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The relationship between feeding and non-nutritive sucking behaviors and speech sound development: A systematic review

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Short Title: SUCKING AND SPEECH SOUND DEVELOPMENT: SYSTEMATIC REVIEW

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1. Abstract

Background

Children with and without Speech Sound Disorders (SSD) are exposed to different patterns of infant feeding (breast/bottle feeding) and may or may not engage in non-nutritive sucking (NNS) (pacifier/digit sucking). Sucking and speech use similar oral musculature and structures, therefore it is possible that early sucking patterns may impact early speech sound development. The objective of this review is to synthesise the current evidence on the influence of feeding and NNS on the speech sound development of healthy full-term children.

Summary

Electronic databases (Pubmed, NHS CRD, EMBASE, MEDLINE) were searched using terms specific to feeding, NNS and speech sound development. All methodologies were considered. Studies were assessed for inclusion and quality by two reviewers. Of 1031 initial results, 751 records were screened and five primary studies were assessed for eligibility, four of which were included in the review. Evidence from the available literature on the relationship between feeding, NNS and speech sound development was inconsistent and inconclusive. An association between NNS duration and SSDs was the most consistent finding, reported by three of the four studies. Quality appraisal was carried out using the Appraisal Tool for Cross-Sectional Studies (AXIS). The included studies were found to be of moderate quality.

Key Messages

This review found there is currently limited evidence on the relationship between feeding, NNS and speech sound development. Exploring this unclear relationship is important because of the overlapping physical mechanisms for feeding, NNS and speech production, and therefore the possibility that feeding and/or sucking behaviours may have the potential to impact on speech sound development. Further

high-quality research into specific types of SSD using coherent clinically relevant assessment measures is needed to clarify the nature of the association between feeding, NNS and speech sound development, in order to inform and support families and healthcare professionals.

2. Introduction

2.1. Background

There is much discussion and debate in the current literature on the advantages of breastfeeding over bottle-feeding, with positive cognitive outcomes often cited for language in later childhood [1-4]. However, evidence on the influence of feeding type on speech sound development is less readily available (e.g., Fox et al. 2002 [5]). Infant feeding (breast, bottle and mixed feeding) and non-nutritive sucking (NNS) (pacifier/digit sucking) are typically concurrent practices in the early lives of infants across the world [6-8]; therefore it is important to consider both of these with regard to the impact on speech sound development. Evidence for an indirect detrimental impact of NNS on speech sound development is indicated with regard to dentition [9-10] and hearing loss resulting from otitis media [11-12]; however the question of a potential direct impact of NNS on speech sound development is of interest due to the shared physical oral mechanisms of these two processes.

The mechanisms for successful bottle and breastfeeding have been described and compared [13], and significant differences in sucking frequency, pressure and muscle activity have been identified and examined [14-15]. Speech develops after these feeding mechanisms have become established and, given the shared musculature between speech and sucking, it is possible that speech sound development could be influenced by infants' early experiences of feeding and NNS [16-17]. If this were the case, there may be observable differences in the speech sound production of children who have different patterns of feeding and NNS. Furthermore, it may be that different patterns of feeding and NNS are associated with Speech Sound Disorder (SSD). In taking a mechanistic view of speech sound development, it is imperative to include both feeding and NNS in this review as either and both have significant influence on infants' early sucking experience. While some studies have described feeding, NNS and anatomical development in terms of atypical dentition and general oral development [18], the evidence of the relationships between the effects of feeding, NNS and speech sound development requires specific exploration to inform our understanding of these closely associated physical mechanisms. Many studies report evidence against a relationship between speech and non-speech mechanisms [19-23]. However, the individual work undertaken in such laboratory or clinic based

pieces of research are not compatible with understanding the complex development of that skillset in the very young child. The complexity of the development taking place during the early postnatal period means that consideration must be given to all three factors (feeding, NNS and speech sound development) as they are distinct but could also overlap and build on each other. Oral feeding from birth through infancy is a highly intensive and enduring physical behaviour. In addition, non-nutritive sucking behaviours often occur concurrently and, can be comparably intensive and enduring from birth through to early childhood. Therefore, these very early intensive sucking behaviours (nutritive and non-nutritive) may have an inevitable influence on the development of motor control and sensorimotor feedback systems for these oral mechanisms and muscle groups. As such, it may be deemed improbable that any use of the oral musculature and articulators, for the purposes of subsequent speech development, from babble through to more refined speech sound productions, could occur in an entirely sterile way. Indeed, there is recent evidence that weak sucking in infants as young as four weeks of age is a significant predictor of persistent SSD at age 8 years [17]. Bunton [21] states that speech motor control is internally driven relating vocal tract changes to acoustic targets, while non-speech motor control is driven by external visuo-spatial or proprioceptive targets. However, within the very nature of clinical therapy Speech Pathologists routinely employ visuo-spatial and proprioceptive cues to support speech production with a high frequency of success [24-26]. It can, therefore, be argued that speech and non-speech motor control cannot reasonably be considered entirely distinct. Indeed, some studies suggest a continuum for development between speech and non-speech tasks [27].

The aim of this systematic review is to synthesise the available evidence about the relationships between feeding (breastfeeding, bottle feeding, mixed feeding methods),NNS behaviours and speech sound development and the incidence of SSD in children from birth to early childhood. This review addresses the following key questions:

- Is there evidence that infant feeding methods and NNS impact the way young children develop speech sounds?

- Is there evidence that children who experience different patterns of NNS as babies have different outcomes in their speech sound development, such as SSD?

This systematic review investigates the literature on feeding and NNS in the development of speech sounds in healthy, full-term, preschool children. For the avoidance of confusion, the term "speech sound development" is consistently written in full, whereas the term "Speech Sound Disorder" is consistently abbreviated to SSD.

2.2. Methods

The review strategy was adapted from the Cochrane Collaboration systematic review methodology and uses a narrative synthesis [28] and guidance from Petticrew & Roberts [29]. A narrative synthesis approach was deemed most appropriate due to the mixed nature (qualitative and quantitative) of the data likely to be retrieved from the included papers. The review was registered on the PROSPERO database (CRD42018106268).

2.3. Identification of Selection Criteria

The Booth & Fry-Smith [30] PICO model (population, intervention, comparison, outcome) guided the development of the search strategy. The population of interest was children from birth into early childhood, with or without identified SSD. Table 1 below lists the inclusion and exclusion criteria. Papers that reported samples including children born prematurely (more than 15% of the total sample), or those with diagnosed congenital disorders, identified learning difficulties, sensorineural hearing loss, or populations that had received speech therapy intervention as part of the reported study were excluded from the review as these factors could also impact on speech sound development. This follows principles set out in similar systematic reviews in comparable cohorts (e.g., Roulstone et al. 2015 [31]). The intervention (behaviour) of interest was infant feeding, comparing outcomes in speech sounds across three comparator interventions – breast-feeding, bottle-feeding and mixed feeding. A second analysis considered presence or absence of NNS and its associations with speech sound outcomes. Only papers reporting both feeding and NNS with regard to speech sound development were included in this review. This systematic review of the current evidence base of journals and

abstracts in this topic area considered all methodologies and settings. Globally accessible articles were examined, providing that they had been published, or were available, in the English language.

2.3.1. Outcomes of Interest

All included studies were required to include an outcome for speech sound development, whether qualitative (e.g., descriptive responses to parent questionnaires) or quantitative (e.g., statistical results obtained from objective clinical speech sound assessments).

[Table 1 about here]

2.4. Search Strategy

The search strategy was designed in consultation with all authors and the search terms following a review of the Cochrane database, PROSPERO and database of abstracts of reviews of effectiveness. Discussions with a specialist speech and language pathologist working with children with SSD facilitated the identification of specific search terms relevant to all possible and appropriate terminology for speech sound development and SSD. A combination of `free text' terms with Boolean operators and truncations were used as follows:

2.4.1 Feeding Search Term

(((((((bottlefe) OR (bottle-fe*) OR (bottle fe*)))) AND (((breastfe*) OR (breast-fe*) OR (breast fe*))))*

2.4.2 Non-Nutritive Sucking Search Term

(((dumm) OR (pacifier*) OR (non-nutritive sucking)))*

2.4.3 Speech Search Term

(((phon) OR (speech) OR (speech disorder*) OR (speech impairment*) OR (speech sound disorder*) OR (speech sound difficult*) OR (speech retard*) OR (speech delay*) OR (speech disabilit*) OR (speech handicap*) OR (speech problem*)))))*

2.5. Findings of the Search Process

2.5.1. Traditional Search Strategy

165 The process and screening results for the database searches are described in Figure 1. Six separate
166 searches were conducted in electronic databases: Pubmed, (inc. PubMed Health, PubMed Central and
167 NCBI Bookshelf Database), NHS CRD https://www.crd.york.ac.uk/CRDWeb/, OVID full text Journals,
168 Embase 1974 to 2018 week 31, Ovid MEDLINE(R) and Epub Ahead of print, In-Process & Other Non-
169 Indexed Citations, and Daily 1946 to July 27, 2018, CINAHL (inc. MEDLINE, Chicano Database, Child
170 Development and Adolescent Studies and AMED (Allied and Complementary Medicine) 1985 to July
171 2018. The PRISMA checklist [32] was followed and a flow chart (Figure 1) details the process of article
172 selection from the formal database searches. Of 981 results, 702 papers were screened (following
173 duplicate removal) and 698 were excluded in accordance with the validity criteria (Table 1). Four full-
174 text articles were assessed for eligibility, two of which were excluded as they did not meet the inclusion
175 criteria. All references from the four full-text papers were reviewed to check for additional articles. No
176 appropriate papers were identified for inclusion in the full paper review stage. Only two papers were
177 retained for inclusion in the narrative synthesis.

178

179 [Figure 1 about here]

180 **Figure 1. PRISMA Flow Chart for Traditional Database Searches**

181

182 ### 2.5.2. Novel 'Google' Search Strategy

183 An additional search of Google, a major search engine [33], was conducted using the simplified search
184 term [infant feeding, speech development and sucking]. Figure 2 shows the PRIMSA flow chart
185 detailing the process of article screening and selection based on the Google search. The first five pages
186 of the Google search, which represented 50 results, were screened for title relevance. Of these results,
187 one article/post was a duplicate from the original formal database search and 48 were rejected; one
188 paper was identified for inclusion in the full article review (see Figure 2). The Google search results
189 also included a website with a bibliography, which was scrutinised. All of the papers had been
190 previously identified in other searches.

In addition to the above searches, one unpublished paper [34], identified through discussions with review colleagues, was included in the screening process and subsequently retained. A total of four papers were included in the full review: two identified from traditional database searches, one from Google and one unpublished paper.

[Figure 2 about here]

Figure 2. PRISMA Flow Chart for Google Search Engine

2.6. Search Validation

The first author (SB) excluded irrelevant articles by screening titles and abstracts (see Figure 1). The remaining abstracts were fully reviewed by the first author and SH independently. Any disagreements were resolved through discussion and when consensus was not met the article was included in the next stage. Four full text articles were then retrieved and further considered against inclusion criteria by the SB and SH.

2.7. Data Extraction

The data extraction was undertaken by the first two reviewers using an adapted version of the published data extraction template for Randomised Control Trials (RCT) and non-RCTs [35]. The results from the data extraction stage were discussed and agreed between the first and second reviewers.

2.8. Quality Appraisal

Selection of the quality appraisal tool was undertaken once the final list of included papers had been obtained and reviewed for their methodology. All four papers used a cross-sectional study design and subsequently the Appraisal Tool for Cross-Sectional Studies (AXIS) was used by SB and SH [36]. This tool was selected as the most appropriate for assessing the quality of the included papers because it has been specifically designed for the critical and quality appraisal of cross-sectional studies. The AXIS

comprises 20 questions to appraise each paper's introduction, methods, results, discussion and other issues related to bias and ethical conduct. The authors assigned a score to each of the categories. 2 for papers that clearly provide the information required by the AXIS tool, 1 if this information is partially present, but not clearly stated, and 0 if it is not present at all. This led to a maximum possible score of 40 on the AXIS. The quality appraisal of the included papers was completed separately by SH and SB and scoring consensus was reached following discussion.

Table 2 below summarises the total quality scores awarded to each paper. Baker *et al* [34] scored highest in the quality appraisal with almost 75% of the maximum score, while Pereira *et al* [37] and Vieira *et al* [38] obtained the lowest scores with just over 50% of the maximum.

[Table 2 about here]

2.8. Data Synthesis

Heterogeneity precluded meta-analysis; therefore, a narrative synthesis was used which summarised the findings descriptively and guided the synthesis.

3. Results

3.1. Review of the Data

The following section describes the presentation of the data in each of the four included papers.

3.1.1. Statistical Techniques

Variation was found in the statistical approaches employed across the four papers (Table 3). In their data tables[39](p.5-6) Barbosa et al [39] provided overall calculated probability, or p values, relating to each variable when compared with age or speech sound assessment classification. Specific p values corresponding to the reported odds ratios and confidence intervals for more specific associations presented in the results are not provided. In contrast, Vieira et al [38] consistently reported associated odds ratios (ORs) with 95% confidence intervals (CIs) alongside their p values. Baker et al [34] and Pereira et al [37] only reported p values.

3.1.2. Methodological Approaches

All four included papers used parent/carer questionnaires to collect data on participant feeding and sucking histories. Both Vieira *et al* [38] and Pereira *et al* [37] reported the use of a 'structured interview' approach. Information is not provided on the interviewer or recording of these data. Barbosa *et al* [39] and Baker *et al* [34] distributed self-administered parent questionnaires. While all studies collected data on presence and duration of feeding and NNS behaviours, only Barbosa *et al* [39] collected data on the frequency of bottle-feeding and pacifier use.

All except one of the papers attempted objective assessment of the participants' speech sound development. Pereira *et al* [37] based their findings solely on parent report and provided no objective measure for the speech sound development of the children in their study. Although Pereira *et al* [37] referenced specific phonemes in their definition of 'speech disorder' or 'speech changes', the single item on their parent questionnaire relating to this measure required only a binary yes/no response and asked simply "difficulties / changes in speech?" without reference to specific sounds or clarification on the authors' intended meaning of 'speech'. As such it is difficult to draw firm conclusions on the basis of this paper due to the potential for variation in respondents' concept of 'speech', and therefore inconsistency in their responses.

3.1.3. Sample Populations

Details of the population samples for each study are provided in Table 3. Only two of the four papers [34, 38] reported any use of exclusion criteria in their sample definitions, and only one of these, hearing loss, was common to both studies (see Table 4). Baker *et al* [34] reported the most comprehensive exclusion criteria, including genetic, medical and developmental factors known to have some association with SSD.

Table 3. Summary Table of Included Studies.

[Table 3 about here]

[Table 4 about here]

3.1.4. Definition of SSD

A key challenge for this review was the disparity in what is meant by the term 'Speech Sound Disorder' between papers. Barbosa *et al* [39] used the terms 'speech disorder(s)' and 'speech processing', the former of which they broadly describe as having the potential to "*impair communication and literacy*"[39](p2). Specific reference to distinct types of SSD was not made; however through their use of the Brazilian speech sound assessment TEPROSIF [40] to "*determine the type and number of errors in the child-age related phonological processes*" [39](p3), the implication was to focus on phonological impairment (PI). Baker *et al* [34] were more explicit in stating their specific focus on children with diagnosed PI, and defined the group as presenting with "*one or more age-inappropriate common phonological error patterns [...] with no evidence of motor speech involvement*"[34](p7). As Baker *et al* [34] themselves acknowledged, "*PI is presumed to be a cognitive-linguistic difficulty involving a difficulty abstracting rules about the phonological system, and the abstract phonological representation of speech rather than an articulation difficulty. As such, it is reasonable to suggest that non-nutritive sucking habits would be unrelated*"[34](p11). Pereira *et al* [37] made reference to both 'speech disorder(s)' and 'speech changes' and acknowledged that they did not distinguish between types of SSD. They provided some definition of their application of the term 'speech disorders' as "*those reported by the parents and/or guardians with respect to the production of the phonemes /t/, /d/, /n/, /l/, /r/, /s/, and /z/, considered comprehensively as they are associated with alterations in the SS* [stomatognathic system]"[37](p2). The repeated emphasis within this paper on the structures and functions of the stomatognathic system, defined by the authors as comprising the functions of suction, swallowing, mastication, respiration and speech[37](p.2), indicated the author's intention to explore 'speech disorders' relating to articulation, rather than those that are cognitive-linguistic in nature. Vieira *et al* [38] also referred to 'speech disorders', 'speech changes' and the SS, as well as 'speech impairment'. They defined their case group as children with "*omissions, substitutions, additions or distortions of phonemes related to functionality*

292 *and associated with the motor aspect of speech production"*[38](p1361). Vieira *et al* [38] specifically stated
293 that "*phonemic productions associated with* […] *chronology of acquisition of children's phonemes*"[38](p1361)
294 (i.e., age-appropriate developmental phonological processes) were not considered pathological. It may
295 be argued that, as with Pereira *et al* [37], this paper focused on articulatory SSD.

296 ### 3.2.5. Definition of Population

297 Exclusion criteria for defining the study samples were not included in either Barbosa *et al* [39] or
298 Pereira *et al* [37] (Table 3). This may mean that their samples included children who had additional
299 difficulties, which, in turn, could have impacted on, or been the underlying cause of, their SSD. Of the
300 four included studies, Baker *et al* [34] presented the most comprehensive exclusion criteria.

301 ### 3.2.6. Confounding Factors

302 Barbosa *et al* [39] acknowledged the likely influence of confounding factors in their study; however
303 they adjust only for gender and age (Table 4). Pereira *et al* [37] considered only gender, age and
304 number of children per household. No information is provided as to whether their statistical analysis
305 accounted for these factors. Baker *et al* [34] collected information for age, gender, hearing,
306 oromuscular structure and function. They also did not state whether these were included in their
307 statistical analysis. Of the four included studies, Vieira *et al* [38] collected information on age, gender,
308 'shift in educational unit', family income, maternal age, maternal schooling and family history of speech
309 impairments. They did not state whether these were included in their statistical analysis.

310 ### 3.2.7. Missing Data

311 Unreported missing data presents a challenge in the interpretation of the data tables in Vieira *et al* [38].
312 When case and control group sample size totals for the different variables are manually calculated the
313 extent of missing data becomes clear. Moreover, when the overall group total (i.e., case and control
314 combined) is calculated for bottle use the number of cases exceeds the reported sample total, indicating
315 some measurement error [38]. This leads to concern about the validity of the analysis and
316 interpretation of the data in this paper. Manual calculations of group totals in Table 2 of Barbosa *et al*
317 [39] indicate missing data across the variables, but this was not acknowledged by the authors. Pereira

318 *et al* [37] also failed to acknowledge the extent of missing data within their report. Their paper
319 presents data on the correlation between NNS and SSD (Table 4). 127 children were reported as having
320 used a pacifier, but only 119 were included in the analysis. Baker *et al* [34] reported the extent of
321 missing data in their analysis.

322 ### 3.2.8. Exposure Measures – Nutritive and Non-nutritive Sucking

323 All four of the included papers reported data on infant feeding type and duration. Three of the four
324 included papers [34, 37, 39] collected data on NNS duration. However, only one [39] collected data on
325 NNS frequency.

326 ### 3.2.9. Outcome Measures – Speech Sound Disorder (SSD)

327 The SSD outcome measurement approach varied across the four papers in this review and although
328 formal assessment was attempted by three studies, the administration quality of the measures was
329 inconsistent. Unusually the questionnaire implemented within Pereira *et al* [37] specifically asked for
330 perceived speech sound changes, but they explicitly chose not to collect this information from the
331 parents of children aged 1-3 years. The modification of the questionnaire for this age group was not
332 defended by Pereira et al [37] and does not find a basis among the current literature, which suggests
333 the potential for identification of SSD within this age bracket [41-43].

334 Barbosa *et al* [39] used the TEPROSIF assessment, which requires the child to imitate a word, either
335 from a spoken phrase or in isolation [40]. Their criteria of "Below Normal" speech sound performance
336 as at least -1 standard deviation represents a liberal cut-off as many other studies have used more
337 stringent criteria [44-46]. It must be assumed that the "Below Normal" group includes a proportion of
338 children who could be considered typically developing in some other studies. As the authors did not
339 provide specific scoring information, further exploration of this issue is not possible. Vieira *et al* [38]
340 also used a published validated assessment, the Children's Language Test [47], to assess speech sound
341 production on both naming and imitation tasks. Only those children who presented with a sound error
342 occurring in both tests were assigned to the 'case' group. The authors implied that children presenting
343 with errors pertaining to age-appropriate phonological processes were not included in the case group

[38]. As scoring information was not presented for the case or control groups, it is not possible to determine or assess the severity of children's speech sound errors within the case group. Baker *et al* [34] provided a clear description and explanation of their selected published assessment tool, the Diagnostic Evaluation of Articulation and Phonology (DEAP) [48]. Following administration of the Phonology Assessment single word naming test, children were assigned to one of four groups based on their obtained DEAP standard score, percentage of consonants correct (PCC) score and error patterns. Only data from the PI group were included in the study. Children assigned to the PI group obtained a DEAP standard score of 6 or less based on their PCC score. A score of 7-13 is understood to fall within the normal range [48].

3.3. Managing Bias

The following section considers risk and evidence of bias across the four included papers.

3.3.1. Sample Baseline Imbalances

Imbalances between groups of baseline variables, such as age and gender, can influence or bias the outcome, and so it is important to consider these when interpreting the reported findings.

Pereira *et al* [37] reported a sample population containing essentially equal genders, although no information was provided on sample selection.

In Vieira *et al* [38] there is a reporting error in the paper. They reported equal overall sample sizes for the case and control groups; however, manual calculations of the group totals from the data presented in their analysis[38](p.1362) indicate a marked group imbalance (see Table 1). There is also a significant gender imbalance within the total sample, which contains 73% more males than females. Vieira *et al* [38] briefly acknowledged this imbalance in their discussion. Baker *et al* [34] also reported a sample gender imbalance, with 55% more males than females in their PI group. The SSD prevalence figures in the wider literature also show a tendency for more boys than girls [17, 49-52].

Barbosa *et al* [39] included in their sample children born prematurely (n=19) and, as acknowledged by the authors, this population are significantly more likely to present with "increased risk of

developmental problems with speech"[39](p4). The inclusion of this population, which constitutes 15% of the total study sample, may have some impact on the results as they potentially comprise almost 1/3 of the reported 'below normal' group. Prematurity is often cited in the wider literature as being associated with speech sound difficulties in later development [53-55].

3.3.2. Recall Bias

Inherent in the methodological use of participant questionnaires is the risk of recall bias [56]. While all four studies in this review employ this data collection approach, only Baker *et al* [34] did not acknowledge the potential limitation. Recall bias is perhaps most problematic with regard to the Pereira *et al* [37] study, which relied solely upon parent report for information on early feeding, sucking and speech sound development and included children up to age 12 years. The remaining studies focussed on the age range 3-5 years; therefore perhaps the influence of recall bias in each case may be considered to be broadly equal.

3.4. Summary of Findings from Included Papers

Although numerical data from the papers was insufficient to undertake meta-analysis, statistical information such as odds ratios and confidence intervals are included in each of the four papers. As previously stated, provision of this information by the authors is inconsistent across the papers.

3.4.1. Feeding Type and Speech Sound Development

Barbosa *et al* [39] suggested an association between bottle feeding and SSD in preschool children, such that delaying bottle use until after age nine months appeared to show some small protective effect (OR: 0.32, 95% CI: 0.10-0.98). Pereira *et al* [37] also reported a significant correlation between speech sound difficulties and bottle feeding (p=0.056). This may indicate a liberal application of their reported adopted 5% significance level[37](p.2). Vieira *et al* [38] found no significant association between feeding type and SSD. Baker *et al* [34] similarly found no association between feeding type and the presence or absence of SSD (specifically PI).

3.4.2. Duration of Feeding Type and Speech Sound Development

Pereira *et al* [37] and Vieira *et al* [38] both collected data on duration of feeding method and speech sound development but did not report on these data within their papers. Baker *et al* [34] suggested a trend whereby longer breastfeeding duration is associated with higher percentage consonants correct (PCC) scores, resulting in more accurate speech sound production for spoken words. Barbosa *et al* [39] reported that children scoring as normal or 1 standard deviation above normal on the "Test para evaluar los procesos fonológicos de simplificación" (TEPROSIF) speech sound assessment tended to have been breastfed for longer than those scoring below expectation for their age [40]. They asserted that delaying bottle feeding until after age 9 months may be to some extent a protective factor against subsequent SSD (OR: 0.32, 95% CI: 0.10-0.98).

3.4.3. Non-nutritive Sucking and Speech Sound Development

Barbosa *et al* [39] suggested an association between NNS and SSD in preschool children. They found that children who sucked their fingers were three times more likely to have speech sound difficulties than children who did not present with this behaviour (OR: 2.99, 95% CI: 1.10-8.00). It is important here to note the wide confidence interval reported for this finding. Pereira *et al* [37] found a correlation between pacifier use and speech sound difficulties (p=0.046). Neither Vieira *et al* [38] nor Baker *et al* [34] found a significant association between NNS and SSD.

3.4.4. Duration of Non-nutritive Sucking and Speech Sound Development

Baker *et al* [34] reported that, while the relationship between NNS and presence of SSD was non-significant, they did identify a trend between longer pacifier use and lower PCC scores. Barbosa *et al* [39] reported that children who used a pacifier for more than three years were much more likely to present with below normal speech sound development (OR: 3.4, 95% CI: 1.08-10.81). Pereira *et al* [37] suggested that using a pacifier for less than one year was not associated with speech sound difficulties, whereas digit sucking persisting for up to four years was positively correlated with the presence of SSD (p= 0.012). Vieira *et al* [38] found no association between NNS and SSD.

4. Discussion

419 This review aimed to examine the evidence of the relationship between infant feeding methods, NNS
420 behaviours and speech sound development in early childhood. The deliberate inclusion of only those
421 papers that address all three aspects of this relationship is due to the high prevalence of concurrent
422 feeding and NNS behaviours in infancy and early childhood [5-7]. To exclude one or other elements
423 would be to disregard significant relevant factors in this association, and risk drawing false conclusions
424 from incomplete information.

425 ### 4.1. Methodological Limitations of this Paper

426 Although clear systematic criteria were used for search and inclusion strategies, it is possible that a
427 number of biases may enter into the process by way of variations in definitions (e.g., SSD) and in
428 general by the specific inclusion criteria. For example, by including only studies that contain both
429 feeding and NNS, the possibility of deriving a fuller understanding of the impact of a single type of
430 sucking behaviour on the development of speech sounds is not possible. For the purposes of this
431 review, we purposely searched for evidence that allowed for the comparison of feeding and NNS. The
432 aim was to develop a picture of the current status of comparative findings.

433 The limited number of studies available for review makes it difficult to draw firm conclusions and
434 develop hypotheses about how differing characteristics and conditions may lead to SSD. It is worth
435 noting that two of the included papers, Vieira *et al* [38] and Pereira *et al* [37], have been translated from
436 the original language. This may have had some impact on the clarity of some of the language and
437 explanations within the papers.

438 ### 4.2. Limitations of Reviewed Studies

439 The following section discusses the limitations of the four studies included in this review.

440 #### 4.2.1. Definition of SSD

441 It is evident that, in terms of the defined outcome of SSD, there is an equal division between the four
442 included papers. Barbosa *et al* [39] and Baker *et al* [34] explored a link between physical oral sucking
443 behaviours (nutritive and non-nutritive) and the cognitive-linguistic aspect of speech sound

development, which, as Baker *et al* [34] acknowledged, is perhaps an unlikely association. Vieira *et al* [38] and Pereira *et al* [37] attempted to explore a possible relationship between physical sucking and the physical act of speech articulation, which may perhaps present a more probable association, and therefore should be the focus of further research in this area. However, it is important to consider that the nature of the chosen speech sound assessment method does not determine the type of SSD a child may have [57]. For example, children with phonological impairments, which may be identified using the phonology subtest of the DEAP [48] can also present with speech motor difficulties and vice versa. Therefore, while the four included studies report findings of atypical speech sound development, these cannot reliably be interpreted as identifying specific types of SSD.

4.2.2 Definition of Population

The lack of exclusion criteria in Barbosa *et al* [39] and Pereira *et al* [37] significantly weakens, in each case, the reliability of their findings and emphasises the importance of clearly defined sample populations for future research in this area. The decision by Baker *et al* [34] to exclude children whose parents were not concerned about their speech may be argued to risk the exclusion of otherwise potentially eligible children from the study on the basis of assumed parent awareness, knowledge or understanding [58].

4.2.3 Confounding Factors

The inclusion of comprehensive confounding factors identified from the literature is crucial in order to isolate the relationship between feeding, NNS and speech sound development as far as possible from these additional factors. Only by including and adjusting for these confounding factors in the statistical analysis can the relationship between NNS and speech sound development be described more accurately.

4.2.4. Missing Data

Unreported missing data was apparent in all but one [34] of the studies included in this review. This presents significant challenges for data interpretation and for the conclusions we are able to draw from the findings.

4.2.5. Exposure Measures – Nutritive and Non-nutritive Sucking

The nature of NNS behaviours vary significantly within and across cultures, with some children engaging only in these behaviours before sleep, while others show persistent behaviours throughout the day [59]. It is surprising that NNS sucking frequency was not reported in more of the papers. The authors of the current review would suggest that future research in this area include information on behaviour frequency as well as duration and causation (e.g., self-soothing behaviour at certain times of the day) in order to provide a comprehensive account of sucking behaviours, with which to then explore speech sound development outcomes in relation to early feeding methods.

4.2.6. Outcome Measures – Speech Sound Disorder (SSD)

While the need for inclusion of objective, formalised outcome measures for SSD in the examination of the relationship between feeding, sucking and speech sound development is evident, the nature of these assessments is also vital in establishing a clear speech sound profile for each child. Of the three studies in this review that completed objective speech sound assessments, all of them focussed on speech sounds at the single word level. There is a substantial and growing body of evidence that advocates the need for broader speech sound assessments to obtain a complete profile of a child's speech sound development; this includes collecting single sound, word, phrase level and connected speech [60]. In considering studies from a broader range of literature, such as those considering either, rather than both, feeding or NNS and speech sound development, no formal speech sound assessment approaches were identified [5, 61-63] and only one study, Baker *et al* [34], used the PCC measure. However, it is important to note the inherent weakness in using PCC as a measure to determine SSD type (e.g., participant assignment to PI group), as PCC scores would be lower among children with any type of SSD. The findings of these studies represent an incomplete picture with regard to patterns of feeding and NNS and any observable impact on speech sound development.

4.2.7. Managing Bias

There is significant inconsistency in the statistical reporting of results across the four included studies in this review. Indeed, the chosen statistical presentation of some of the results may be considered to risk reporting bias. As illustrated in section 3.2.1 above, ORs are reported by only two of the four studies [38-39], and only one of these consistently reported confidence intervals [38]. This paucity of accurate, consistent statistical reporting can lead to misrepresentation of the results, complicates the interpretation of the findings and can be misleading [64].

Recall bias is inherent in studies reliant on participant questionnaires for data collection, and applies to each of the four studies included in this review. A way to address this would be to carry out a prospective study, such as the Avon Longitudinal Study of Parents and Children [65].

4.3. Conclusions

This review has established that the current evidence around the relationship between infant feeding, NNS and speech sound development is very limited, of questionable quality and provides inconsistent findings. Greater clarity is required with regard to the nature of SSD being explored and coherence of approaches to outcome measurement. While the limited evidence examined within this review suggests some association between persistent NNS behaviors and the presence of SSD, the strength of this association is not clear. The question of a relationship between feeding type and SSD per se remains unanswered; however when duration is considered, there is some limited evidence for a protective effect of longer breastfeeding duration.

4.4. Potential Impact of Review Findings

The studies included in this review explore two distinct types of SSD: PI and articulation disorder. Several different classifications of SSD are presented in the literature [52, 66-67]. It has been suggested that an association between physical sucking and physical speech articulation may present a more logical relationship than that between physical sucking and cognitive speech sound processing [34]. The potential impact of the findings of this review is that further research is required to explore the

relationship between the physical aspects of sucking and speech sound development. This work should use more precise and detailed measures for sucking behaviours and speech sound development with explicit consideration of the different classifications of SSD. Fundamental to this is the careful consideration of the many documented confounding variables involved in this proposed association [17]. Future research should aim to provide clinically relevant findings that might be easily and usefully applied to the clinical settings where these populations receive support. An optimal outcome measurement approach would include detailed speech sound assessment from single sound imitation through to connected speech samples [60]. Ideally, these data would be captured through video recording in order to facilitate precise and accurate transcription by a qualified Speech and Language Pathologist (SLP). Audio recording of the data with the assessment administration and transcription completed by a qualified SLP is recommended as a minimum requirement for future research in this area.

5. Appendix

Appendix 1. PRISMA Flow Chart – Search Engine Searches

6. Supplementary Material

7. Statements

7.1 Acknowledgments

Not applicable.

7.2. Statement of Ethics

The authors have no ethical conflicts to enclose.

7.3. Disclosure Statement

The authors have no conflicts of interest to declare.

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7.5. Author Contributions

SB, TD and YW discussed the aim and objectives of this review. SB completed the initial searches and shortlisted at the abstract stage. SB and SH reviewed the included papers and completed the Quality Appraisal separately for subsequent discussion. SH was a major contributor in writing the final manuscript. All authors read, edited and approved the final manuscript.

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9. Figure Legends

Fig. 1. PRISMA Flow Chart for Traditional Database Searches.

Fig 2. PRISMA Flow Chart for Google Search

Table 1. Table 1. Inclusion and Exclusion Criteria

Table 2. Table 2. Quality Assessment Criteria and Scoring

Table 3. Table 3. Summary Table of Included Studies.

Table 4. Table 4. Summary Table of Exclusion Criteria Reported by Included Papers