Identificar desarrollo atípico: ¿una tarea de los trabajadores de guardería?

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Resumen
Identificar los signos tempranos de déficit del neurodesarrollo es importante para asegurar diagnóstico oportuno e intervención temprana. Los trabajadores de guarderías pueden estar en posición de privilegio para notar potenciales desvíos del desarrollo, pero no está claro si pueden reconocer definidamente los sutiles signos tempranos del desarrollo atípico. Sesenta trabajadores de guarderías examinaron videos hogareños de niños muy pequeños con síndrome de X frágil y niños con desarrollo típico. Los resultados mostraron que la mayoría de los trabajadores en guarderías pueden identificar desarrollo típico y atípico en general y podrían por lo tanto tener un rol importante en la identificación temprana. La experiencia especial de trabajo y el entrenamiento pedagógico avanzado parecieron impulsar la sensibilidad de los trabajadores de guarderías para detectar figuras atípicas en el desarrollo temprano y proveer vigilancia diaria efectiva.

Palabras clave: Trabajadores de guardería. Déficit del desarrollo. Identificación temprana. Análisis retrospectivo de video. Síndrome del X frágil

La comprensión científica y la conciencia de niños pequeños en riesgo de desórdenes del desarrollo han aumentado sustancialmente en las últimas décadas. El creciente número de estudios sobre el desarrollo infantil temprano ha provisto nuevos hallazgos empíricos. Que han alterado las perspectivas sobre fenómenos edad-específicos, mecanismos, y perfiles asociados con varias alteraciones del desarrollo y genéticas (Johnson et al. 2015; Marschik et al. 2013; Messinger et al. 2013; Thomas et al. 2009). Los avances relacionados con estudios sobre desórdenes del desarrollo detectados tarde (LDDDs), por ejemplo, son particularmente evidentes en la investigación sobre el trastorno del espectro autista (TEA; Bölte et al. 2016, 2013; Bontinck et al. 2018; Bussu et al. 2018; Gliga et al. 2014; Messinger et al. 2013; Roeyers 2018). Los avances en la investigación también han contribuido a lo que parece ser mayor reconocimiento de la significancia de la detección temprana del desarrollo atípico para posibilitar intervención y apoyo oportuno.
En muchos países europeos, donde el número de madres que trabajan se mantiene en aumento, la necesidad de servicios de guarderías públicas aumenta firmemente. En Austria, por ejemplo, la proporción de infantes menores de 2 años de edad que asisten a centros de día se duplicó desde 2007 a 2017. En algunas áreas urbanas, tanto como el 45 % de los niños de edad 0-2 años asisten a guarderías, de los cuales más del 90% pasan 6-10 hs. por día con cuidadores profesionales (Statistik Austria 2018). Estos trabajadores de guarderías podrían entonces ser vistos entre las personas más importantes en el cuidado para muchos niños pequeños. En consecuencia, parecería importante investigar el papel potencial de los trabajadores de guarderías en la detección temprana de desarrollo desviado en niños pequeños. Por ejemplo, podría ser factible convocar a los trabajadores de guarderías en el proceso de tamizaje de TEA? Tales temas han sido resaltados en un número de publicaciones recientes (Branson et

al. 2008; Dereu et al. 2012; Janus et al. 2018; Janvier et al. 2016; Larsen et al. 2018a, b; Nordahl-Hansen et al. 2018, 2013).

En efecto, los resultados de algunos estudios sugieren que los trabajadores de guarderías tienen la competencia para reportar con exactitud signos tempranos de autismo aplicando listas de verificación bien diseñadas (Dereu et al. 2010; Larsen et al. 2018b). También podría haber algunas ventajas potenciales en incorporar la ayuda de los trabajadores de guarderías porque con frecuencia este personal tiene entrenamiento y conocimiento del desarrollo infantil temprano, y considerable experiencia práctica en trabajar con niños de edades similares y diversos perfiles del desarrollo. Dado que los déficits del desarrollo afectan al menos a 7-9 % de los niños pequeños (Olusanya et al. 2018; Zablotsky et al. 2017), puede ser importante involucrar los trabajadores de guarderías como potenciales detectores de discapacidad del desarrollo. Al revés de los padres, es más factible que sean más objetivos. El aporte de su vigilancia diaria puede complementar nuestra comprensión del período prodrómico de LDDDs y potencialmente contribuir a identificación más temprana, siendo entonces de importante interés público y científico.

El Síndrome del X Frágil (SXF) es uno de estos LDDDs. Similar a varios otros desórdenes, tal como el síndrome de Rett (RTT) y TEA, las presentaciones relacionadas físicas y del comportamiento son con frecuencia sutiles y difíciles de detectar al principio, lo cual por supuesto hace la identificación temprana un mayor desafío. Pese a ello, signos atípicos tempranos que emergen en los primeros años de vida en diferentes dominios se han reportados como perceptibles para los padres de niños con SXF (Hinton et al. 2013; Zhang et al. 2017). Algunos de estos signos son frecuentemente presentados y capturados en videos hogareños. Con un procedimiento de puntos de referencia analizando retrospectivamente videos hogareños de niños con SXF, nosotros, entre otros investigadores, demostramos que estos signos pueden ser fácilmente identificados y clasificados por profesionales (Zhang et al. 2018; ver también Baranek et al. 2005). Fenotipos tempranos de SXF presentan un amplio espectro de formas neuroconductuales atípicas en varios dominios (ej., motor, cognición, lenguaje- habla, social-comunicación), que no son específicos de SXF (Haessler et al. 2016; Hagerman 2002; Kidd et al. 2014; Marschik et al. 2014; Raspa et al. 2017; Roche et al. 2018; Zhang et al. 2017). Dado que los signos tempranos de SXF se superponen con peculiaridades de otros niños con otras alteraciones genéticas o del desarrollo, identificar estos signos requiere sensibilidad para el desvío del desarrollo en general y no necesariamente ser experto en un síndrome específico.

En el presente estudio, intentamos utilizar secuencias de video hogareño mostrando comportamiento en diferentes áreas del desarrollo en niños con desarrollo típico y niños con SXF como material para captar la preocupación de los trabajadores de guarderías en el desarrollo temprano. Adaptamos un proceso de revisión de videos usado por Burford et al. (2003) agregando determinaciones de referencia de las características del neurodesarrollo. Específicamente, nos propusimos investigar si los trabajadores en guarderías perciben la diferencia entre niños con desarrollo típico y atípico. Además, quisimos encontrar si ellos podían identificar en forma precisa las características típicas y atípicas del desarrollo temprano. Dado que los trabajadores de guarderías tienen diferente entrenamiento y experiencia de base, también nos propusimos responder si estos factores modifican las percepciones de los trabajadores sobre los fenómenos del desarrollo temprano.

Métodos

Participantes

Doce centros de día y una escuela vocacional para maestros de jardín en Graz, Austria y regiones circundantes fueron contactados. Siete centros con 36 trabajadores (21 maestros de jardín, un varón; 15 cuidadores, todas mujeres) y la escuela con 25 estudiantes para maestros (todas mujeres) acordaron participar. De las 21 maestras, 8 recibieron entrenamiento adicional pedagógico avanzado o en necesidades especiales. El entrenamiento profesional para maestras de jardín de infantes en Austria es de 5 años, y para cuidadores 6 meses. Ninguno de los 15 cuidadores en el estudio recibió entrenamiento adicional relativo al cuidado de niños. Al momento de recolectar los datos, la edad media de los maestros fue 42 años (DS=11.40), y la de los cuidadores fue 43 años (11.50). Los 25 estudiantes pre-servicio habían recientemente finalizado el penúltimo año de entrenamiento, y tenían en promedio 17 años (rango 17-19). Su currículum en el penúltimo año incluye 2 hr supervisadas de entrenamiento activo por semana en centros de día. Un estudiante pre-servicio abandonó por razones personales, quedando la muestra final con 60 participantes (21 maestros, 15 cuidadores, 24 estudiantes de enseñanza pre-servicio).
Sumado a los niveles de entrenamiento, recolectamos datos de la experiencia laboral de los participantes, incluyendo la extensión general y si el participante tenía experiencia profesional con niños <2 años de edad y/o con alteración del desarrollo.

Materiales

El proyecto Ciudadano Científico está enlazado a un proyecto mayor sobre desarrollo temprano de individuos con SXF (Zhang et al. 2017). Para el estudio actual, extrajimos datos de nuestra base de datos GUARDIAN (Graz University Audiovisual Research Database for the Interdisciplinary Analysis of Neurodevelopment). Seleccionamos grabaciones de 13 niños con SXF y 7 niños con desarrollo típico (DT), de los cuales estaban disponibles videos de los primeros 24 meses de vida. Todos los niños con SXF recibieron un diagnóstico clínico y genético, y todos los videos fueron grabados antes del diagnóstico. Todos los niños con DT eran de nacimientos simples, nacidos a término de embarazos y partos sin particularidades. Al momento de la preparación de este manuscrito, todos eran mayores de 7 años de edad, ninguno había recibido un diagnóstico de ningún desorden del desarrollo ni genético, ni tampoco alguno de sus familiares. Se redondearon 35 horas de grabación (25 horas de niños con SXF), incluyendo contenidos de rutinas diarias o reuniones familiares especiales, que fueron recortados en clips por un asistente de la investigación. Los clips se cortaron según lineamientos, que fueron por cambios en la posición del niño (ej., en prono/supino, sentarse en una hamaca, caminar con soporte) y la presencia/ausencia del cuidador en la escena; para detalles técnicos de los clips por favor ver Zappella et al. (2015). Siguiendo los objetivos de este estudio, inicialmente seleccionamos 40 clips en el grupo SXF y 20 en el grupo DT cubriendo diferentes bandas etarias de los primeros 2 años de vida (1-6, 7-12, 13-18, y 19-24 meses). Los clips fueron elegidos para presentar comportamientos en diferentes áreas del desarrollo (ej, motor, social-comunicación)

del niño objetivo. Después de una prueba piloto y de pruebas intramuros, retuvimos 15 clips para recortar la duración del experimento (por favor ver los datos de la colección más abajo, dejando 10 videos de niños con SXF (videos SXF de aquí en más) y 5 de niños con DT (videos DT de aquí en más). Estos 5 videos DT fueron comparables a 5 de los 10 videos SXF en edad y bases (Tabla 1). Todos los clips tenían una extensión de 1 minuto (± 5 segundos).
El presente estudio fue aprobado por el Institutional Review Board de la Universidad Médica de Graz, Austria (29-054 ex 16/17). Los participantes y padres que compartieron sus datos dieron su consentimiento informado escrito parea participar en el estudio y la publicación de los resultados.

Recolección de datos

La revisión de videos se llevó a cabo para cada participante individualmente en una habitación separada, tranquila en el jardín de infantes o escuela donde el participante trabajaba o era entrenado. A los participantes se les dio información de base acerca del estudio y se les instruyó para observar marcadores de desarrollo temprano que pudieran contribuir a la identificación oportuna de desórdenes del desarrollo diagnosticados tarde. Después de firmar el consentimiento informado, los participantes fueron invitados a revisar los 15 videoclips de los niños entre 8 y 23 meses de edad. También fueron informados que algunos de los niños se estaban desarrollando típicamente, y que algunos fueron posteriormente diagnosticados con un desorden del desarrollo. El término (síndrome X frágil" no fue mencionado. Los participantes fueron estimulados a comentar cada video basados en su experiencia con niños de edad similar mientras tenían en cuenta los diferentes dominios del desarrollo. El orden para presentar los 15 video clips fue randomizado entre los participantes. Antes de pasar el video, el participante fue informado de la edad y sexo del niño, pero no si el niño había sido diagnosticado. Cada video fue pasado dos veces en una computadora, controlado por un investigador. El participante fue invitado a tomar notas después de la primera vuelta. Después de una segunda vuelta, el participante fue invitado a comentar sobre cualquier aspecto llamativo del niño, tanto aspectos positivos como preocupaciones. Nunca se pidió al participante que juzgara desarrollo normal versus anormal. El siguiente clip se mostraba cuando el participante estaba listo. Se hacían pausas según necesidad, pero no durante un mismo clip. La revisión de los 11 clips llevó alrededor de 60 minutos para cada participante. Un investigador guió el procedimiento de revisión, tomó notas, y pasó el programa de la computadora sin entrar en discusión acerca de los comentarios del participante. El registro del audio sobre la pantalla se realizó con Corel VideoStudio X9, usando la función de "Captura de Pantalla".

Transcripción y puntuación

Los 60 registros de audio fueron luego transcriptos verbatim en el laboratorio iDN por el autor I.K.T. Alrededor de 15% de los materiales de audio fueron randomizadamente seleccionados y transcriptos por otro asistente de la investigación para chequear la exactitud y consistencia para el punteo de las transcripciones. No hubo diferencias

significativas. Entonces, toda la puntuación estuvo basada únicamente en las transcripciones de I.K.T.

Tres miembros del equipo de investigación experimentados iDN (D.Z., P.B.M., y C.E.) revisaron los 15 video clips sin conocer los antecedentes médicos de los niños. Ellos delinearon separadamente todas las características dignas de atención para cada niño objetivo, siendo particularmente positivas o avanzadas para la edad (ej., caminar de lado/en borde a los 10 meses de edad) así como las atípicas o inadecuadas para la edad (ej., referencia a hitos del desarrollo demorados y/o comportamiento anormal). Las características cubrían neurofunciones diferentes de varias áreas del comportamiento (ej., motor, social, verbal, apariencia física, movimientos repetitivos). Pese a que conductas típicas o acorde a la edad (ej., sentarse sin apoyo) fueron observadas en los clips de niños con SXF, ninguno fue señalado como destacado o avanzado para la edad. Entonces, ninguna característica positiva o avanzada para la edad fue punteada para ningún video clip SXF. Para los videos DT se puntuaron características positivas y atípicas. Los tres autores también nombraron una o dos "características primarias" para cada clip, en referencia a la característica más llamativa observada en la escena. Luego discutieron sus análisis y alcanzaron consenso sobre una lista de puntos de referencia para cada clip. Cada lista consiste de al menos tres características además de una primaria. Múltiples características fueron identificadas frecuentemente en la misma área del desarrollo (ej., motor, social) para un video. En dos clips de niños con SXF y uno de un niño DT, dos características primarias fueron identificadas (Tabla 1).

Para examinar los comentarios de los participantes, implementamos un esquema de puntuación adaptando el concepto de optimalidad introducido por Prechtl (1980) y la metodología analítica inductiva del corpus de Lindseth y Norberg (2004). Scores más altos indican mayor congruencia con la lista de referencia. Cada comentario hecho sobre un clip fue asignado un score entre 0 y 7; los detalles de puntuación se muestran en la Tabla 2.

En el siguiente paso, D.Z. y I.K.T. examinaron separadamente todas las 60 transcripciones. El coeficiente de correlación intraclase (ICC) entre los puntuadores fue .94 (IC 95%, .90- .97), basado en el modelo de medidas promedio, acuerdo absoluto, y efectos mixtos de doble sentido. Luego se discutieron las discrepancias hasta llegar a consenso, Además, contamos el número de participantes para cada video que comentaron que el niño objetivo era normal o no expresaban preocupación acerca del desarrollo del niño (Tabla 1).

Tabla 1 Características de los puntos de referencia de videos de pacientes con SXF (Síndrome X frágil) y DT (desarrollo típico del niño).

	Código de video[a]	Edad (meses)	Áreas con características de la lista de referencia[b]							n/61
			Mot	Fis	MR	S/C				
						Fac	Int	Len	Voc	
FXS videos	SXF1	8	++		+	+			+	17
	SXF2	9		+	+	++	+		+	13
	SXF3	9	++	+				+	+	15
	SXF4	10	++			++	+	+		7
	SXF5	10	+		++		+			9
	SXF6	12	++		++	+	+	+		6
	SXF7	12	+			++				26
	SXF8	13	+		+		++		+	6
	SXF9	16	+		+	++		+		7
	SXF10	23	+		+		+	++	+	21
DT videos	DT1	8	+		+		+	++		31
	DT2	9	+		++		+		+	48
	DT3	10	+						++	46
	DT4	16	++				+	++		9
	DT5	23					++	+		48

++ Esta área consiste en las características primarias definidas para este video sobre la lista de referencia. Para tres videos (FXS4. FXS6 y TD4), dos características principales han sido identificadas.
+ Esta área consiste de características no primarias identificadas para ese video.
n/60: Número de participantes que consideraron al niño como normal
La lista de referencia incluye características específicas de edad (en orden alfabético) en cuatro áreas (en letra remarcada negrilla) **Mot** (motor) de acuerdo a edad avances positivos (voltear de lado, arrastre, caminar, manipulación fina, estar de pie independiente, buen control postural). Características inadecuadas para la edad (no movimientos anti gravitatorios, no manipulación, no sentarse sin apoyo, no estar de pie independiente, no caminar libre, Tocando objetos sin agarrarlos). Y además mala postura- desplomado/hipotónico, grande protrusión de lengua. **Fis** (apariencia física) orejas muy grandes, estrabismo. **RM** (movimientos repetitivos), balanceo de cuerpo, sacudidas de manos y brazos, manos abriéndolas y cerrándolas, sacudir la cabeza. **S/C** (social comunicación), con (a) Fac (expresión facial), sonrisa torpe, llorando como expresión facial durante vocalización placentera, mirada vacía, expresión facial no adaptativa, expresión facial con retardo. (b) Int (interacción): características positivas (atento, paciente, comprometido en el juego, interactivo y responsivo) y características negativas (reacciones exageradas, ausencia de adaptación y reacción en el juego social, no hay respuestas cuando le hablan, pasivo/sin iniciativa en el juego social, hostilidad con otros niños; (c) Len (lenguaje): inadecuadas declaraciones para la edad, neologismos; (d) Voc (vocalizaciones) vocalizaciones inspiratorias y espiratorias, vocalización de tono agudo, voz ronca, vocalización monótona/repetitiva, ecolalia, voz apagada, vocalización no modulada.
[a] Videos con resultados de desarrollo neurológico típicos de niños: TD1 a TD5 fueron comparables con FXS1, FXS2, FXS4,FCS9, FXS10 respectivamente en edad ascendente corregida.
[b] Múltiples características fueron frecuentemente identificadas en la misma área para un video.

Análisis de datos

Los datos fueron analizados con SPSS, versión 25 (SPSS Inc, Chicago, IL). Se realizó test de Mann- Whitney para comparar la diferencia de dos grupos independientes si no podía asumirse distribución normal. T test fue utilizado para comparar la diferencia entre dos medias de distribución normal. Coeficiente de correlación de Pearson ® fue calculado para estimar la asociación de dos variables continuas con distribuciones

normales. Correlación de orden rango de Spearman (rho) fue aplicada para estimar la asociación entre dos variables si no podían asumirse distribuciones normales para ambas. Por medio de ANOVA se comparó las medias entre tres o más grupos independientes.

Tabla 2 Esquema de puntuación para videos con SXF (Síndrome X frágil) y DT (desarrollo típico-normal)

Primera puntuación	Definición	Primera puntuación	Definición
7	Correcta identificación de 3 o más características Incluyendo las características primarias	7	Correcta identificación de 2 o más características Incluyendo las características primarias
6	Correcta identificación de 2 características Incluyendo las primarias	6	Correcta identificación de la característica primaria o muchas no primarias
5	Correcta identificación de la característica primaria o muchas no primarias	5	Correcta identificación de ninguna característica primaria
4	Correcta identificación de ninguna característica primaria	4	No identificación de alguna característica pero haciendo comentarios de características relacionadas
3	No identificación de alguna característica pero haciendo comentarios de características relacionadas[a]	3	No se encuentra nada digno de llamar la atención
2	No hay observaciones irrelevantes[b]	2	No hay observaciones irrelevantes
1	No hay interpretación contraria a la características[c]	1	No hay interpretación contraria a las características
0	Ninguna preocupación	0	Solamente interpretación contraria a las características

Interpretación contraria lleva a deducción de un punto. Por ejemplo si un participante identifica correctamente una característica primaria de Síndrome X Frágil (SXF) todavía el paciente referido puede tener una característica normal (interpretación opuesta). La Calificación podría ser 4 puntos (5 puntos y deducción de 1 punto).

[a] Por ejemplo una característica de la lista de referencia de un video fue "mirada vacía". Un participante comentó "El mira de algún modo extraño, yo me pregunto si él puede ver todo, quizá él es ciego".

[b] Los participantes hacen comentarios rastreables para la escena, pero falla para identificar alguna característica de la lista de referencia.

[c] Interpretación opuesta: una conducta obviamente atípica, parece como normal o viceversa

Eta cuadrado (η^2) fue elegido para reportar el tamaño de efecto de U test y ANOVA. Se informó Cohen d para el tamaño de efecto de t-test. Alpha fue establecido en .05, dos colas para todos los análisis.

Resultados

¿Desarrollo normal versus atípico?

Más participantes comentaron que el niño objetivo estaba desarrollándose normalmente en los videos DT (Md=46 de 60, rango 9-48; Tabla 1) comparado con los videos SXF (Md=11, rango 6-26), Mann Whitney U= 5.50, Z= 2.40, p=.013, con tamaño de efecto grande η^2 =.38. Para todos excepto uno de los videos DT, más de la mitad de los participantes remarcaron espontáneamente que el niño objetivo se estaba desarrollando normalmente. Este video mostró a un niño de 16 meses, que no produjo vocalizaciones, ni caminaba o se paraba independientemente en el clip.

Consistencia entre los participantes y el análisis de puntos de referencia

El análisis inicial de los scores brutos medios de los 5 videos DT así como los 10 videos SXF reveló distribuciones normales. El score medio bruto para los videos DT entre participantes fue 4.04 (DS= 1.24) y 3.13 (DS=.93) para los videos SXF. El score estándar

medio bruto de videos DT (Z_{TD}) y SXF (Z_{SXF}) estuvo significativamente correlacionado, r=.43, p=.001. Esto es, participantes que tuvieron score alto (o bajo) en los videos DT también era factible que lo tuvieran en los videos SXF. El score general de un participante es la suma estándar de Z_{TD} y $Z_{SXF,}$ ej., score general= Z {Z_{TD}+ Z_{SXF}}. El score general estuvo en rango entre -2.23 a 2.26 entre los participantes. Un score general más alto en un participante indica mayor consistencia con el análisis de punto de referencia de los videos. Examinamos las siguientes variables:

Nivel de entrenamiento

Los scores generales difirieron significativamente entre maestros con o sin entrenamiento pedagógico adicional o en necesidades especiales. (**st**; maestros $^{st+}$ y maestros $^{st-}$ de aquí en más), cuidadores, y maestros estudiantes pre-servicio, F (3, 56)= 5.45, p=.002, η^2 =.23. Una post comparación Tukey reveló significativamente mayores scores generales para los maestros $^{st+}$ (M= 1.15, DS=.96) comparado con maestros $^{st-}$ (M=.05, DS=.92, d= 1.17, p= .043), cuidadores (M=-.18, DS=.99, d= 1.36, p=.008), y con estudiantes pre-servicio (M=-.30, DS=.82, d=1.62, p=.001). No se encontró diferencia estadísticamente significativa en el score general entre ninguno de los dos grupos entre maestros $^{st-}$, cuidadores, o estudiantes pre-servicio.

Experiencia laboral

Los estudiantes pre-servicio fueron excluidos de este análisis porque aún no habían comenzado a trabajar diariamente. La extensión de la experiencia laboral de los 34 trabajadores de guardería no tuvo distribución normal. La mediana fue 7.5 años (rango 2-34). De los 20 maestros, la mediana fue 7.5 años (rango 2-34); de los 14 cuidadores, fue 8.0 años (rango 2-19). La diferencia no fue significativa, Mann- Whitney U= 136.00, Z=.141, p=.888. Los scores generales de los trabajadores en guarderías se correlacionó moderadamente con la extensión de su experiencia de trabajo, rho Spearman= .34 (p=.052). No se encontró diferencia significativa en el score general entre participantes con experiencia laboral mayor (ej., extensión de experiencia por encima de la mediana, n= 16; score general M=.51, DS=.96) o más corta (n=18, score general M=.08, DS=1.03), t-test de Student de muestra independiente arrojó t= 1.26, p=.218.

Experiencia relevante para la tarea

Experiencia relevante para la tarea requiere: (a) cantidad razonable de experiencia general de trabajo como trabajador de guardería (ej., extensión de la experiencia al menos de 5 años), y, (b) experiencia profesional trabajando con niños pequeños (ej., menos de 2 años) y/o (c) niños con desórdenes del desarrollo. Doce maestros y 6 cuidadores cumplían con estas condiciones y fueron clasificados como el grupo con "rica" experiencia relevante en la tarea, incluyendo 8 de los maestros $^{st+}$ **.** Los scores medios generales del grupo "rico" fue .68 (DS=.94). Los restantes 8 cuidadores que nunca trabajaron con niños pequeños ni con niños con desorden del desarrollo, o que tenían menos de 5 años de experiencia profesional como trabajador en guardería fueron tomados en el grupo de experiencia de trabajo "limitada". La media de su score

general fue-.16 (DS=.92). La diferencia del score general entre los grupos "rico" y "limitado" fue significativa, t=2.65, p=.012, d=.90. Esto es, participantes con rica experiencia de tareas tuvieron scores más altos. El análisis más avanzado reveló que los maestros con rica experiencia `para la tarea (M=.98, DS=.89) puntuaron más alto que los maestros con experiencia limitada (M=-.21, DS=.95), t= 2.86, d=1.29, p= .010. Los scores de los cuidadores con experiencia rica (M=.08, DS=.75) o limitada (M=-.11, DS=.95), sin embargo, no fueron significativamente diferentes, t=.41, p=.688. En otras palabras, la experiencia relevante para la tarea influenció los scores de los maestros, pero no los de los cuidadores.

DISCUSIÓN

El presente estudio, basado en nuestras previas investigaciones y estudios metodológicamente similares (Burford et al. 2003; Dereu et al. 2012; Marschik et al. 2012; Zhang et al. 2017, 2018), presenta un enfoque novedoso para investigar la preocupación de los trabajadores de guardería sobre el desarrollo temprano y la fase prodrómica de desórdenes del desarrollo reconocidos tarde. Sacamos partido de la visión y conocimientos de los participantes sobre los hitos del desarrollo infantil temprano y los desvíos cualitativos del desarrollo normal durante los 2 primeros años de vida. Pese a que los participantes no fueron explícitamente invitados a diferenciar entre desarrollo normal o atípico, sus comentarios indicaban que ellos claramente percibían las diferencias entre DT y niños con desarrollo desviado, en este caso SXF. Preocupaciones menos significativas pero confirmación más espontánea de normalidad fue hecha en los niños DT comparados con SXF. Es decir, desarrollo normal o aberrante en general fue perceptible para los trabajadores de guardería.

Este hallazgo es similar a los de Marschik et al. (2012) en un estudio que estuvo enfocado en la percepción acústica Gestalt. Ellos encontraron que tanto los especialistas en lenguaje y los no-profesionales pudieron distinguir vocalizaciones típicas de atípicas expresadas por niños pequeños. En el estudio actual, la mayoría de los participantes notaron que *algo* no estaba bien al observar a un niño que luego fue diagnosticado con SXF, pero al tener que especificar *qué* estaba mal, ahí aparecieron las diferencias entre los diferentes grupos. Específicamente, las observaciones de los maestros $^{st+}$ (ej, con entrenamiento especializado adicional en desarrollo atípico) fueron más consistentes con las determinaciones de puntos de referencia de los científicos del desarrollo. Los maestros $^{st-}$ (ej, sin entrenamiento adicional) se desempeñaron menos bien y se compararon con los estudiantes pre-servicio en el penúltimo año de entrenamiento, y, también, similar a los cuidadores (ej., con entrenamiento profesional mucho más corto). Esto, no sorprendentemente, sugiere que el entrenamiento especializado más allá de la preparación general de cuidado en guardería puede resaltar la sensibilidad para reconocer características típicas y atípicas en el desarrollo temprano (ver también Daniels and Mandell 2014; Lee et al. 2005; Reilly et al. 2015).

Frente a estos hallazgos, queda la pregunta acerca de qué rol tiene la experiencia, ¿si es que tiene alguno? Interesantemente, los maestros y los cuidadores que habían trabajado más tiempo en unidades de cuidado de día no puntuaron significativamente mejor que aquellos con menos experiencia. Esto fue replicado por los hallazgos en

maestros $^{st-}$ los estudiantes pre-servicio que tuvieron desempeño bastante similar, ambos con un nivel comparable de entrenamiento, mientras que los maestros habían tenido práctica más extensa. En otras palabras, la duración de la experiencia no demostró ser crítica para desarrollar "ojos sensibles" para determinar fenómenos tempranos del desarrollo. En cambio, el *perfil* de experiencia pareció jugar un rol vital en este aspecto. En nuestro estudio actual, no fue el cuidado de niños en guardería el que importó, sino en cambio, la "experiencia relevante para la tarea", esto es, tener experiencia práctica con ciertos grupos de niños (ej., niños menores de 2 años y/0 niños con una discapacidad del desarrollo) estuvo asociado con puntuaciones más altas. Pese a esto, debe ser puntualizado que la experiencia relevante para la tarea pareció tener impacto sólo si había una sólida base de entrenamiento. Entonces, sólo los maestros, pero no los cuidadores, parecieron beneficiarse de la experiencia en la tarea al desempeñarse mejor al compararse con sus compañeros con menos experiencia. Esto sugiere que sin suficiente entrenamiento general, sólo acumular experiencia podría no inducir realmente mejor mirada para el desarrollo temprano del niño.

Incidentalmente, peo no inesperado, los 8 maestros $^{st+}$ estuvieron en el grupo de los 16 participantes con rica experiencia. Estos 8 maestros que buscaban entrenamiento avanzado, también practicaban concordantemente su conocimiento en su trabajo diario. Por lo tanto el entrenamiento especializado y las prácticas relacionadas pueden entonces haber sumado en el reconocimiento de características típicas versus atípicas en el desarrollo temprano. Para los trabajadores de guarderías que interactúan intensamente con niños de diferentes edades y perfiles de desarrollo cada día, notar que *algo* puede estar mal con el desarrollo de un niño podría no ser un desafío. Sin embargo, para poder más precisamente especificar las anomalías del desarrollo que pueden ser indicadoras de una discapacidad del desarrollo, el entrenamiento especializado y experiencias ricas parecen ser factores determinantes.

Notablemente, los scores brutos evaluados acorde a las escalas de puntuación (Tabla 2) indicaron que muchos de los maestros $^{st-}$, los cuidadores, y los estudiantes pre-servicio fallaron en reconocer alguna característica específica de la lista de investigación de puntos de referencia en la mayoría de las secuencias de video (ej, haber puntuado en promedio menor de 4 para los videos SXF y menor de 5 para los videos DT). Las características que fueron salientes para los profesionales que establecieron los puntos de referencia parecían haber escapado a los ojos de muchos de los participantes. Este hallazgo parecería traer a colación el problema de la confiabilidad de esperar que los trabajadores de guarderías detecten signos específicos de advertencia de una posible discapacidad del desarrollo en niños muy jóvenes. Similarmente, estudios enfocando en niños mayores revelaron que el acuerdo de los maestros con otros reportando fenómenos del desarrollo variaron a través de diferentes contextos (Ehlers et al. 1999; Macari et al. 2018; Mattila et al. 2009; Nordahl- Hansen et al. 2013, 2014; Posserud et al. 2006). En contraste, un estudio previo demostró que enfermeras especialistas con extensa experiencia en vigilancia de niños en el hogar y el hospital eran muy capaces de detectar signos tempranos de déficit del desarrollo (ej., RTT) y distinguir los infantes de sus controles con desarrollo típico (Burford et al. 2003). Esto resalta la potencial importancia de estar calificado en entrenamiento y práctica. Los hallazgos también están en línea con nuestro estudio actual en que aquellos trabajadores de guardería con entrenamiento y experiencia avanzados estuvieron mejor habilitados para

identificar características tempranas típicas y atípicas del desarrollo. Otro estudio de una cohorte extensa de Dereu et al. (2010) utilizó una checklist especialmente desarrollada para trabajadores en guardería infantil para tamizar niños con TEA. Después de un corto entrenamiento destacando los signos tempranos de TEA y cómo usar la checklist, los cuidadores fueron probados en su confiabilidad y sensibilidad como informantes de signos tempranos de TEA. Este estudio junto con el nuestro sugieren que los trabajadores de guardería tienen el potencial de distinguir desarrollo típico y atípico en general. Para obtener la mejor información de parte de los trabajadores de guarderías que apoye la identificación temprana del desarrollo atípico, usar una checklist de signos específicamente diseñada y proveer entrenamiento relacionado puede ser efectivo y deseable.

Estamos bien al tanto de que nuestro estudio tiene algunas limitaciones. El diseño experimental utilizando breves secuencias de video no replica completamente el ámbito natural, donde los trabajadores de guardería tienen la posibilidad de observar cada niño a lo lardo de una serie de contextos durante días, semanas, y meses. Más aún, algunos de nuestros participantes podrían no haberse sentido confortables en una situación experimental, que podría haber influenciado negativamente su desempeño. Sin embargo, la revisión de videos provee una aproximación sólida del mundo real y puede entregar información valiosa dentro de un tiempo razonable. Dado que todos los participantes tenían el mismo material para evaluar, las distinciones entre ambos grupos podrían ser atribuidas a sus diferentes sensibilidades para varias características del desarrollo. Es bastante improbable que la simple provisión de ambientes de observación más extensos y relajados llevará a significativamente mejores observaciones de las normalidades y peculiaridades del desarrollo temprano. En su campo de trabajo diario, los trabajadores de guarderías podrían ser reticentes para alarmar padres o profesionales de la salud acerca del desarrollo atípico de un niño debido a razones éticas o personales, aún si ellos han notado signos adversos. Entonces, investigadores y profesionales de la salud necesitan recolectar activamente datos de una manera eficiente de trabajadores de guardería calificados si tienen la intención de aprovechar información crítica adicional que no pueden obtener de los padres (ver también Garcia-Primo et al. 2014; Macari et al. 2018). También el pequeño tamaño muestral es otra limitación relacionada al presente trabajo.

Por último, pero no menos importante, las investigaciones en los signos tempranos de LDDDs han sido hechas en su mayoría retrospectivamente, incluyendo empleo de cuestionarios y checklists así como análisis de videos hogareños. Esto ha sido gradualmente reemplazado por enfoques prospectivos para un número de desórdenes, siendo el principal TEA (ej, Bölte et al. 2013; Isaksson et al. 2018; Loth et al. 2016; Messinger et al. 2013; Ozonoff et al. 2015). Debido a las limitaciones y beneficios ampliamente discutidos de ambos enfoques (Marschik and Einspieler 2011; Ozonoff et al. 2011; Palomo et al. 2006), los métodos prospectivos no podrán reemplazar completamente a los retrospectivos. Especialmente cuando se estudian enfermedades y desórdenes raros tales como RTT, principalmente causados por mutaciones de novo en el gen MECP2, para el cual obtener poblaciones de alto riesgo es poco realista (Marschik et al. 2018), los procedimientos retrospectivos son aún de gran valor.

Conclusión

Los trabajadores de guarderías infantiles no son profesionales de la salud entrenados y no es su responsabilidad identificar niños con desarrollo atípico. En cambio, con su interacción intensiva diaria con niños pequeños de diferentes perfiles de desarrollo, podrían estar entre los primeros en notar desarrollo aberrante en niños muy pequeños. Nuestro sistema de salud podría beneficiarse mucho de los trabajadores de guarderías designando miembros del personal con entrenamiento avanzado y experiencia en cada institución de cuidado infantil, lo que facilitaría la vigilancia diaria del desarrollo y podría ayudar a los profesionales de la salud a identificar a menor edad desórdenes del desarrollo que de otra forma son identificados tarde.

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ORIGINAL PAPER

Identifying Atypical Development: A Role of Day-Care Workers?

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Abstract

Identifying the early signs of developmental disability is important for ensuring timely diagnosis and early intervention. Day-care workers may be in a prime position to notice potential developmental deviations, but it is unclear if they can accurately recognize subtle early signs of atypical development. Sixty day-care workers examined home-videos of very young children with fragile X syndrome and typically developing children. Results indicated that most day-care workers can distinguish typical and atypical development in general and might therefore have an important role in early identification. Special work experience and advanced pedagogical training appeared to boost day-care workers' sensitivity to detect atypical features in early development and to provide effective daily surveillance.

Keywords Day-care workers · Developmental disability · Early identification · Retrospective video analysis · Fragile X syndrome

Scientific understanding and public awareness of young children at risk for developmental disorders have substantially increased in the past few decades. The growing number of studies on early human development have provided new empirical findings that have altered perspectives on the age-specific phenomena, pathways, and profiles associated with several developmental and genetic disorders (e.g., Johnson et al. 2015; Marschik et al. 2013; Messinger et al. 2013; Thomas et al. 2009). The advancements related to studies on late detected developmental disorders (LDDDs), for example, are particularly evident in research on autism spectrum disorder (ASD; e.g., Bölte et al. 2016, 2013; Bontinck et al. 2018; Bussu et al. 2018; Gliga et al. 2014; Messinger et al. 2013; Roeyers 2018). Research advances have also contributed to what appears to be greater acknowledgment of the significance of earlier detection of atypical development to enable timely intervention and support.

In many European countries, as the number of working-mothers keeps climbing, the need of public childcare services increases steadily. In Austria, for example, the proportion of infants and children under 2 years of age who are attending day-care centres doubled from 2007 to 2017. In some urban areas, up to 45% of children aged 0 to 2-years attend day-care, of whom more than 90% spend 6–10 h per day with professional caregivers (Statistik Austria 2018). These day-care workers could thus be seen as among the most important care persons for many young children. As a consequence, it would seem important to investigate the potential role of day-care workers in the early detection of

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deviant development in young children. For example, might it be feasible to enlist day-care workers in the process of screening children for ASD? Such issues have been highlighted in a number of recent publications (Branson et al. 2008; Dereu et al. 2012; Janus et al. 2018; Janvier et al. 2016; Larsen et al. 2018a, b; Nordahl-Hansen et al. 2018, 2013).

Indeed, results from some studies suggest that day-care workers have the competency to accurately report early signs of autism by applying well-designed behavioural checklists (Dereu et al. 2010; Larsen et al. 2018b). There might also be some potential advantages in integrating the help of day-care workers because such personnel often have training and knowledge of early child development and have considerable hands-on experience in working with children of comparable ages and diverse developmental profiles. Given that developmental disabilities affect at least 7 to 9% of young children (Olusanya et al. 2018; Zablotsky et al. 2017), it may be important to involve day-care workers as potential screeners of developmental disability. Unlike parents, day-care workers are likely to be more objective. The input from their daily surveillance may complement to our understanding of the prodromal period of LDDDs and potentially contribute to earlier identification, hence be of important public and scientific interest.

Fragile X syndrome (FXS) is one of these LDDDs. Similar to several other disorders, such as Rett syndrome (RTT) and ASD, syndrome related behavioural and physical features are often subtle and elusive to detect at first, which of course makes early identification more challenging. Still, atypical early signs emerging in the first years of life across different developmental domains have been reported to be perceptible to parents of children with FXS (e.g., Hinton et al. 2013; Zhang et al. 2017). Some of these signs are frequently presented and often captured by home videos. With a benchmark procedure retrospectively analysing home videos of children with FXS, we, among other researchers, demonstrated that these signs can be readily identified and classified by professionals (Zhang et al. 2018; see also Baranek et al. 2005). Early phenotypes of FXS present a broad spectrum of atypical neurobehavioural features across various domains (e.g., motor, cognition, speech-language, social-communication), which are not specific to FXS (e.g., Haessler et al. 2016; Hagerman 2002; Kidd et al. 2014; Marschik et al. 2014; Raspa et al. 2017; Roche et al. 2018; Zhang et al. 2017). As early signs of FXS overlap with peculiarities of children with other developmental and genetic disorders, to identify these signs requires sensitivity to deviant development in general and not necessarily expertise in a specific syndrome.

Pertinent to the current study, we intended to utilize home-video footage showing behaviours of different developmental areas of typically developing children and children with FXS as material to tap on day-care workers' general awareness of early development. We adapted a video reviewing procedure used by Burford et al. (2003) by adding benchmark assessments of neurodevelopmental features. Specifically, we aimed to investigate whether day-care workers perceive young children with typical or atypical development differently. In Addition, we intended to find out whether they are able to accurately identify typical and atypical features in early development. As day-care workers have different training and experience backgrounds, we also aimed to answer whether these factors modify day-care workers' perceptions of early developmental phenomena.

Methods

Participants

Twelve day-care centres and one vocational school for kindergarten teachers in Graz, Austria and surrounding regions were contacted. Seven centres with 36 day-care workers (21 kindergarten teachers, one male; 15 carers, all female) and the school with 25 pre-service student teachers (all female) agreed to participate. Of the 21 teachers, 8 received additional advanced pedagogical or special-needs training. The professional training for kindergarten teachers in Austria is 5 years, and for carers 6 months. None of the 15 carers in this study had received additional training related to child-care. At the time of data collection, the mean age of teachers was 42 years (SD = 11.40), and that of the carers was 43 years (SD = 11.50). The 25 pre-service student teachers had recently finished the penultimate year of their training, and were on average 17 years of age (range 17–19). Their curriculum in the penultimate year includes 2 h supervised hands-on training per week at day care centres. One pre-service student teacher dropped out due to personal reasons, leaving the final sample with a total of 60 participants (21 teachers, 15 carers, 24 pre-service student teachers).

In addition to levels of training, we collected data of work experiences of the participants, including the overall length, and whether the participant had professional experience with children younger than 2 years of age and/or with a developmental disorder.

Materials

This Citizen Scientist project is linked to an umbrella project on early development of individuals with FXS (Zhang et al. 2017). For the current study, we extracted data from our real-world database GUARDIAN (Graz University Audiovisual Research Database for the Interdisciplinary Analysis of Neurodevelopment). We selected recordings of 13 children with

FXS and 7 typically developing children (TD), of whom videos of the first 24 months of life were available. All children with FXS had received a clinical and genetic diagnosis, and all the videos were recorded prior to diagnosis. All TD children were singletons, born at term with uneventful pregnancy and delivery. At the time of this manuscript preparation, all were older than 7 years of age, none has received a diagnosis of any developmental or genetic disorder, nor did any of their known family relatives. A total of 35 (rounded) hours recordings (25 h from the children with FXS), including footage of daily routines or special family gatherings, were first cut into clips by a research assistant. Clips were cut by settings, which were defined by changes of the child's position (e.g., in prone/supine, sitting in rocker, walking with support) and the presence/absence of the caregiver(s) in the scene; for technical details on clipping please see Zappella et al. (2015). Following the aims of this study, we initially selected 40 clips of the FXS corpus and 20 of the TD corpus covering different age bands of the first 2 years of life (1–6, 7–12, 13–18, and 19–24 months). The clips were chosen to present behaviours in different developmental areas (e.g., motor, social-communication) of the target child. After a pilot trial and intramural testing, we retained 15 clips to trim the experiment duration (please see data collection below), leaving 10 videos from children with FXS (FXS videos from here after) and 5 from TD children (TD videos from here after). These 5 TD videos were comparable to 5 of the 10 FXS videos in age and setting (Table 1). All clips had a length of 1 min (± 5 s).

The present study was approved by the Institutional Review Board of the Medical University of Graz, Austria (29-054 ex 16/17). Participants and parents who shared their data gave their informed written consent to participate in the study and to publication of the results.

Data Collection

Video reviewing took place for each participant individually in a separate quiet room at the kindergarten or school where the participant worked or was trained. The participants were given background information about the study and instructed to look for early developmental markers that might contribute to timely identification of late-diagnosed developmental disorders. After signing the informed written consent, the participants were asked to review the 15 video clips from young children between 8 and 23 months of age. They were also informed that some of the children were typically developing, and some were later diagnosed with a developmental disorder. The term "fragile X syndrome" was not mentioned. The participants were encouraged to comment on each video based on their experience with similar aged children while taking into account the different developmental domains. The order of presenting the 15 video clips was randomized across participants. Before playing each video, the participant was informed of the child's gender and age, but not whether the child had received a diagnosis. Each video was played twice on a computer, controlled by an experimenter. The participant was encouraged to take notes after the first run. After the second run, the participant was asked to comment on anything worthy of attention about the child, both positive notes and concerns. The participant was never requested to judge normal versus atypical development. The next clip was played when the participant was ready. Pauses were made when necessary, but not during the same clip. The review of all 15 clips took about 60 min for each participant. An experimenter guided the review procedure, took notes, and ran the computer program without engaging in any discussion on the participant's comments. On-screen audio recording was taken by Corel VideoStudio X9, using the "Screen Capture" function.

Transcription and Scoring

All 60 audio recordings were later transcribed verbatim in the iDN lab by the author I.K.T. About 15% of the audio materials were randomly selected and transcribed by another research assistant to check on the accuracy and consistency for the scoring of the transcriptions. No meaningful difference has been revealed. Thus, all scoring was solely based on the transcripts from I.K.T.

Three senior experienced team members of the research group iDN (D.Z., P.B.M., and C.E.) reviewed the 15 video clips without knowing the medical backgrounds of the children. They separately outlined all features worthy of attention for each target child, being particularly positive or age-advanced (e.g., coasting/sideward walking at 10 months of age) as well as atypical or age-inadequate (i.e., referring to delayed developmental milestones and/or abnormal behaviours). The features covered different neurofunctions of various developmental areas (e.g., motor, social, verbal, physical appearance, repetitive movements). Although typical and age-appropriate behaviours (e.g., sitting without support) were observed in the clips of children with FXS, none was pointed out as remarkable or age-advanced. Thus, no particularly positive or age-advanced feature was listed for any of the FXS videos. For TD videos, both positive and atypical features were listed. The three authors also named one or two "primary feature(s)" for each clip, referring to the most salient feature(s) observed in the scene. They then discussed their analyses and reached consensus on a benchmark list for each clip. Each list consists of at least three features besides the primary one. Multiple features were frequently identified in the same developmental area (e.g., motor, social) for one video. For two clips from children with FXS and one from a TD child, two primary features were identified (Table 1).

Table 1 Benchmark features for the FXS and TD videos

	Video code[a]	Age (months)	Areas with features on the benchmark list[b]							n/60
			Mot	**Phy**	**RM**	**S/C**				
						Fac	Int	Lan	Voc	
FXS videos	FXS1	8	++		+	+			+	17
	FXS2	9		+	+	++	+		+	13
	FXS3	9	++	+				+	+	15
	FXS4	10	++			++	+	+		7
	FXS5	10	+		++		+			9
	FXS6	12	++		++	+	+	+		6
	FXS7	12	+			++				26
	FXS8	13	+		+		++		+	6
	FXS9	16	+		+	++		+		7
	FXS10	23	+		+		+	++	+	21
TD videos	TD1	8	+		+		+	++		31
	TD2	9	+		++		+		+	48
	TD3	10	+						++	46
	TD4	16	++				+	++		9
	TD5	23					++	+		48

++This area consists of the primary feature defined for this video on the benchmark list. For three videos (FXS4, FXS6, and TD4), two primary features have been identified

+This area consists of non-primary features identified for this video

n/60: Number of participants regarded the child as normal

The benchmark list includes age-specific features (in alphabetical order) in four areas (in bold): **Mot** (motor): age-advanced/positive features (coasting/sideward walking, fine manipulation, standing up independently, good postural control), age-inadequate features (no antigravity movements, no manipulation, not sitting without support, not standing-up free, not walking free, tapping objects without grasping), and in addition, slumped posture/hypotonia, long-lasting tongue protrusion; **Phy** (physical appearance): oversized ears, strabismus; **RM** (repetitive movements): body rocking, hand/arm flapping, hand opening and closing, head shaking; **S/C** (social/communication) with (a) Fac (facial expression): awkward smile, crying-like facial expression during pleasure vocalizations, empty gaze, non-adaptive facial expression, sluggish facial expression; (b) Int (interaction): positive features (attentive and patient, engaged in play, interactive and responsive) and negative features (exaggerated reaction, lack of adaptive reaction in social play, no response when spoken to, passive/no initiative in social play, roughness with other children); (c) Lan (language): age-inadequate utterances, neologisms; (d) Voc (vocalizations): expiratory and inspiratory vocalizations, high-pitched vocalizations, hoarse voice, monotonous/repetitive vocalizations, echolalia, pressed voice, unmodulated vocalizations

[a]Videos from children with neurotypical outcome, TD1 to TD5, were comparable to FXS1, FXS2, FXS4, FXS9, and FXS10, respectively, in age and setting. Videos are ordered by ascending age (completed months)

[b]Multiple features were frequently identified in the same area for a video

To assess the comments of the participants, we implemented a scoring scheme adapting the optimality concept introduced by Prechtl (1980) and the inductive corpus analytical methodology of Lindseth and Norberg (2004). Higher scores indicate greater congruity with the benchmark list. Each comment made on a clip was assigned a score between 0 and 7; details on scoring are given in Table 2.

In the next step, D.Z. and I.K.T. separately assessed all 60 transcripts. Intraclass correlation coefficient (ICC) between the scorers was .94 (95% CI, .90–.97), based on the model of average measures, absolute-agreement, and two-way mixed-effects. Discrepancies were then discussed until consensus was reached. In addition, we counted for each video the number of participants who commented that the target child was normal or expressed no concern about the child's development (Table 1).

Data Analysis

Data were analysed with SPSS, version 25 (SPSS Inc, Chicago, IL). Mann–Whitney U test was run to compare the difference of two independent groups if normal distributions could not be assumed. *T* test was used to compare the difference between two means of normal distributions. Pearson correlation coefficient (r) was calculated to estimate the association of two continuous variables with normal

Table 2 Scoring scheme for FXS and TD videos

Scale for comments on FXS videos		Scale for comments on TD videos	
Raw score	Definition	Raw score	Definition
7	Correct identification of 3 or more features including the primary feature	7	Correct identification of 2 or more features including the primary feature
6	Correct identification of 2 features including the primary feature	6	Correct identification of the primary feature or multiple none-primary features
5	Correct identification of the primary feature or multiple none-primary features	5	Correct identification of a none-primary feature
4	Correct identification of a none-primary feature	4	No identification of any relevant feature, but making feature-related comments
3	No identification of any feature, but making feature-related comments[a]	3	Finds nothing worthy of attention
2	No irrelevant remarks[b]	2	No irrelevant remarks
1	No opposite interpretation of the features[c]	1	No opposite interpretation of the features
0	No concern	0	Only opposite interpretations of the features

Opposite interpretation leads to point deduction. For example, if a participant correctly identified the primary feature of an infant with FXS, yet referred to the child's another atypical feature as normal (i.e., opposite interpretation), the score for this clip would be 4 points (5 points, 1 point deduction)

[a]For example, a feature on the benchmark list of a video was "empty gaze". A participant commented "He looks somehow strange to me. I wonder if he could see at all? Maybe he is blind?"

[b]The participant makes comments traceable to the scene, but fails to identify any feature on the benchmark list

[c]Opposite interpretation: an obviously atypical behaviour seen as normal, or vice versa

distributions. Spearman rank order correlation (rho) was applied to estimate the association between two variables if normal distributions could not be assumed for both. ANOVA was run to compare means among three or more independent groups. Eta squared (η^2) was chosen to report the effect size of U test and ANOVA. Cohen's d was reported for the effect size for t-test. Alpha was set to .05, two-tailed for all analyses.

Results

Normal Versus Atypical Development?

More participants commented that the target child was developing normally for TD videos (Md = 46 out of 60, range 9–48; Table 1) compared to FXS videos (Md = 11, range 6–26), Mann–Whitney U = 5.50, Z = 2.40, p = .013, with large effect size η^2 = .38. For all but one TD video, more than half of the participants spontaneously remarked that the target child was normally developing. This video showed a child of 16 months of age, who did not produce linguistic vocalizations, nor walked or stood up independently on the clip.

Consistency Between Participants and the Benchmark Analysis

Initial analysis of the mean raw scores of the 5 TD videos as well as the 10 FXS videos revealed normal distributions. The mean raw score for the TD videos across participants was 4.04 (SD = 1.24) and 3.13 (SD = .93) for the FXS videos. The standard mean raw score of TD (Z_{TD}) and FXS videos (Z_{FXS}) was significantly correlated, r = .43, p = .001. That is, participants who scored high (or low) on the TD videos were also likely to do so on the FXS videos. The overall score of a participant is the standard sum of Z_{TD} and Z_{FXS}, i.e., overall score = Z [Z_{TD} + Z_{FXS}]. The overall score ranged from − 2.23 to 2.26 across participants. A higher overall score of a participant indicates higher consistency with the benchmark analysis of the videos. We examined the following variables:

Training Status

The overall scores differed significantly among teachers with or without additional pedagogical or special-needs training (st; teachers^{st+} and teachers^{st-} from here after), carers, and pre-service student teachers, $F_{(3, 56)} = 5.45$, p = .002, η^2 = .23. A Tukey posthoc comparison revealed significantly higher overall scores for teachers^{st+} (M = 1.15, SD = .96) compared to teachers^{st-} (M = .05,

SD = .92, d = 1.17, p = .043), to carers (M = − .18, SD = .99, d = 1.36, p = .008), and to pre-service student teachers (M = − .30, SD = .82, d = 1.62, p = .001). No statistically significant difference of the overall score between any of the two groups was found among teachers^{st-}, carers, or pre-service student teachers.

Work Experience

The pre-service student teachers were excluded from this analysis as they had not yet started to work on a daily basis. The length of work experience of the 34 day-care workers[1] was not normally distributed. The median was 7.5 years (range 2–34). Of the 20 teachers, the median was 7.5 years (range 2–34); of the 14 carers, it was 8.0 years (range 2–19). The difference was not significant, Mann–Whitney U = 136.00, Z = .141, p = .888. The overall scores of the day-care workers correlated moderately with the length of their work experience, Spearman's rho = .34 (p = .052). No significant difference of the overall score was found between participants with longer (i.e., length of experience above median, n = 16; overall score M = .51, SD = .96) or shorter (n = 18, overall score M = .08, SD = 1.03) work experiences, Student's independent sample t-test yielded t = 1.26, p = .218.

Task Relevant Experience

Task relevant experience requires: (a) a reasonable amount of general work experience as a day-care worker (i.e. length of work experience being at least 5 years), and, (b) has professional experience working with young children (i.e., under 2-year-old) and/or (c) children with a developmental disorder. Twelve teachers and 6 carers met the above conditions and were classified as the group as having "rich" task relevant experience, including all of the 8 teachers^{st+}. The mean overall scores of the "rich" group was .68 (SD = .94). The remaining 8 teachers and 8 carers who had never worked with young children nor children with a developmental disorder, or, who had less than 5 years of professional experience as a day-care worker were taken into the "limited" task relevant experience group. The mean of their overall score was -.16 (SD = .92). The difference of the overall score between the "rich" and "limited" groups was significant, t = 2.65, p = .012, d = .90. That is, participants with rich task relevant experiences had higher scores. Further analysis revealed that teachers with rich task relevant experience (M = .98, SD = .89) scored higher than teachers with limited task relevant experience (M = − .21, SD = .95), t = 2.86, d = 1.29, p = .010. Scores of the carers with rich (M = .08, SD = .75) or limited (M = − .11, SD = .95) task relevant experience, however, were not significantly different, t = .41, p = .688. In other words, task relevant experience influenced the scores of the teachers, but not the carers.

Discussion

The present study, based on our previous research and methodologically similar studies (Burford et al. 2003; Dereu et al. 2012; Marschik et al. 2012; Zhang et al. 2017, 2018), presents a novel approach for investigating day-care workers' awareness on early development and the prodromal phase of late recognized developmental disorders. We tapped on the participants' views and knowledge of early developmental milestones and qualitative deviances from normal development during the first 2 years of life. Although participants were not explicitly asked to differentiate between normal and atypical development, their comments indicated that they clearly perceived differences between TD and children with deviant development, FXS in this case. Significantly less concerns but more spontaneous confirmation of normality were addressed to TD compared to children with FXS. That is, normal or aberrant development in general was perceptible to the day-care workers.

This finding is similar to those of Marschik et al. (2012) in a study that focused on acoustic Gestalt perception. They found that both speech-language pathologists and non-professionals were able to distinguish typical vocalizations from atypical vocalizations expressed by young children. In the current study, most of the participants noticed that *something* was wrong when they were observing a child who was later diagnosed with FXS, but when it came to specifying exactly *what* was wrong, that was where differences among the different participant groups emerged. Specifically, observations from teachers^{st+} (i.e., with additional specialized trainings in atypical development) were most consistent with the benchmark assessments of developmental scientists. Teachers^{st-} (i.e. without additional training) performed less well and comparable to the pre-service student teachers in the penultimate year of training, and, again similar to the carers (i.e., with much shorter professional training). This, not surprisingly, suggests that specialized training beyond the general childcare preparation may underpin the sensitivity to recognize typical and atypical features in early development (see also Daniels and Mandell 2014; Lee et al. 2005; Reilly et al. 2015).

Given these findings, it leaves open the question concerning what, if any, role does work experience play? Interestingly, teachers and carers who had worked longer in day care units did not score significantly better than those with

[1] For one of the 21 teachers and one of the 15 carers, data of the length of work experience was missing, leaving 20 teachers and 14 carers for the comparison.

shorter work experience. This was echoed by the findings that teachers^{st-} and the pre-service student teachers performed rather similarly, both having comparable level of training, while the teachers having practiced longer. In other words, the duration of experience did not prove to be critical to developing "sensitive eyes" for assessing early developmental phenomena. Rather, the *profile* of experience seemed to play a vital role in this respect. In our current study, it was not childcare experience per se that mattered, but rather having "task-relevant experience", that is, having practical experience with certain groups of children (e.g., children younger than 2 years and/or children with a developmental disability) that was associated with higher scores. Still, it must be pointed out that the task-relevant experience appeared to have an impact only if a solid training basis was already present. That is, only the teachers, but not the carers, seemed to have benefited from the task-relevant experience in that they performed better compared to their counterparts with less experience. This suggests that without sufficient general training, accumulating experience alone might not actually induce better insight into children's early development.

Incidentally, yet not unexpectedly, all 8 teachers^{st+} fell into the group of 16 participants with rich task-relevant experience. These eight teachers who pursued advanced trainings had also accordantly practiced their know-how in their daily work. Both specialized training and related practices may therefore have accounted for their performance in recognizing typical versus atypical features in early development. For day-care workers who intensively interact with children of different ages and developmental profiles day in and day out, to notice that *something* may be wrong with a child's development might be unchallenging. However, in order to more precisely specify developmental anomalies that may be indicative of a developmental disability, specialized training and rich experiences seem to be critical factors.

Notably, the raw scores assessed according to the scoring scales (Table 2) indicated that many of the teachers^{st-}, the carers, and the pre-service student teachers failed to recognize any specific feature listed by the benchmark assessment for most video sequences (i.e., having scored on average lower than 4 for the FXS videos and lower than 5 for the TD videos). The features which were salient to the professionals who set the benchmarks appeared to have escaped the eyes of many of the participants. This finding would seem to call into question the reliability of asking day-care workers to detect specific warning signs of a possible developmental disability in very young children. Similarly, studies focusing on older children revealed that teachers' agreement with other informants on reporting developmental phenomena varied across different contexts (Ehlers et al. 1999; Macari et al. 2018; Mattila et al. 2009; Nordahl-Hansen et al. 2013, 2014; Posserud et al. 2006). In contrast, a previous study demonstrated that specialist nurses with extensive experience in home and hospital surveillance of infants were well able to detect early signs of developmental disability (i.e., RTT) and distinguish the infants from typical developing controls (Burford et al. 2003). This highlights the potential significance of qualification both in training and practice. The findings are also in line with our present study in that those day-care workers with advanced training and experience were better able to identify early typical and atypical developmental features. Another study on a large cohort by Dereu et al. (2010) used a specially developed checklist for childcare workers to screen children at risk for ASD. After a targeted short training highlighting early signs of ASD and how to use the checklist, the childcare workers were proven to be reliable and sensitive informants of early signs of ASD. This and our present study together suggest that day-care workers have the potential to distinguish typical and atypical development in general. To gain most from day-care workers' input to support early identification of atypical development, using specifically designed checklist of signs and providing related training may be effective and desirable.

We are well aware that our study brings along some limitations. The experimental design using brief video sequences does not fully resemble natural settings, where day-care workers may be able to observe each child across a range of contexts over days, weeks, and months. Moreover, some of our participants might not have felt comfortable in an experimental situation, which could have negatively influenced their performance. Nonetheless, reviewing videos provides a sound approximation of the real world setting and can deliver valuable information within a reasonable time. As all the participants had the same materials to assess, distinctions among groups might be attributed to their different sensitivities to various developmental features. It is rather unlikely that simply providing more extensive and relaxed observational settings will lead to significantly better insights into normalities and peculiarities in early development. In their daily work field, day-care workers might be reluctant to alarm parents or healthcare professionals about atypical development of a child due to ethical and personal reasons, even if they would have noticed adverse signs. Thus, researchers and healthcare professionals need to actively collect data in an efficient way from qualified day-care workers if they intend to profit from additional critical information which they cannot gain from parents (see also Garcia-Primo et al. 2014; Macari et al. 2018). Also, the small sample size is another limitation related to the present work.

Last but not least, investigations into the early signs of LDDDs have mostly been done with retrospective approaches, including utilizing questionnaires and checklists, as well as analyses of home videos. This has been gradually overtaken by prospective approaches for a number of disorders, ASD being at the forefront (e.g., Bölte et al.

2013; Isaksson et al. 2018; Loth et al. 2016; Messinger et al. 2013; Ozonoff et al. 2015). Due to well-discussed limitations as well as benefits of both approaches (e.g., Marschik and Einspieler 2011; Ozonoff et al. 2011; Palomo et al. 2006), the prospective methods will unlikely fully replace the retrospective ones. Especially when studying rare diseases and disorders such as RTT, mainly caused by de novo mutations of the *MECP2* gene, for which obtaining high-risk samples is unrealistic (Marschik et al. 2018), retrospective procedures are still of great value.

Conclusion

Day-care workers are not trained healthcare professionals and it is not their responsibility to identify children with an atypical development. Rather, with their intensive daily interaction with young children of different developmental profiles, they might be among the first to notice aberrant development in very young children. Our healthcare system might benefit most from day-care workers by allocating staff members with advanced training and experience in every childcare institution, who will facilitate daily developmental surveillance and may help healthcare professionals to identify otherwise late recognized developmental disorders at a younger age.

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Compliance with Ethical Standards

Conflict of interest The authors declared no conflicts of interest with respect to the research, authorship, and/or publication of this article.

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