

THEORY/REVIEW MANUSCRIPT

Joint Attention in the Context of Hearing Loss: A Meta-Analysis and Narrative Synthesis

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Abstract

Joint attention is important for children's language development. We report two meta-analyses that demonstrate that the congruency in hearing status between parent and child affects the establishment and maintenance of joint attention. Dyads consisting of hearing parents and children with hearing loss, achieve fewer and briefer moments of joint attention in comparison to dyads of hearing parents and hearing children and dyads of deaf parents and deaf children. The theoretical and practical implications of these differences are discussed and placed in the context of two narrative syntheses. The first one focusing on parental strategies used to achieve and maintain moments of joint attention and the second one on the relation between joint attention and spoken language proficiency. We also expect that this review may serve as the start of quest towards a more detailed description (taxonomy) and operationalization of joint attention in the context of hearing loss.

Introduction

Parents play an important role in creating an optimal language learning environment for their child (Gogate et al., 2000). Joint attention, that is, shared focus between parent and child, is an important feature of this learning environment. Once joint attention is established, parent and child have a communicative context in which linguistic information about events or objects in the environment can effectively be exchanged. In typically developing hearing children associations between several aspects of joint attention, such as mutual gaze and object looking, and later vocabulary sizes have been reported (e.g., Carpenter et al., 1998; Morales et al., 2000; Abney et al., 2020). For typically developing hearing children with hearing parents,

moments of joint attention are often characterized by simultaneous and coordinated exchanges of auditory and visual information (Gogate et al., 2000; Chen et al., 2019a). The coupling between children's sustained attention to objects and parents naming the object is considered a key factor in children's learning of words (Yu et al., 2018).

Children differ, however, in what constitutes their optimal word learning environment. While hearing children may benefit from the synchronous presentation of auditory and visual information, the situation may be different for children with hearing loss. It is likely that these children benefit from a different combination of multimodal cues (e.g., visual and tactile cues) or that they learn words more easily when information is provided

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in a sequential rather than synchronous way. These differences may not only affect the way in which parent and child exchange linguistic information during episodes of joint attention, it may also impact the establishment of joint attention (i.e., the number of successful joint attention episodes and/or the number of successful attempts to achieve joint attention).

Most children with hearing loss (~96%) grow up with hearing parents (Mitchell & Karchmer, 2004). The incongruity in hearing status between parent and child may impact the ease of establishment and maintenance of joint attention, because hearing parents may need to adapt their intrinsic ways of achieving and maintaining joint attention (Swisher, 1992). If this adaptation is less successful, hearing parents and their children with hearing loss may achieve fewer or briefer moments of joint attention. This will decrease children's access to the linguistic input and this may correlate negatively with the spoken language development of children with hearing loss (Dirks & Rieffe, 2019). Whether (and how) a congruity in hearing status between parent and child impacts the establishment and maintenance of joint attention as well as its relation to spoken language proficiency has been a topic of study over the past few years. To the best of our knowledge, the outcomes of these different studies have never been systematically reviewed or summarized. Therefore, the aim of the present review is to provide both a quantitative and narrative overview of the studies that investigated joint attention and its relation to spoken language proficiency in the context of hearing loss.

The Concept Joint Attention

Joint attention is not only considered important for language development, but also plays an important role in other areas of children's development. As such, joint attention has been studied from different disciplines and perspectives. Consequently, and because joint attention is not a single process but involves a cluster of different social skills, cognitive skills, and motivations of both social partner's (i.e., the parent and child; Carpenter et al., 1998), different views on what joint attention is and how it is achieved exist (Siposova & Carpenter, 2019). In the current review, we adopt a broad definition of joint attention and define it as a complex of social cognitive behaviors (including gaze, pointing, and visual attention) that lead to moments in time at which parent and child synchronously focus at the same object, action, event, or person (Akhtar & Gernsbacher, 2007). In our definition, we do not include any statement as to whether parents and children need to be aware of each other's attentional state (Chen et al., 2020). This is in contrast with the more "social" definition of joint attention in which intentionality and/or mutual awareness of the social partners' attentional state is a critical component (Gabouer & Bortfeld, 2021). In this context, it is also relevant to mention that in the present paper, we use terms like joint attention, joint engagement, and coordinated attention interchangeably.

The Development of Joint Attention

During the child's first 2 years of life, joint attention is thought to emerge gradually and incrementally in interaction with the child's emotional and social development, as well as resulting from a cognitive development involving skills like processing, attention and self-regulation (Morales et al., 2000). From very early on, already in their first 4 months of life, hearing infants start to engage with their hearing mothers in sustained periods

of face-to-face or mutual gaze (eye) contact (Johnson & de Haan, 2011, p.137). Between the ages of 9 and 12 months, children start to explore their environment. At that point, dyadic attention shifts to triadic attention in which parent and child start to coordinate and systematically divide their attention between objects or events in the environment and their social partner (Carpenter et al., 1998; Wille et al., 2019). It should be noted, however, that these episodes of joint attention do not occur frequently until children are about 15–18 months old (Carpenter et al., 1998). In general terms, deaf children follow a similar developmental pathway of joint attention as their typically developing hearing peers (Spencer et al., 1992).

Given its multicomponent nature, it is not always easy to tease apart what aspects of joint attention, that is, the individual skills of the child (e.g., gaze and gesture following, sustained attention) or the parent's motivations, responsiveness, and sensitivity predict the child's later social and language development. As recently reported by Abney et al. (2020), children's later vocabulary development depends on traits of both the parent and the child. In their study, both parent responsiveness and infant sustained attention when the child was 9 months of age were important aspects of joint attention that predicted vocabulary size of these children at 12 and 15 months of age. Children's ability to respond to bids of joint attention by their parent at 6, 8, 10, and 18 months of age has been reported to predict vocabulary size (measured with the MacArthur-Bates Communicative Development Inventories [NCDI; Fenson et al., 1993]) at 30 months of age, even when controlling for early language status (Morales et al., 2000). Interestingly, there is also some evidence suggesting that *responding* to joint attention may be predictive of receptive vocabulary size and that *initiating* joint attention may be predictive of expressive vocabulary size (Mundy & Gomes, 1998; Markus et al., 2001).

The Present Study

Given its important role for language development, joint attention has been studied in atypically developing children as well. For deaf children with hearing parents specifically, it has been hypothesized that differences in the accessibility of modalities through which parent and child perceive the world and communicate, impact the establishment and maintenance of joint attention. This may reduce children's access to linguistic input. Indeed, several studies do report fewer and/or briefer moments of joint attention in interactions between hearing parents and children with hearing loss than in interactions between deaf parents and deaf children or hearing parents and hearing children. Most of these studies are based on small sample sizes, however, and also the magnitude of the "joint attention disadvantage" is unknown. Using both quantitative and narrative analyses, the main aim of the present study is to provide an in-depth overview of these issues. More specifically, this review is set out to investigate the following four questions:

- (1) Do we observe quantitative differences between parent-child dyads with incongruity in hearing statuses (deaf/hard of hearing children with hearing parents: DH dyads) and parent-child dyads with congruity in hearing statuses (hearing children with hearing parents [HH dyads] or deaf/hard of hearing children with deaf parents [DD dyads]) in their establishment of joint attention (meta-analysis 1)?
- (2) Do we observe quantitative differences between parent-child dyads with incongruity in hearing statuses (DH

dyads) and congruency in hearing statuses in their maintenance of joint attention (HH dyads and DD dyads; meta-analysis 2)?

- (3) Do the potential quantitative differences as addressed with question 1 and question 2 relate to different strategies that parents use to achieve joint attention (narrative synthesis 1)?
- (4) Is there a relation between the establishment and/or maintenance of joint attention and spoken language proficiency in children with hearing loss (narrative synthesis 2)?

In addition to these four theoretical questions, this systematic review also provides an overview of the different definitions, operationalizations, settings, tasks, and participants in which joint attention has been studied in the context of hearing loss. How and whether such aspects impact cross-study comparisons on joint attention and the generalizability of the results is a topical debate currently held within the field (e.g., Siposova & Carpenter, 2019; MacGowan et al., 2020; Morales et al., 2000; Gabouer & Bortfeld, 2021).

Methods

We used the preferred reporting items for systematic reviews and meta-analysis statement to organize the meta-analyses and narrative syntheses (Moher et al., 2009). Effect size calculations and statistical analyses on the effect size measures were done in R (R Core Team, 2018). All data and scripts used for analyses are open available via our Open Science Framework (OSF) Project Page, accessible via: <https://osf.io/2qk6y/>.

Eligibility Criteria

Studies were eligible and included in the meta-analyses or narrative syntheses if they met all of the following criteria:

1. The study reports on an empirical measure of joint attention. Joint attention should be measured via videotaped (semi)structured or free play/interactional sessions. Note that studies using the still-face paradigm in very young infants are also included because eye-gaze in young infants is often seen as a precursor for joint attention.
2. The study involves parent-child dyads. This means that studies with other dyad groups (e.g., peer dyads, sibling dyads, researcher-child dyad, teacher-child dyad, or therapist-child dyad) were excluded.
3. The study must involve at least one participant group with unilateral or bilateral hearing loss.
4. The children are aged between 0 and 6 years (early intervention group). This age range was chosen given the importance of the early years of a child's life in terms of their development and the parent-child relation.

Additionally, studies are excluded if they met one (or multiple) of the following criteria:

1. The study solely based its conclusion on questionnaires.
2. The study is a case study (or a report of multiple case studies).
3. The study involves deaf or hard of hearing children with additional disabilities.

Information Sources and Search Strategy

Using a predefined search strategy (see our OSF-page), we conducted a first systematic search on the Web of Science in April 2020. This search yielded 133 potentially relevant unique articles. This search was followed by a second search using three additional information sources (PsycInfo, PubMed, and Scopus) in May 2020. This second search yielded 97 additional potentially relevant unique articles. Additionally, we received one record via personal communication with one of the authors of an included article (this article was published after our search period). In total, we ended up with 231 potentially relevant unique articles.

Study Selection Procedure

All 231 articles were first screened on their title and abstract by three of the authors of this review ([blinded for review]). If, by screening the title and/or abstract, it became clear that the study did not meet the eligibility criteria, the study was excluded. Based on this screening, 166 articles were excluded (initial inter-rater reliability: 90%, disagreements were discussed among the authors to reach consensus). For the remaining 65 articles, [blinded for review] read the methods and results sections carefully in order to decide whether or not the study met the eligibility criteria. After reading these 65 articles and discussing disagreements (37 articles) on inclusion judgment, 26 studies met the eligibility criteria and were included in our database for the meta-analyses and/or narrative syntheses. For a visual overview of the search procedure, see Figure 1. Note that studies had to report quantitative measures of joint attention success rate and/or duration in dyads with congruent hearing status and dyads with incongruent hearing status to be included in one of the meta-analyses (see the study selection spreadsheet our OSF-page for an overview of the decisions made during the study selection procedure).

Sample Description

The 26 papers (27 studies, one paper reports two studies) included in our database refer to 12 unique study samples. This means that there is overlap across the participants described in 14 of the studies included in this review. For one paper (Spencer et al., 1992), it is unclear whether or not the described data overlaps with data reported in other included papers. For more information, see Table 1.

Demographic Information

For the studies included, we extracted information on general demographic variables (country of study, race/ethnicity of included samples) and dyad characteristics (child age, parental education level, parent gender, child gender, child degree of hearing loss, CI-use, primary communication mode). In this section, we only describe and summarize those variables relevant to our Discussion. Detailed information regarding the other extracted variables can be found in our Supplementary Methods section and Supplementary Tables at our OSF-project page (Supplemental Table S1: general demographic information; Supplemental Table S2: parent and child characteristics).

The studies that were included in our database are conducted in six different countries, with the majority of studies conducted in the United States ($N=18$ studies, 11 unique samples). The remaining included studies have been conducted in Belgium

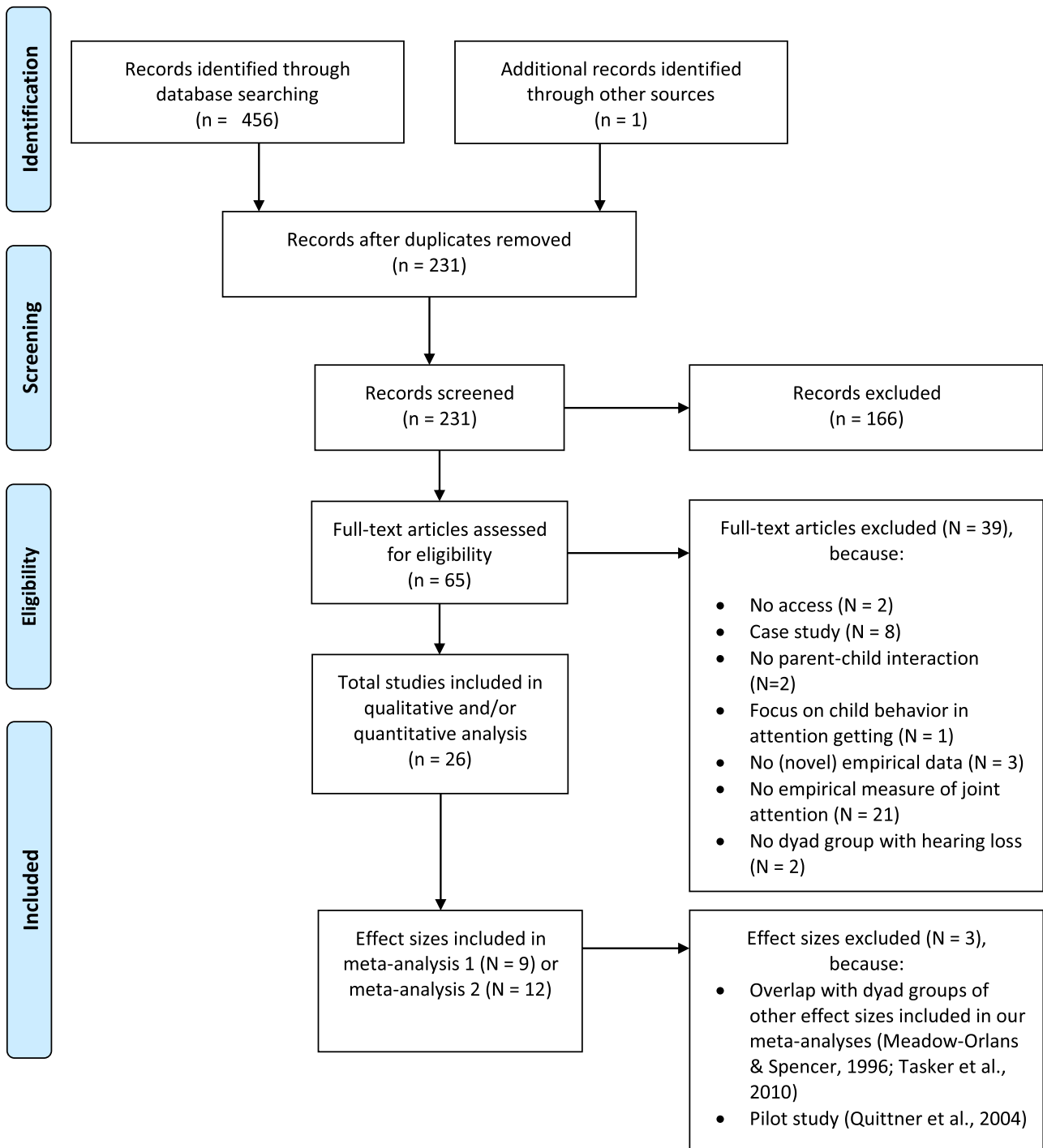


Figure 1. Flowchart indicating data inclusion and data exclusion at each stage of the literature search procedure.

($N=3$ studies, 2 unique samples), Canada ($N=2$ studies, 1 unique sample), the United Kingdom ($N=1$ studies), the Netherlands ($N=1$ study), and Sweden ($N=1$ study). One study includes participants from both the United Kingdom and Australia (Harris & Mohay, 1997). About 17 of the 27 studies provide information on the race/ethnicity of their participating dyads. In 6 of these 17 studies (five unique samples) the dyads are Caucasian. About 11 studies (six unique samples) report that their sample includes participants with Caucasian, Hispanic, African American, Asian

and Asian American race/ethnicity. Note, however, that for nine of these studies (five unique samples) with mixed samples, the samples are predominantly Caucasian (see Table 2 for an overview).

In the majority of studies, the dyads are mothers (rather than fathers) with their child. The mean percentage of participating mothers across 25 studies (two studies do not report parent gender) is 93%, with 18 studies (13 unique samples) in which the dyads consist of only mothers and their child. Only two studies

Table 1 General demographic information of the studies included in our final database on joint attention in children with hearing loss

Study	Included	Overlap study samples
[1] Barker et al., 2009	Narrative 2	[1][3]
[2] Wille et al., 2019	MA2 and narrative 1	—
[3] Cejas et al., 2014	MA2 and narrative 2	[1][3]
[4] Chen et al., 2020	MA1; MA2; and narrative 1	—
[5] Depowski et al., 2015	MA2	[5][7][8]
[6] Dirks and Rieffe, 2019	MA1; MA2; narrative 2	—
[7] Gabouer et al., 2018	Narrative 1	[5][7][8]
[8] Gabouer et al., 2020	MA1; Narrative 1	[5][7][8]
[9] Gale and Schick, 2009	MA1; MA2; narrative 2	—
[10] Harris and Chasin, 2005	Narrative 1	—
[11] Harris and Mohay, 1997	Narrative 1	—
[12] Koester, 2001	Narrative 1	[12][13][17] [25]
[13a] Koester and Lahti-Harper, 2010 part I	Narrative 1	[12][13][17] [25]
[13b] Koester and Lahti-Harper, 2010 part II		
[14] Lieberman et al., 2014	Narrative 1	—
[15] Loots and Devisé, 2003	Narrative 1	[15][16]
[16] Loots et al., 2005	MA1; MA2	[15][16]
[17] Meadow-Orlans and Spencer, 1996	MA2 ^a	[12][13][17] [25]
[18] Nowakowski et al., 2009	MA1	[18][24]
[19] Prendergast and McCollum, 1996	MA1	—
[20] Prezbindowski et al., 1998	MA2	—
[21] Quittner et al., 2004	MA2 ^a	—
[22] Roos et al., 2016	Narrative 1	—
[23] Spencer et al., 1992	Narrative 1	Unclear
[24] Tasker et al., 2010	MA1 ^a	[18][24]
[25] Waxman and Spencer, 1997	Narrative 1	[12][13][17] [25]
[26] Waxman et al., 1996	Narrative 1	—

^aOriginally, we had planned to include these studies in either meta-analysis 1 or 2, but eventually we had to exclude the studies because of overlap between the participating dyads of this study and another study included in the same meta-analysis (Tasker et al., 2010), the longitudinal nature of the study (Meadow-Orlans and Spencer, 1996) or because it was a pilot study (Quittner et al., 2004). HL = hearing loss; MA1 = meta-analysis 1; MA2 = meta-analysis 2.

(Loots & Devisé, 2003; Wille et al., 2019) explicitly compare differences in joint attention between fathers and mothers.

Other situations reported were semistructured play, face-to-face interaction, shared book reading and spontaneous interaction (Supplemental Table S3).

Study Characteristics

In this section, we provide a summary of the study characteristics that are relevant for our Discussion. For additional characteristics and an overview of the extracted information per study, we refer to the supplementary Methods section and Supplementary Tables at our OSF-project page (Supplemental Table S3: study characteristics).

The total number of dyads participating in the studies ranges from 8 to 276 dyads, with a median number of 26 dyads per dyad group per study. The median and range for each of the dyad combinations is: DD dyads, median = 8 dyads (range = 4–20); hearing children with deaf parents (HD dyads), median = 17 dyads (range = 6–20); DH dyads, median = 10 dyads ranging (range = 3–180); HH dyads, median = 10 dyads (range = 4–96).

Five of the 27 studies describe longitudinal data. Four of these five studies describe data of the Macturk et al. (1993) sample for which data were collected when children were 6, 9, 12, 15, and 18 months of age. The other longitudinal study is the study by Roos et al. (2016) who reports on data from children of 2 months to 18 months of age.

Almost all studies (17 studies, 12 unique samples) investigate parent-child interaction in a free object play session either at the child's home (9 studies, 8 unique samples), at the lab/clinic (12 studies, 6 unique samples) or a combination of both settings (4 studies, 3 samples; 2 studies did not report the study setting).

Definitions and Operationalizations of Joint Attention

Table S4 in the Supplementary tables section at our OSF-page provides an overview of the used definitions, terminologies, and operationalizations of joint attention among the included studies. Despite subtle differences most studies operationalize joint attention as a sequence of predefined events: (1) an episode of joint attention starts with a bid for attention by the parent or child; (2) The parent or child responds to this bid within a certain amount of time after the initiation has started (ranging from 3 to 15 s, with most studies requiring a response within 3 or 5 s); (3) the response results in shared interest for an object, event or each other; (4) the episode ends when the parent or child disengages from the shared state for a certain amount of time (ranging from 300 ms to 15 s).

We also have information on the instruments that researchers used to operationalize joint attention. The most frequently used instrument to code joint attention is Adamson's code for joint attention states. Four studies (three unique samples) developed their own coding scheme. Most studies, except the studies by Tasker et al. (2010) and Nowakowski et al. (2009; overlapping samples), use one code for all dyad groups. Tasker et al. (2010) and Nowakowski et al. (2009), however, set different response time criteria depending on the child's hearing status. Joint attention in their HH dyads is established if the social partner responds within 5 s of the other person's bid for attention and

Table 2 Numeric overview of demographic variables and study characteristics (total included studies = 27, unique studies samples = 12)

Variable	Number of studies (unique samples in brackets)	
Country	United States	18 (11)
	Belgium	3 (2)
	Canada	2 (1)
	United Kingdom	1 (1)
	Netherlands	1 (1)
	Sweden	1 (1)
	United Kingdom and Australia	1 (1)
Race/ethnicity	Caucasian	6 (5)
	Mixed (Hispanic, African American, Asian, Asian American, Caucasian)	2 (1)
	Mixed, but predominantly Caucasian	9 (5)
	Not reported	10 (9)
Parent gender	Mothers only	18 (13)
	Mixed, but predominantly mothers	5 (4)
	Mothers and fathers balanced	2 (2)
	Not reported	2 (2)
Study design	Cross sectional	22 (17)
	Longitudinal	5 (2)
Setting	Lab/clinic	12 (6)
	Home	9 (8)
	Combination of home, lab/clinic	4 (3)
	Not reported	2 (2)
Situation	Free (object) play session	17 (12)
	Semistructured play	4 (2)
	Face-to-face interaction	3 (2)
	Semistructured play (and free object play)	1 (1)
	Shared book reading (and free object play)	1 (1)
	Spontaneous interaction	1 (1)

terminates when the partner's attention is lost for more than 5 s. In DH dyads, the 5-s criterion is increased to 15 s (see Discussion).

Effect Size Calculation

For the studies that were included in one of the two meta-analyses, we first calculated, for each study, the effect size (Hedges' *g*) for the standardized mean difference (SMD) between dyads with congruency in hearing status and dyads with incongruency in hearing status. The effect sizes were calculated with the mean number of successful joint attention episodes/mean success rate (meta-analysis 1) or mean time spend in joint attention/mean duration of joint attention episodes (meta-analysis 2) and the corresponding standard deviation scores for the dyad groups, using the *mes2* function in the R *compute.es* package (Del Re, 2013). The effect size is calculated so that a negative value indicates that dyads with incongruency in hearing status reach fewer moments of joint attention (meta-analysis 1) or spend less time in joint attention (meta-analyses 2) than dyads with congruency in hearing status.

Results

In what follows, we present the results relating to the four main questions addressed in this paper. The first two questions assess

whether quantitative differences exist in (1) the establishment or (2) the maintenance of joint between parent-child dyads with incongruency in hearing status and parent-child dyads with congruency in hearing status. These two questions are addressed using quantitative syntheses (meta-analyses). The third question assesses whether these potential quantitative differences in the establishment or maintenance of joint attention relate to different strategies that parents use to achieve and maintain joint attention. The fourth question assesses the relation between the establishment and maintenance of joint attention and spoken language proficiency in children with hearing loss. These third and fourth questions are addressed using narrative syntheses (see Holzinger et al., 2020 for a similar distinction between quantitative and narrative syntheses).

Meta-Analysis 1: Establishment of Joint Attention

The first meta-analysis addresses the difference in the total number of established joint attention episodes and the success rate of parent-initiated bids for joint attention¹ between dyads with incongruency in hearing status (DH dyads) and dyads with congruency in hearing status (HH dyads and DD dyads). The meta-analysis includes nine effect sizes (from seven studies, see Figure 1) that came from 104 DH dyads (incongruent hearing status), 21 DD dyads, and 76 HH dyads (congruent hearing status). The ages of the children covered by the effect sizes in this

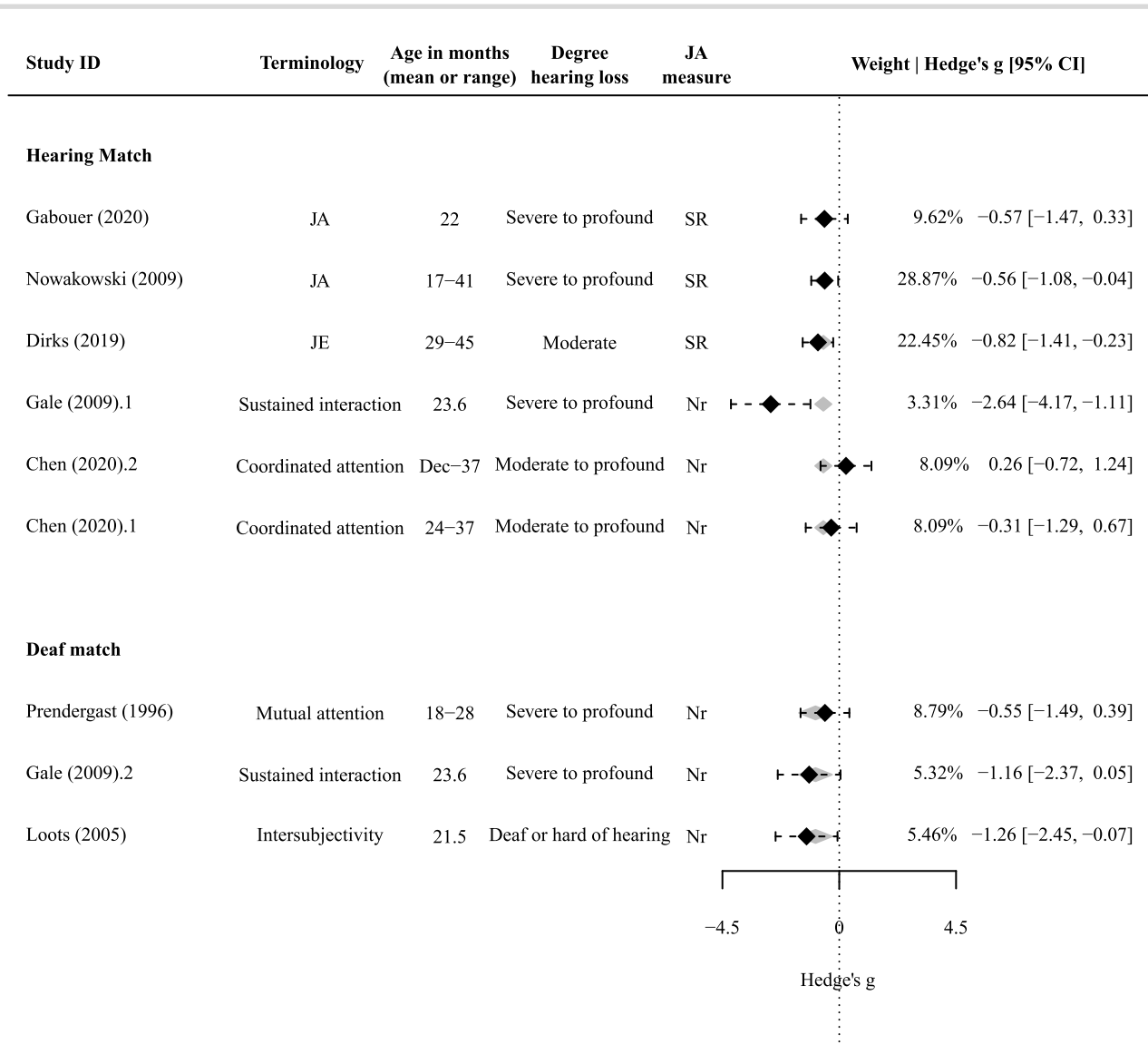


Figure 2. Forest plot showing overall and individual average effect sizes (Hedge's *g*) and 95% confidence intervals. A negative effect indicates that the incongruent dyads were less successful in their establishment of joint attention. JA = joint attention; JE = joint engagement; SR = success rate; Nr = number of successful established episodes.

meta-analysis ranged from 12 to 45 months and the degree of children's hearing loss ranged from moderate to profound (see [Supplemental Table S2](#) at our OSF-page).

To investigate our first research question we ran a hierarchical meta-analytic random effects model (using the *rma.mv* function from the *metaphor* package, Version 2.0 in R, [Viechtbauer, 2010](#)) in which the SMD was fitted as a function of the binary moderator Control Group, with DD dyads coded as $-\frac{1}{2}$ and with HH dyads coded as $+\frac{1}{2}$. The random-effects structure contained a random intercept for Study ($N=7$), by DH sample ($N=7$)² by observation/effect size ($N=9$). The model estimate of the overall weighted SMD (intercept) was -0.76 ($SE=0.18$, $95\% \text{ CI}=[-1.11, -0.41]$) and statistically significantly different from zero ($z=-4.26$; $p=2.1 \cdot 10^{-5}$), which indicates that dyads with incongruency in hearing status, on average, had a lower number or lower success rate of joint attention episodes as compared to dyads with congruency in hearing status ([Figure 2](#)). The confidence interval ranged from -1.11 to -0.41 indicating

a large to medium difference (in terms of Cohen's *d* effect sizes [Cohen, 1988](#)).

We also explored whether the standardized mean differed when DH dyads are compared to HH dyads ($N=6$) or DD dyads ($N=3$). The model estimated that this difference is smaller for comparisons between DH dyads and HH dyads than for comparisons between DH dyads and DD dyads. However, the estimate was not statistically significantly different from zero ($SMD=+0.30$; $SE=0.36$; $z=0.84$; $p=0.40$; $95\% \text{ CI}[-0.40, +1.0]$).

We also assessed the presence of a publication bias (i.e., tendency of a higher publication rate for studies with statistically significant results than studies with statistically nonsignificant results) via funnel plot asymmetry ([Egger et al., 1997](#)). No evidence was found for or against funnel plot asymmetry (*regtest* function in the *metaphor* package, $z=-1.1$, $p=0.27$) in our sample. Also, a visual inspection of the funnel plot ([Figure 3](#)) did not direct towards a publication bias.

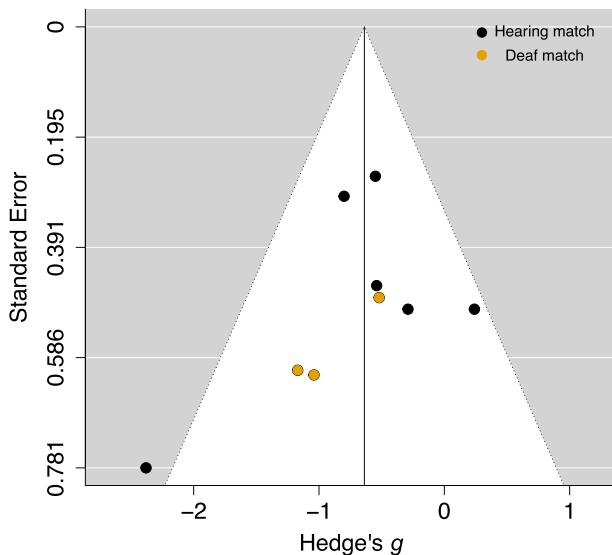


Figure 3. Funnel plot showing standard error of the Hedge's g effect size as a function of the effect size (studies on the establishment of joint attention). The vertical line indicates the mean weighted effect size. Dots in black are individual effect sizes from studies that compare DH dyads to HH dyads, dots in yellow represent individual effect sizes from studies that compare DH dyads to DD dyads. The triangle-shaped unshaded region represents a pseudo confidence interval region with Bounds equal to ± 1.96 SE.

As a final note, we would like to add that because of the overall low number of studies that investigated the establishment of joint attention in the context of hearing loss, we decided to combine the “success rate measure” and “total number of joint attention episodes measure” in one model. We realize that there are conceptual differences between these two measures. For example, success rate is a proportion measure including both failed and successful bids for joint attention, whereas the measure of total number of joint attention episodes only counts successful moments. Relatedly, success rate implies that joint attention episodes are the result of a bid for joint attention by one of the dyad members, this bid for attention is not implied in the total number of joint attention episodes measure (e.g., in these cases joint attention may have also occurred without an explicit bid). We did not further explore whether this difference in outcome measure moderates the overall outcome of this first meta-analysis, because Cochran's Q -test for the total amount of variance between the studies was not significant ($Q(7) = 11.74$, $p = 0.11$), meaning that we have no evidence that the true effect sizes do differ (or do not differ) between the studies in our sample.

Meta-Analysis 2: Maintenance of Joint Attention

The second set of results addresses differences related to the time spend in joint attention between dyads with incongruency in hearing status (DH dyads) and dyads with congruency in hearing status (DD dyads and HH dyads).

This second meta-analysis included 12 effect sizes (from eight studies) that came from 274 DH dyads (incongruent hearing status), 17 DD dyads, and 162 HH dyads (congruent hearing status). The ages of the children covered by the effect sizes in this meta-analysis ranged from 12 to 45 months and the degree of children's hearing loss ranged from moderate to profound (see Supplemental Table S2 at our OSF-page).

To answer the second research question, we ran a hierarchical meta-analytic random effects model in which we fitted the SMD as a function of the binary moderator control group, with DD dyads coded as $-\frac{1}{2}$ and with HH dyads coded as $+\frac{1}{2}$. The random-effects structure contained a random intercept for study ($N = 8$), by DH sample ($N = 10$)³ by observation/effect size ($N = 12$). The model estimate of the overall weighted SMD (intercept) was -0.44 ($SE = 0.20$, $95\% \text{ CI} = [-0.83, -0.053]$) and statistically significantly different from zero ($z = -2.23$; $p = 0.026$), which indicates that DH dyads with incongruency in hearing status, on average, spend less time in episodes of joint attention as compared to dyads with congruency in hearing status (Figure 4). The confidence interval ranges from -0.83 to -0.053 indicating a large to small difference (in terms of Cohen's d effect sizes; Cohen, 1988).

Our exploratory model comparison estimated that the difference in maintenance of joint attention is larger for comparisons between DH dyads and HH dyads ($N = 9$) than for comparisons between DH dyads and DD dyads ($N = 3$). However, the estimate was not statistically significantly different from zero (SMD = -0.68 ; $SE = 0.39$; $z = -1.72$; $p = 0.086$; $95\% \text{ CI} [-1.45, +0.096]$).

Cochran's Q -test for the total amount of variance between the studies (heterogeneity) was not significant ($Q(10) = 16.03$, $p = 0.10$), meaning that we have no evidence that the true effect sizes differ (or do not differ) between the studies in our sample. Also, we found no evidence for or against funnel plot asymmetry (Figure 5 and *regtest* function in the *metafor* package, $z = 1.03$, $p = 0.30$) in our sample (Egger et al., 1997).

Narrative Synthesis 1: Strategies to Establish or Direct (Joint) Attention

Another frequently addressed topic in studies on joint attention in children with hearing loss is *what strategies* lead to episodes of joint attention. In discussing these strategies, most studies distinguish between visual (e.g., sign, entering the child's visual space, wait until the child looks), tactile (e.g., tap on the child's body or floor behind the child), visual/tactile (e.g., tap and sign) and auditory/oral strategies used by parents to achieve their child's attention.

Most studies report that deaf parents of deaf children (Waxman et al., 1996; Koester 2001; Loots & Devisé, 2003; Koester & Lahti-Harper, 2010; Roos et al., 2016; Wille et al., 2019) and deaf parents of hearing children (Koester, 2001; Loots & Devisé, 2003; Gabouer et al., 2018) use more tactile and visual-tactile strategies to gain their child's attention than hearing parents of both deaf and hearing children⁴. Roos et al. (2016), for example reports that “Deaf parents with deaf children (2 to 18 months) are very visually oriented [...], not only when they are signing” (Roos et al., 2016, p. 17). Hearing parents on the other hand, are more likely to use oral or auditory strategies to establish moments of joint attention with their deaf child (Wille et al., 2019) or to re-establish eye contact with their deaf or hearing child (Koester, 2001). The longitudinal data of Koester and Lahti-Harper (2010) suggests that differences in maternal strategies to re-establish eye contact with their child are consistent over development (9, 12, and 18 months of age). They add to their conclusions that deaf mothers of hearing children use more auditory strategies during free play interactions as compared to deaf mothers of deaf children. Lieberman et al. (2014) investigated what strategies deaf parents of deaf children use to ensure that their signs were visible to their deaf children, either in a play setting or in a

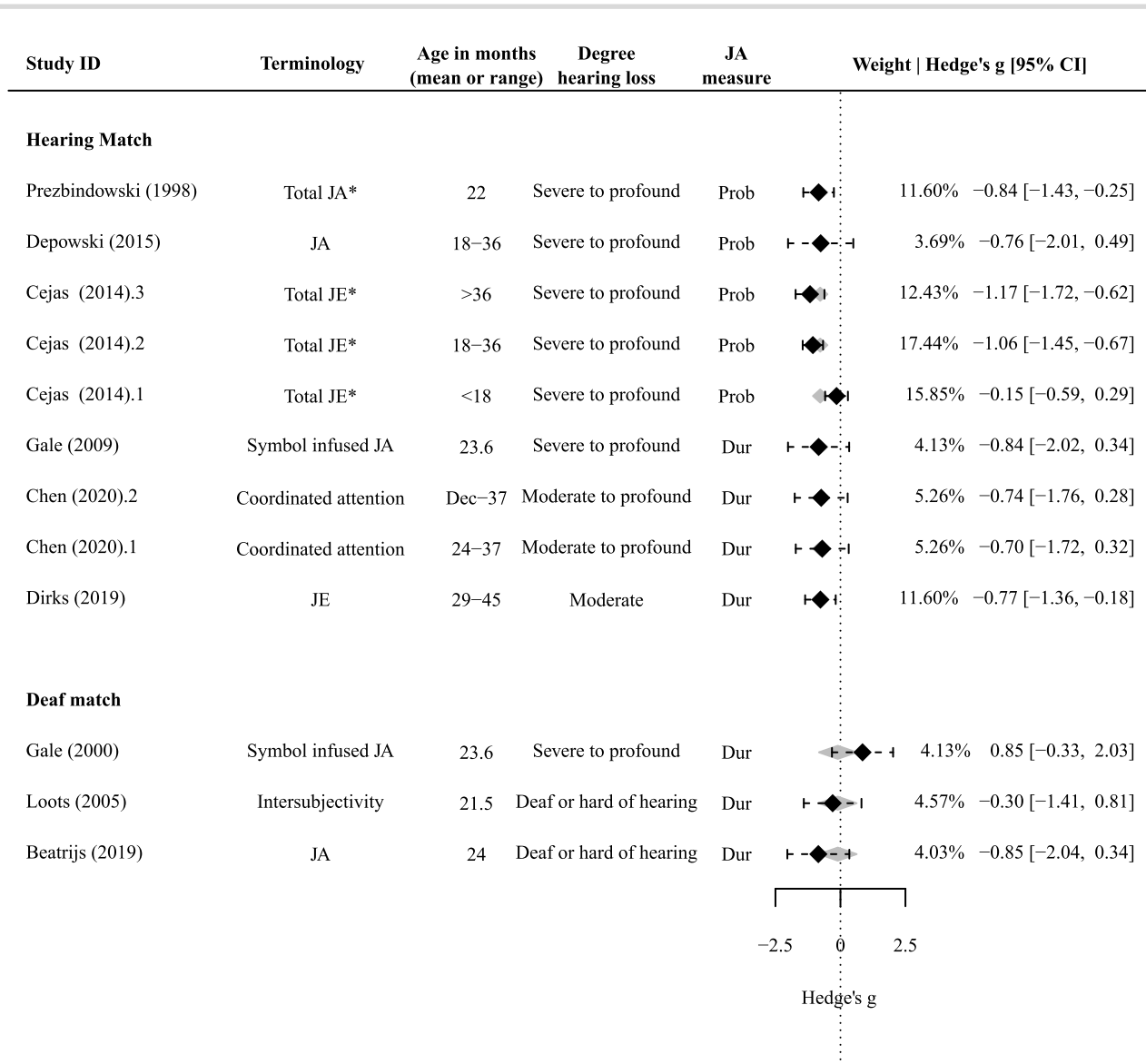


Figure 4. Forest plot showing overall and individual average effect sizes (Hedge's g) and 95% confidence intervals. A negative effect indicates that the congruent dyads spent longer time in episodes of joint attention. JA = joint attention; JE = joint engagement. Total JE or Total JA (studies marked with an *) is the sum of time dyads spent in supported joint attention, coordinated joint engagement, symbol infused supported joint engagement, and symbol-infused coordinated joint engagement states.

book reading setting. In both settings, maternal prompts were most often congruent with conventional signing and involved little additional overt behavior to direct the child's gaze. In the book reading condition, they observed that mothers were most effective in getting their child to look up when they tapped or waved at the child, or when the mother lifted her hands to begin an utterance.

Finally, comparing hearing parents of deaf children with hearing parents of hearing children, Gabouer et al. (2020) found no evidence for a difference in parent's use of touch to initiate joint attention. They did observe, however, that the overall use of touch was greater for hearing parents with deaf children than for hearing parents with hearing children.

Studies by Harris and colleagues report that movement (object movement or parent movement) plays an important role in 18-month old deaf children's episodes of attention. This holds both for deaf children with hearing parents and deaf children with deaf parents. Physical contact (tactile) and the use of noisy

objects (auditory) played a relatively small part in the episodes of attention of these children (Harris & Mohay, 1997; Harris & Chasin, 2005). Consistent with these findings, Waxman and Spencer (1997) report that the frequency of presenting objects and moving objects is higher in dyads with a deaf member (DH dyads and HD dyads) as compared to HH dyads. Also, for all three groups, they observe a decrease in the use of this strategy from 9 to 12 months and from 12 to 18 months.

A more passive strategy used by mothers to gain joint attention is to wait for their child's (visual) attention. Both Koester (2001) and Spencer et al. (1992) report that there is a tendency for deaf mothers to wait longer for their child to look back than hearing mothers.

A slightly different approach in describing ways in which parents and children come to coordinated attention is provided by Chen and colleagues (Chen et al., 2020). Chen and colleagues distinguish between gaze following pathways (looking at partners' eyes to infer gaze direction) to coordinated attention and

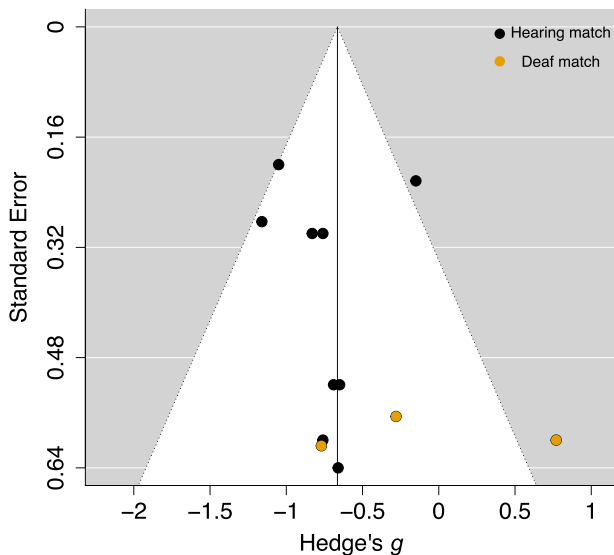


Figure 5. Funnel plot showing standard error of the Hedge's g effect size as a function of the effect size (studies on time spend in joint attention). The vertical line indicates the mean weighted effect size. Dots in black are individual effect sizes from studies that compare DH dyads to HH dyads, dots in yellow represent individual effect sizes from studies that compare DH dyads to DD dyads.

hand following pathways (manual actions on objects) to coordinated attention. They observe that, in parent-led attention episodes, children with hearing loss (24–37 months) used both their (hearing) parent's gaze and manual directions whereas the two groups of hearing children (chronological-age matched and hearing-age matched) rely mostly on their hearing parent's hand actions to achieve coordinated attention. In child-led coordinated attention episodes, all parents mainly follow their child's gaze direction to achieve coordinated attention, regardless of whether the child's hand cues are available or not.

In summary, compared to hearing parents with hearing children, parents of young deaf children seem to make more frequently use of visual and tactile strategies to gain their child's attention. Whether deaf and hearing parents differ in frequency and success of usage of these strategies is not clear yet, although (Wille et al., 2019) report that the duration of successful interaction moments was longer in dyads of deaf children and a deaf parent. Lieberman et al. (2014) report that the most successful strategy of deaf mothers of deaf children to get their child to look up from a book was tapping, waving or starting a new utterance by raising their hands. Movement of objects or parents to ensure visibility by the child while gaining attention is another strategy reported to be used by both hearing and deaf parents of deaf children. From the deaf child's perspective, following parental gaze and hands are both important in establishing and maintaining coordinated attention.

Narrative Synthesis 2: Joint Attention and Language

Given the fundamental role of joint attention for language development, several studies also investigated whether correlations exist between measures of joint attention and measures of children's spoken language proficiency. A study by Barker et al. (2009) reports a relationship between child language proficiency and the time that parent and child spend in joint attention.

They find that that the differences in time spend in parent-child communication between HH dyads and DH dyads disappears after controlling for language proficiency (as indexed via the Reynell Developmental Language Scales [Reynell & Gruber, 1990] and the MacArthur-Bates Communicative Development Inventories [NCDI; Fenson et al., 1993]). In addition, the percentage of children that uses language (manual and oral) during episodes of joint attention is lower for children with severe to profound hearing loss (CI candidates; 38% of the children, mean age = 2.96 years, SD = 0.95 years) than for hearing children (73% of the children, mean age = 2.76 years, SD = 0.94).

Two of the three studies that investigated the link between joint engagement and spoken language proficiency report a relationship that is statistically different from zero between these measures. In Dutch children with and without moderate hearing loss, the duration of joint engagement episodes is positively correlated with children's language ability (as indexed via the Reynell Developmental Language Scales—Dutch Version [Schaerlaekens et al., 1993] and the Schlichting language test [Schlichting et al., 1995]; Dirks & Rieffe, 2019). Similarly, Cejas et al. (2014) report that both deaf (CI candidates) and hearing children between 18 and 36 months of age with higher levels of language proficiency (as indexed via the Reynell Developmental Language Scales [Reynell & Gruber, 1990] and the MacArthur-Bates Communicative Development Inventories [NCDI; Fenson et al., 1993]) spend more time in total symbol-infused joint engagement states than children with lower levels of language proficiency. The third study, however, finds no statistical evidence for a link between time spend in symbol-infused attention states and language proficiency (number of different word types produced by the toddlers, mean length of utterances in words, NCDI scores) in deaf (no CI) and hearing 22-month old children (Gale & Schick, 2009).

In sum, this second narrative synthesis showed that the duration of episodes of (symbol-infused) joint attention is related to children's spoken language proficiency in three out of four studies.

Discussion

The present paper provided an in-depth overview of the studies that have investigated joint attention in parent-child dyads with congruent and incongruent hearing statuses. The two meta-analyses showed that on average dyads consisting of hearing parents and children with hearing loss (incongruency in hearing statuses), achieve fewer and briefer moments of joint attention as compared to dyads of hearing parents and hearing children and dyads of deaf parents and children with hearing loss (congruency in hearing statuses). The estimates of the magnitudes for these differences ranged from medium to large (establishment of joint attention) and from small to large (maintenance of joint attention). The two subsequent narrative syntheses, focused on differences in parental strategies used to achieve joint attention and to relations between joint attention and spoken language proficiency in children with hearing loss. These syntheses showed that deaf parents of children (deaf and hearing) make more frequently use of visual and tactile strategies to gain their child's attention compared to hearing parents with hearing children. What strategies are most successful for the establishment of joint attention in the different dyads groups and whether these strategies differ for dyads with congruent versus dyads with incongruent hearing status is not clear yet. As for the relation between joint attention and spoken language

proficiency, three out of four studies that investigated this interplay did observe a positive correlation between joint attention and general measures of spoken language proficiency.

Given the importance of joint attention for children's cognitive and social development, the current findings underline the need to better understand *what* strategies may facilitate or enhance the establishment and maintenance of joint attention in dyads with incongruity in hearing statuses (i.e., clinical relevance), a question that has not been explicitly addressed yet. This is not to say that it will be easy to identify strategies that facilitate or enhance the establishment or maintenance of joint attention. The multilayered nature of joint attention and the bidirectional interactions with cognition and behavior important for parent-child interaction (e.g., reciprocity, parental sensitivity, parental responsiveness, language proficiency, turn-taking, sustained attention; Tomasello & Farrar, 1986; Tamis-LeMonda, Kuchirko, & Song, 2014; Yu et al., 2018; Dirks & Rieffe 2019) make it difficult to disentangle the different cues (e.g., eye-gaze, point, vocalizations, movement, gestures) and behaviors that play a role in the maintenance and establishment of joint attention.

Another complicating factor in understanding differences in joint attention between dyads with congruity in hearing statuses and dyads with incongruity in hearing statuses is that most studies included in this review remain relatively a-specific as to *what exactly constitutes* the challenge in establishing and maintaining joint attention in dyads with incongruity in hearing statuses (hearing parents with children with hearing loss). Relevant questions to further investigate in this context may be (1) whether the establishment and/or maintenance of joint attention in dyads with incongruity in hearing statuses versus dyads with congruity in hearing statuses may be more difficult because the timing of (communicative) events is different for hearing members (synchronous timing of events) and deaf members (sequential timing of events) of the dyads or (2) whether the establishment and/or maintenance of joint attention is affected because the members of dyads with incongruity in hearing statuses employ a different combination of physical senses (hearing, sight, touch) to establish and/or maintain joint attention.

Joint Attention and Spoken Language Development in Children with Hearing Loss

Another goal of this review was to increase our understanding on the relation between joint attention and spoken language proficiency in children with hearing loss growing up with hearing parents. These children often show delays in their language development and it has been hypothesized that these delays may correlate with their joint attention challenges. This hypothesis builds on the assumption that, especially in the case of word learning (mapping labels onto objects), episodes of joint attention create optimal learning opportunities. After all, in the context of word learning, referential cues (e.g., eye gaze, pointing) may direct the child's attention to a specific object in the environment. The situation is complex, however, and different cues (referential and nonreferential) interact. In typically developing hearing children, there is quite some work that investigates how these different cues interact during word learning and how the weighing of eye-gaze (referential cue) versus mutual exclusivity and perceptual saliency of objects (nonreferential cue) changes over time (see review by Cetinçelik et al., 2021). Such fine-grained analyses on the weighing and use of referential versus nonreferential cues have not been conducted in children with hearing loss. The studies that investigated the link between

joint attention and (spoken) language proficiency in the context of hearing loss only assessed these links at a broader level, that is, by correlating joint attentional episodes with general measures of children's language ability. A next step may thus be to further investigate how deaf and hard of hearing children weigh referential cues and nonreferential cues in the context of word learning and whether their weighing is different from hearing children because of differences in attention allocation across modalities.

The Measurement and Operationalization of Joint Attention

Our study shows that there is little variation in the tasks used to study joint attention. The majority of studies assessed joint attention in a free play session either at the child's home or at the lab/clinic. The testing locations (home versus lab/clinic) were almost equally distributed across the included studies with slightly more studies conducted in the lab/clinic than at the children's homes. Although most studies did not control for situation or setting, a recent study of MacGowan et al. (2020) demonstrates that the task used (free play versus semistructured) may impact joint attention outcomes: HH dyads displayed more joint attention episodes than DH dyads during free play tasks, but there was no evidence for such difference using semistructured tasks⁵. Also, HH dyads achieved more moments of joint attention when joint attention was assessed at home as compared to the lab/clinic, suggesting that also the setting affects joint attention outcomes.

In our meta-analyses we observed that all studies operationalize joint attention as a sequential set of events involving a bid for attention and a response to this bid by the social partner within a certain amount of time that leads to shared interest (indexed via eye gaze) in an object, event of each other. Most studies tracked this sequence of events with scene cameras that have a temporal resolution of seconds. One study, however, used a novel eye-tracking technology in which parent and child gaze behavior was tracked with a temporal resolution of fractions of a second (Chen et al., 2020). Chen et al. explain that tracking eye gaze with such high temporal precision fits modern views on joint attention in which joint attention depends on a tight and fast coupling of sensorimotor dynamics and bodily adjustments between the two members of a dyad.

At this point it may be relevant to emphasize that there are currently no standardized or validated measures of joint attention (see also MacGowan et al., 2020). Also, it is yet an empirical question whether or not joint attention should be operationalized differently for different learner populations. Within our set of studies only the studies conducted by the research lab of Tasker (Nowakowski et al., 2009; Tasker et al., 2010) decided to operationalize joint attention differently in HH dyads versus DH dyads. In these studies, the time frame within the members of the dyad had to respond to bids of joint attention was prolonged (from 5 to 15 s) in dyads with a deaf member in comparison to dyads with hearing members only. The authors decided to do so because they assumed that dyads with a deaf member adhere to a sequential timing of communicative events, in which it takes longer to achieve joint attention (Tasker et al., 2010, p. 518). If this assumption is true, it may be that previous studies that did not operationalize joint attention differently for HH dyads and HD dyads missed a lot of joint attention episodes simply because it took dyads with a deaf member longer to achieve a moment of joint attention. In other words, whether or not joint attention

should be operationalized differently in HH dyads versus HD dyads is an important question to further investigate.

Generalizability of the Outcomes

There are several aspects that limit the generalizability of our conclusions. Firstly, our systematic overview of studies reveals that research groups tend to publish multiple times on the same groups of dyads (only 12 unique samples across 26 articles). This overlap between samples across different papers is not always clear, which may have biased past interpretations and comparisons of results.

Secondly, all studies included in this review were conducted in Western, educated, industrialized, rich and democratic (WEIRD) countries. Joint attention may be culturally defined. In the Western world eye contact is important for the establishment and maintenance of joint attention (Cetinçelik et al., 2021), which is in line with our conclusion that all studies operationalized joint attention in terms of eye gaze (see section “The measurement and operationalization of joint attention”). Other cultures or population groups (e.g., blind people) may achieve joint attention using different senses (Siposova & Carpenter, 2019). Such bias towards studies on visual joint attention is also noted in the literature on joint attention in typically developing children (Akhtar & Gernsbacher, 2007). Cultural differences may not only involve different senses, but also the number of people that are typically involved in interactions as well as the number of objects and events that interaction partners simultaneously focus on. Akhtar and Gernsbacher (2007) for example point out that some cultures spend more time in polyadic (group) interactions than dyadic interactions. It is yet unknown whether such cultural differences can be generalized to dyads with incongruent hearing status and if so, how the number of interactional partners impacts the establishment and maintenance of joint attention in dyads with incongruent hearing status across cultures. These are relevant questions, because even in Western middle-class societies children spend lots of time in polyadic interaction (e.g., with both their parents, with siblings or with peers).

A third concern is that the majority of participating dyads consisted of mothers and their child. Only two studies actively recruited fathers and subsequently assessed gender related differences in the strategies used to achieve joint attention (Loots & Devisé, 2003; Wille et al., 2019). Both studies do report differences between fathers and mothers in their strategies used to achieve joint attention (e.g., deaf fathers were found to display more an attitude of waiting than deaf mothers, which is also in line with findings in hearing-hearing dyads, Barton and Tomasello, 1994). This overlook of fathers is in line with the outcomes of a recent systematic review by Szarkowski and Dirks (2021) who conclude that it is yet impossible to state the influence(s) that fathers have on the development of their deaf or hard of hearing children.

Future Directions

Our meta-analyses in combination with the narrative syntheses revealed that dyads with incongruity in hearing statuses achieve fewer and briefer moments of joint attention as compared to dyads with congruity in hearing statuses, and that it is currently unclear what strategies lead to more and longer moments of joint attention in this group. Also, relatively few

studies investigated the link between joint attention and language proficiency. To better understand how joint attention plays a role in the language development of deaf or hard of hearing children with hearing parents, more longitudinal studies with larger samples are needed. These studies may also focus on specific aspects of language development (e.g., word learning).

In the first part of this discussion, we concluded that the field may benefit from a better conceptual understanding of what interactions between cognition and behavior lead to difficulties in the establishment and maintenance of joint attention in dyads with incongruity in hearing statuses. Neuroimaging techniques may provide an opportunity to study joint attention and its interaction with other cognitive processes (e.g., attention or memory) and behaviors from within an integrative framework. To the best of our knowledge, there are no neuroimaging studies on joint attention in deaf or hard of hearing children. A pilot study on typically developing hearing toddlers by Hutman et al. (2016) investigated how neural correlates of object discrimination (necessary for object word learning) differed between situations with and without joint engagement. The authors conclude that neural correlates of joint engagement can be of use in evaluating the risk for social learning impairments during a time frame in which behavioral markers of atypical development are both subtle and emergent and thus difficult to assess (Hutman et al., 2016). In other words, such an approach may turn out to be valuable for assessments in clinical contexts.

Novel headmounted eye-tracking technologies may provide another opportunity to study joint attention in a more ecological valid context. With these novel technologies, joint attention can be studied with a temporal precision of fractions of seconds. Such high temporal resolution allows researchers to study how subtle differences in the temporal ordering of (communicative) events between hearing and deaf members of a dyad may affect the establishment and maintenance of joint attention and what role synchronicity plays during this process (Chen et al., 2019b). At the same time, the headmounted eyetrackers provide unique first-person views (rather than third-person views) on what the members of a dyad actually perceive and pay attention to. These videos may lead to corpora of parent-child interaction that provide opportunities for integrative approaches in which the effects of several factors (e.g., environmental, behavioral, sensory, nonreferential, and referential cues) on the establishment and/or maintenance of joint attention can be studied simultaneously. For an example of such corpus we refer to the ECOLANG corpus of the UCL Language and Cognition laboratory led by Gabriella Vigliocco (Language and Cognition Lab – Ecological language, 2021).

Finally, our study demonstrated that the studies included in this review used slightly different definitions of the concept of joint attention. This makes it difficult to understand what consequences (in)congruity in hearing statuses has on the establishment and maintenance of joint attention at a theoretical level (for a similar observation in a systematic review on synchrony—a concept closely related to joint attention—see Leclère et al., 2014). Pointing out these issues, we expect that this review may serve as the start of quest towards a more detailed description (taxonomy) of what is meant by joint attention in the context of hearing loss, how it can be best operationalized in this context and whether the operationalization should depend on the hearing status of the dyad members (see also “The measurement of joint attention”).

Notes

1. Number of successful established joint attention bids divided by the total number of attentional bids.
2. We assigned a number to each unique sample of participants. In Gale and Schick (2009) and Chen et al. (2020), outcomes of the same hearing mismatch groups are compared to both a hearing match and deaf match control group. We control for this dependency between effect size by adding “DH sample” to the random effects structure of the model.
3. As for meta-analysis 1 DH sample ID was added to the random effect structure of the model to control for dependency between the effect sizes for those studies that compared one DH group to both a HH group and DD group (Gale and Schick, 2009; Chen et al. 2020).
4. Waxman and Spencer (1997): Wave in the visual field occurred rarely for all groups. The DD dyads exceeded all other groups for this strategy and de Deaf mothers with hearing kids exceeded the two groups with hearing mothers. Also this strategy increased between 12 and 18 months.
5. Note that this study was published after our search period and that we decided not to include the outcomes of this study in our analyses because of overlap of participating dyads with (Tasker & Schmidt, 2008; Nowakowski et al., 2009; Tasker et al., 2010).

Supplementary Data

Supplementary material is available at *Journal of Deaf Studies and Deaf Education*.

*Studies marked with an asterisk are included in at least one of the two narrative syntheses.

**Studies marked with two asterisks are included in at least one of the two meta-analyses.

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Conflicts of Interest

No conflicts of interest were reported.

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