effects. The limited follow-up allowed for only a brief examination of the intervention's sustainability. Future studies should therefore address these limitations in order to determine whether treatment effects vary at exit and follow-up. It is also critical to evaluate the efficacy of SCI against that of other treatments.

Conclusions

These results allow us to claim that change can be brought about in core developmental problems among preterm children with ASD, using a low-intensity intervention targeting social and communication skills. Even though such core areas of impairment are not easy to change, the intervention had an appreciable effect. Most of the participants improved significantly in socio-communicative skills, cognitive development, language, and adaptive behavior, and ASD signs were reduced. Our study adds to the literature which suggests that short-term focused interventions can reduce ASD impairments (Ingersoll, 2012). Accordingly, research into the longer-term effects of the intervention and a larger population of preterm children are needed, in order to examine possible mediators and moderators of intervention outcomes on children's social and communication abilities.

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Author Contributions ABM, RCB, and MMM designed the study; ABM and RCB wrote the manuscript; ABM, conducted the intervention program; ABM, MMM, AHF, SMD, PMG assessed families and children; ABM, MMM and EDV carried out statistical analyses and interpreted the results; MMM, AHF, EDV, CJR and MP collaborated in writing the manuscript. All authors have read and approved the final manuscript.

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Declarations

Conflict of interest All authors declare they have no conflicts of interest.

Ethical Approval Ethical approval was given by the Research Ethics Committee of the University of Salamanca, Spain (201700031949), and the Clinic Hospital of Salamanca, Spain (PI9910/2017).

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Discusión y conclusiones

DISCUSIÓN GENERAL

Esta tesis examinó el efecto de un programa de intervención temprana en el funcionamiento social y comunicativo, el desarrollo cognitivo y el lenguaje en menores pequeños prematuros y a término con signos de TEA, para lo cual se basó en información previa sobre los procedimientos focalizados que han aportado evidencia de eficacia. En el estudio sobre la intervención; y sobre las demandas y puntos de vista de familias y profesionales sobre los servicios de detección y atención temprana, los menores de los grupos de intervención obtuvieron ganancias significativamente mayores antes y después del tratamiento en la imitación de objetos, el juego, el lenguaje y la comunicación que el grupo de control. Además, no hubo diferencias entre los niños prematuros y los nacidos a término con TEA en ninguna de las áreas, y los análisis individuales respaldaron los resultados obtenidos en los análisis de grupo.

Como se ha indicado, el proceso de elaboración y puesta en marcha del programa de intervención comprendió dos estudios previos. En primer lugar, se realizó un metaanálisis de la literatura. Este estudio pretendió contribuir a la literatura existente que evalúa la eficacia de las prácticas focalizadas en cuanto a la mejora de las habilidades sociales y comunicativas específicas de los menores con TEA. Los resultados sugieren que los programas de intervención en estudios experimentales, con un diseño de grupo o de caso único, que se centran en la mejora de las habilidades socio-comunicativas, producen efectos positivos. En concreto, estos efectos se encuentran en las prácticas en las que los padres y/o profesores participan activamente junto al terapeuta principal. Además, el efecto de la intervención aumenta entre los participantes que empiezan a participar en los programas a una edad temprana. Estas prácticas demostraron un efecto positivo en el desarrollo de las habilidades comunicativas. Por tanto, el uso de ellas permitiría desarrollar un programa para el desarrollo de habilidades de los menores con

TEA, ya que producen efectos positivos, reducen los costes y los tiempos de espera, dos aspectos fundamentales en la satisfacción de los padres y profesionales de los menores con TEA (Bejarano-Martín et al., 2020b). Los resultados obtenidos del tamaño del efecto global de las prácticas focalizadas son similares a los de otros programas de intervención estudiados ampliamente en la literatura (por ejemplo, el Análisis de Conducta Aplicado (ABA), el Modelo Denver, el PECS).

El tamaño del efecto encontrado en el metaanálisis global de las prácticas focalizadas fue comparable al de otros metaanálisis de intervenciones sociales y comunicativas realizados hasta la fecha (g = 0,51, Gates et al., 2017; g = 0,47, Reichow et al., 2012). La asignación aleatoria de grupos fue posible en la mayoría de los estudios de diseño grupal. Por lo tanto, todos los estudios de diseño de grupo incluidos en los análisis cumplieron los estándares de calidad metodológica. La metarregresión de los análisis de moderadores de los estudios de diseño de grupos aportó pruebas de la importancia de considerar las características de los participantes para mejorar el efecto de los programas aplicados. Estos resultados apoyan las sugerencias de otros estudios (véase Siu et al., 2016) de que las intervenciones deben aplicarse lo antes posible. Sin embargo, estos datos muestran que, aunque el aumento de la dosis de tratamiento tuvo un impacto positivo en el efecto de la intervención, los resultados no fueron estadísticamente significativos. Estos resultados son coherentes con Virues et al. (2010).

Los tamaños del efecto fueron significativos cuando, además del terapeuta, participaron en la intervención los padres, los cuidadores o los profesores. Estos resultados están en consonancia con los resultados de otros estudios en los que se evaluó el papel de los padres o cuidadores en la intervención (Estes et al., 2015, Pickles et al., 2016), y que también aportan pruebas de una reducción de los niveles de estrés familiar (Keen et al., 2010). La participación de los padres o cuidadores ha sido propuesta como

uno de los componentes clave de la intervención por numerosos estudios (Zwaigenbaum et al., 2015; Casagrande & Ingersoll, 2017), ya que son las personas con las que los niños con TEA pasan la mayor parte de su tiempo. Tener en cuenta las prioridades y preferencias de los cuidadores produce resultados positivos (Leadbitter et al., 2017), ya que tienen una mejor comprensión de los retos a los que se enfrenta el niño en su vida diaria. Además, la inclusión de los cuidadores reduciría muy probablemente los costes de los servicios públicos, al disminuir el número de horas de intervención que los terapeutas dedican a cada niño. En consecuencia, dar a los cuidadores un papel activo no sólo ayudaría al niño con TEA, sino que también conduciría a una reducción del estrés parental a largo plazo y de las demás cargas relacionadas con el TEA (Keen et al., 2010).

Por tanto, los resultados de este estudio sugirieron el uso de las prácticas de intervención focalizada en habilidades como gestos, atención conjunta, juego, imitación y contacto visual. Además, el iniciar la intervención de forma temprana, y la participación activa de los padres, parece que tienen un papel importante en el incremento del efecto y, por tanto, la eficacia del programa.

En segundo lugar, se estudiaron las perspectivas de los padres y profesionales de menores con TEA. El objetivo de este estudio era analizar las características de los servicios de detección, diagnóstico e intervención que reciben los menores con TEA, y comparar y contrastar la satisfacción general reportada por 1.223 familias y 760 profesionales, con el fin de proporcionar un marco basado en la evidencia para la creación y puesta en marcha de un programa de atención temprana, y así poder incorporar las opiniones de estos grupos en estas actividades. En lugar de pretender ser representativo de toda la Unión Europea, este estudio trató de obtener una muestra representativa de la mayoría de los países que participaron en el proyecto ASDEU (2015-2018), a fin de realizar un análisis global que fuera útil para la gestión de nuevas hipótesis y cambios.

Aunque la satisfacción general con los servicios de padres y profesionales de menores con fue positiva, los profesionales se mostraron más satisfechos que los familiares. Estas diferencias podrían deberse al hecho de que las familias tienen que lidiar con el proceso no solo de obtener el reconocimiento y la aceptación del hecho de que hay algo que no funciona en el desarrollo de su hijo/a, sino también con la espera de los servicios, así como con la gran cantidad de servicios y visitas médicas que necesitan los menores con TEA, todo lo cual da lugar a mayores niveles de estrés (Burke y Goldman 2015; Summers et al., 2007). Por ejemplo, se encontraron diferencias significativas en la satisfacción con el número de sesiones de intervención recibidas. En base a su experiencia, los padres reportaron recibir menos de la mitad de tiempo en las sesiones de intervención que las reportadas por los profesionales, lo que mostraría cómo las experiencias personales vividas en los servicios podrían afectar a la satisfacción.

Las familias informaron de que la demora media entre la detección y el diagnóstico (18,1 meses) era mucho mayor que entre el diagnóstico y el tratamiento (5,8 meses), y el 14,8% de las familias informaron de que su hijo había iniciado un programa de intervención (privado o público) antes de recibir un diagnóstico formal. Aun así este dato informa que de media pasan unos 20-23 meses desde las primeras preocupaciones hasta recibir un programa de intervención. El hecho de que las familias informaran de respuestas más tardías y lentas que los profesionales sugeriría que existen desfases en los servicios y que es necesario dotar al personal profesional de recursos técnicos y humanos (programas de formación y herramientas) que agilicen los procesos de detección y diagnóstico y reduzcan los retrasos en el acceso a dichos servicios. Las familias que tuvieron un acceso temprano a los servicios y experimentaron menos retrasos tendieron a valorar más positivamente los servicios. La mayoría de las familias que participaron en el estudio informaron de que, tras preocuparse por su hijo y comunicar sus inquietudes a

un pediatra, tuvieron que esperar, primero, a que un servicio especializado les diagnosticara el TEA y, después, a que se les aplicara un programa de intervención. El programa de intervención objetivo de la presente tesis se inició a los 18 meses, unas semanas después de que se detectaran las primeras dificultades, y sin un diagnóstico formal específico, lo que redujo en gran medida los tiempos de espera, hasta prácticamente su desaparición.

Este estudio muestra que la implicación activa de los padres aumenta la satisfacción de las familias con los servicios, un hallazgo coherente con otros estudios que muestran que la implicación de los padres es fundamental para la satisfacción con los programas de intervención (McIntyre y Zemantic, 2017; Stadnick, Drahota y Brookman-Frazee, 2013). En los últimos años, también se ha demostrado que la implicación activa, no solo aumenta la satisfacción con el servicio, sino que también mejora los resultados de la intervención, por ejemplo, aumentando el progreso en la adquisición de habilidades (Ingersoll & Wainer, 2013; Kasari, Gulsrud, Paparella, Hellemann, & Berry, 2015; Pickles et al., 2016). Todos estos factores hacen que la participación de los padres en las intervenciones reduzca la carga económica para la familia, el sistema sanitario y la sociedad, junto con el estrés asociado a tener un hijo con TEA (Kasari et al., 2015). Estos resultados sirvieron para reforzar la idea de que el programa de intervención debía tener en cuenta en todo momento las perspectivas de las familias, así como su inclusión en el programa de forma activa.

Una vez analizados ambos estudios y teniendo en cuenta los resultados mencionados, se puso en marcha el programa de intervención temprana. Los resultados obtenidos fueron similares a los reportados por otros estudios dirigidos a mejorar las habilidades sociales y de comunicación en menores con TEA (Landa, 2018; Rogers y Vismara, 2014). Aunque la trayectoria de desarrollo estudiada fue corta (de pre a post

tratamiento, 5 meses), la trayectoria de los menores prematuros con TEA fue cercana a la trayectoria del grupo de control, post tratamiento. Este hallazgo puede sugerir que fue la intervención temprana y no la simple maduración lo que mejoró estas habilidades (Marsden y Torgerson, 2012). Los menores prematuros con TEA tienden a mostrar deficiencias en sus trayectorias de desarrollo social y comunicativo, cognitivo o del lenguaje, siendo estas trayectorias descendentes con el tiempo y aumentando así la diferencia con otros menores con nacimiento prematuros (Chen et al., 2020; Nguyen et al., 2018).

La comparación de ambos grupos con TEA (prematuros y a término) no pareció mostrar diferencias. Del mismo modo, no se encontraron diferencias antes del tratamiento. Esto podría deberse a que ambos grupos presentan las mismas características en cuanto a los signos de TEA y sus deficiencias asociadas. Cuando se compararon los tres grupos, hubo diferencias significativas entre los grupos de intervención y el grupo de control en aquellos casos en los que las diferencias pretratamiento habían sido mayores. Los análisis individuales mostraron que, a excepción de la atención Conjunta, la media de los menores prematuros con TEA fue significativa. Estos resultados podrían apoyar el análisis de grupo del tamaño del efecto. La falta de cambios fiables dentro del grupo de prematuros sin TEA sugiere que la trayectoria de este grupo fue estable a lo largo del tiempo.

Este es el primer estudio piloto de un programa de intervención temprana en menores prematuros con TEA. Dado que se trata de un grupo con una alta prevalencia de TEA (7%; Agrawal et al., 2018), la investigación futura debería centrarse en el estudio de los programas de intervención temprana que producen mejores resultados.

LIMITACIONES E INVESTIGACIÓN FUTURA

Hay que señalar algunas limitaciones que nos permiten vislumbrar las posibles líneas de investigación futura relativa a la atención temprana. En primer lugar, la presencia de grupos preexistentes (pretérmino con TEA frente a pretérmino a término con TEA frente a pretérmino) significó que había grupos no equivalentes antes del tratamiento. Sin embargo, estas diferencias se compensaron con los análisis estadísticos. Utilizando un diseño de medidas repetidas, cada sujeto era su propio control a la hora de calcular las ganancias, y el papel del pre-test actuaba como covariable, reduciendo así el efecto de las diferencias. Los ensayos controlados aleatorios (ECA) son importantes para mostrar el efecto de una intervención en circunstancias ideales. Sin embargo, es importante mostrar que, dentro de un grupo específico (niños prematuros), un tratamiento es eficaz en un entorno naturalista en el que las circunstancias no siempre son ideales. Los ensayos aleatorios también incluyen un grupo de control en lista de espera que proporciona pruebas más sólidas. Con un grupo de control en lista de espera, los resultados pueden compararse directamente con el grupo experimental. Sin embargo, tener un grupo de control en lista de espera no siempre es la mejor opción para iniciar la intervención del programa lo antes posible. Las investigaciones futuras deberían estudiar a los niños prematuros con TEA en el contexto de un diseño de ensayos aleatorios.

En segundo lugar, el programa de intervención fue un estudio piloto que fue elaborado en base a dos estudios previos, y se basó en un tamaño de muestra pequeño en cada grupo. A pesar de la alta prevalencia de TEA en el grupo de prematuros, aproximadamente 143 menores nacen con prematuridad cada año en la región donde se realizó el estudio (datos del Instituto Nacional de Estadística de España para el año 2018). Con una prevalencia de TEA del 7% (Agrawal et al., 2018), habría un máximo de 10 bebés prematuros con TEA al año. Debido a la pequeña muestra de niños prematuros con

TEA, no fue posible tener un grupo de control con características similares (con TEA). Aunque limitados por el pequeño tamaño de la muestra, los hallazgos de este estudio son, no obstante, prometedores debido a los resultados positivos encontrados en las características básicas tempranas del autismo en los niños prematuros con TEA. Por lo tanto, se utilizaron estadísticas robustas, ya que éstas funcionan bien para los datos extraídos de una amplia gama de distribuciones de probabilidad, especialmente las que no son normales (Daszykowski et al., 2007). Además, no se incluyó en el estudio un grupo de control a término. Sin embargo, los objetivos principales estaban relacionados con la comparación de los niños que asistían a las condiciones de "TEA" y "Prematuridad". Los datos basados en un tamaño de muestra mayor y con un grupo de control a término permitirán un análisis más profundo de los factores que afectan a los resultados óptimos del tratamiento en esta población.

En tercer lugar, los niños fueron reclutados a los 18 meses si mostraban signos preocupantes de TEA en el M-CHAT-R/F y el ADOS-2. Tras el programa de intervención, el diagnóstico de TEA se confirmó a los 24 meses de edad. Aunque la literatura reciente sugiere que el diagnóstico de TEA podría estabilizarse a esta edad, todavía hay controversia sobre la edad a la que el diagnóstico de TEA es estable (de 24 a 36 meses). Sin embargo, el objetivo principal del programa fue comenzar lo más pronto posible y sin retrasos entre los servicios de detección e intervención. Los estudios futuros deberían incluir un seguimiento a los 30-36 meses para confirmar el diagnóstico de TEA y apoyar los resultados obtenidos en el campo de los TEA.

Por último, no hubo datos de seguimiento para evaluar el impacto a largo plazo de los efectos del tratamiento. El seguimiento limitado sólo permitió examinar brevemente la sostenibilidad de la intervención. Por lo tanto, los estudios futuros deberán abordar estas limitaciones para determinar si los efectos del tratamiento varían a la salida y en el

seguimiento. También es fundamental evaluar la eficacia de este programa frente a la de otros tratamientos.

CONCLUSIONES

Esta tesis doctoral ha planteado como objetivo general desarrollar, implantar y evaluar un programa de intervención temprana dirigido a menores en riesgo de trastorno del espectro autista por prematuridad. Nos hemos esforzado por que el estudio esté bien fundamentado, no solo en cuanto a qué procedimientos pueden ser los más apropiados para enseñar habilidades deficitarias en los bebés en riesgos, sino que también hemos buscado criterios, que apoyen nuestro planteamiento que surgen directamente de la experiencia de los profesionales y de las expectativas de las familias. Humildemente consideramos que ésta es una forma innovadora y, por supuesto, integradora de abordar la búsqueda de nuevos enfoques de intervención preventiva para tratar los signos tempranos de TEA.

Nuestro camino no ha hecho más que empezar. Por el momento, nuestro estudio de encuesta nos ha permitido (1) conocer las opiniones y perspectivas de familias de menores con TEA y profesionales que trabajan con este grupo en relación con los servicios, así como conocer aquellos puntos en los que las familias y profesionales tenían mayor o menor satisfacción; (2) conocer qué técnicas focalizadas a la mejora de habilidades de comunicación social son más eficaces; y (3) poner en marcha de un programa de intervención temprana para menores con TEA en base a los resultados obtenidos en los estudios anteriores. A la luz de los resultados, se obtienen las siguientes conclusiones:

- Las Prácticas de Intervención Focalizadas (PIF) actualmente disponibles, dirigidas a mejorar las habilidades socio-comunicativas generalmente afectadas en los niños pequeños con TEA (imitación, atención conjunta y juego), son efectivas, adecuadas y suficientemente idóneas para ser utilizadas como tratamiento en los servicios de intervención temprana.

- Los cuidadores y/o los profesores podrían desempeñar un papel activo en los PIF a la hora de obtener efectos positivos. Además, la participación activa de los cuidadores o profesores podría reducir los costes de los servicios públicos, al disminuir el número de horas de intervención que los terapeutas dedican a cada niño. Además, dado que estos PIF se centran en habilidades específicas, el tiempo de aplicación sería más corto, algo que podría, a su vez, llevar a reducir el coste del tratamiento y hacerlo así más asequible.
- Aunque las familias y los profesionales de la comunidad del autismo están ampliamente satisfechos con los servicios, se encontraron sin embargo diferencias entre estos dos grupos. En particular, las familias de los menores con TEA informaron de una menor satisfacción general con los servicios, así como de mayores edades de los niños y mayores retrasos en el acceso a los servicios que los profesionales que trabajan habitualmente con niños con TEA.
- Los resultados sugieren que, tanto en las familias como en los profesionales, una mayor satisfacción con los servicios se asocia con edades más bajas de detección y diagnóstico, ya que esto permite que la intervención comience antes.
- Son los padres los que siguen siendo cruciales para la detección de los primeros signos de TEA. Las familias nos dicen que se necesitan profesionales colaboradores, inclusivos y autocríticos, y que deben participar en todos los aspectos de la atención a su hijo/a.
- Las políticas de los servicios y las investigaciones futuras deberían centrarse en reducir los retrasos en el acceso a los servicios, por ejemplo, mediante la

aplicación de programas de detección precoz específicos para los TEA, con el fin de aumentar la satisfacción de las familias con los servicios y, por tanto, posiblemente reducir su estrés y mejorar su bienestar.

- Estos resultados nos permiten afirmar que se puede producir un cambio en los problemas centrales del desarrollo de los niños prematuros con TEA, utilizando una intervención de baja intensidad dirigida a las habilidades sociales y de comunicación.
- A pesar de que estas áreas centrales de deterioro no son fáciles de cambiar, la intervención tuvo un efecto apreciable. La mayoría de los participantes mejoraron significativamente en las habilidades sociocomunicativas, el desarrollo cognitivo, el lenguaje y la conducta adaptativa, y se redujeron los signos de TEA.
- En consecuencia, es necesario investigar los efectos a más largo plazo de la intervención y de una población más amplia de niños prematuros, con el fin de examinar los posibles mediadores y moderadores de los resultados de la intervención en las habilidades sociales y comunicativas de los niños.

CONCLUSIONS

The general objective of this doctoral thesis is to develop, implement and evaluate an early intervention program aimed at children at risk of autism spectrum disorder due to prematurity. We have tried to ensure that the study is well grounded, not only in terms of what procedures may be most appropriate for teaching deficient skills to at-risk infants, but we have also sought criteria, in support of our approach, that arise directly from the experience of professionals and the expectations of families. We humbly consider this to be an innovative and, of course, integrative way of approaching the search for new preventive intervention approaches to address early signs of ASD.

Our journey has only just begun. For the moment, our survey study has allowed us (1) to know the opinions and perspectives of families of children with ASD and professionals working with this group in relation to the services, as well as to know those points in which families and professionals were more or less satisfied; (2) to know which techniques focused on the improvement of social communication skills are more effective; and (3) to implement an early intervention program for children with ASD based on the results obtained in the previous studies. In light of the results, the following conclusions are drawn:

- Focused Intervention Practices (FIPs) aimed at improving the social communicative skills generally affected in young children with ASD (imitation, joint attention and play), are effective, adequate, and suitable enough to be used as treatment in early-intervention services.
- Caregivers and/or teachers could play an active role in FIPs when it comes to obtaining positive effects. Moreover, active participation by caregivers or teachers could reduce the costs incurred by public services, by reducing the number of intervention hours that therapists devote to each child. In

addition, since these FIPs focus on specific skills, application time would become shorter, something that could, in turn, lead to lowering the cost of treatment and thus rendering it more affordable.

- Although families and professionals in the autism community are broadly satisfied with services and that children's ages were lower and delays in access to services were shorter than in other studies, differences were nevertheless found between these two groups. In particular, families of children with ASD reported lower overall satisfaction with and higher child ages and longer delays in access to services than did professionals who routinely work with children with ASD.
- The results suggest that, in both families and professionals, greater satisfaction with services is associated with low ages of detection and diagnosis, as this enables intervention to begin sooner.
- Parents who are still crucial for the detection of the first ASD signs. Families are telling us that there is a need of collaborative, inclusive and self-critical professionals, and that they should be involved in every aspect of care for their child.
- Service policies and future research should focus on reducing delays in access to services, through, say, the implementation of early ASD-specific detection programmes, in order to increase families' satisfaction with services and thereby possibly reduce their stress and improve their wellbeing.
- These results allow us to claim that change can be brought about in core developmental problems among preterm children with ASD, using a low-intensity intervention targeting social and communication skills.

- Even though such core areas of impairment are not easy to change, the intervention had an appreciable effect. Most of the participants improved significantly in socio-communicative skills, cognitive development, language, and adaptive behavior, and ASD signs were reduced.
- Accordingly, research into the longer-term effects of the intervention and a larger population of preterm children are needed, in order to examine possible mediators and moderators of intervention outcomes on children's social and communication abilities.

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