

Appendices

Appendix A. Published systematic review (Burr *et al*, 2020)

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

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14 Short Title: SUCKING AND SPEECH SOUND DEVELOPMENT: SYSTEMATIC REVIEW

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30 **Keywords:** systematic review, speech sound development, speech disorder, infant feeding, non-
31 nutritive sucking.

32 **1. Abstract**

33 **Background**

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

40

41 **Summary**

42 Electronic databases (Pubmed, NHS CRD, EMBASE, MEDLINE) were searched using terms specific to
43 feeding, NNS and speech sound development. All methodologies were considered. Studies were
44 assessed for inclusion and quality by two reviewers. Of 1031 initial results, 751 records were screened
45 and five primary studies were assessed for eligibility, four of which were included in the review.

46 Evidence from the available literature on the relationship between feeding, NNS and speech sound
47 development was inconsistent and inconclusive. An association between NNS duration and SSDs was
48 the most consistent finding, reported by three of the four studies. Quality appraisal was carried out
49 using the Appraisal Tool for Cross-Sectional Studies (AXIS). The included studies were found to be of
50 moderate quality.

51

52 **Key Messages**

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

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

60

61 **2. Introduction**

62 **2.1. Background**

63 There is much discussion and debate in the current literature on the advantages of breastfeeding over
64 bottle-feeding, with positive cognitive outcomes often cited for language in later childhood [1-4].

65 However, evidence on the influence of feeding type on speech sound development is less readily
66 available (e.g., Fox et al. 2002 [5]). Infant feeding (breast, bottle and mixed feeding) and non-nutritive
67 sucking (NNS) (pacifier/digit sucking) are typically concurrent practices in the early lives of infants
68 across the world [6-8]; therefore it is important to consider both of these with regard to the impact on
69 speech sound development. Evidence for an indirect detrimental impact of NNS on speech sound
70 development is indicated with regard to dentition [9-10] and hearing loss resulting from otitis media
71 [11-12]; however the question of a potential direct impact of NNS on speech sound development is of
72 interest due to the shared physical oral mechanisms of these two processes.

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

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

107

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

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

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

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

120 **2.2. Methods**

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

126 **2.3. Identification of Selection Criteria**

127 The Booth & Fry-Smith [30] PICO model (population, intervention, comparison, outcome) guided the
128 development of the search strategy. The population of interest was children from birth into early
129 childhood, with or without identified SSD. Table 1 below lists the inclusion and exclusion criteria.
130 Papers that reported samples including children born prematurely (more than 15% of the total
131 sample), or those with diagnosed congenital disorders, identified learning difficulties, sensorineural
132 hearing loss, or populations that had received speech therapy intervention as part of the reported study
133 were excluded from the review as these factors could also impact on speech sound development. This
134 follows principles set out in similar systematic reviews in comparable cohorts (e.g., Roulstone et al.
135 2015 [31]). The intervention (behaviour) of interest was infant feeding, comparing outcomes in speech
136 sounds across three comparator interventions – breast-feeding, bottle-feeding and mixed feeding. A
137 second analysis considered presence or absence of NNS and its associations with speech sound
138 outcomes. Only papers reporting both feeding and NNS with regard to speech sound development were
139 included in this review. This systematic review of the current evidence base of journals and abstracts in
140 this topic area considered all methodologies and settings. Globally accessible articles were examined,

141 providing that they had been published, or were available, in the English language.

142 **2.3.1. Outcomes of Interest**

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

146

147 [Table 1 about here]

148 **2.4. Search Strategy**

149 The search strategy was designed in consultation with all authors and the search terms following a
150 review of the Cochrane database, PROSPERO and database of abstracts of reviews of effectiveness.

151 Discussions with a specialist speech and language pathologist working with children with SSD
152 facilitated the identification of specific search terms relevant to all possible and appropriate
153 terminology for speech sound development and SSD. A combination of 'free text' terms with Boolean
154 operators and truncations were used as follows:

155 **2.4.1 Feeding Search Term**

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

157 **2.4.2 Non-Nutritive Sucking Search Term**

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

159 **2.4.3 Speech Search Term**

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

163 **2.5. Findings of the Search Process**

164 **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.

191 In addition to the above searches, one unpublished paper [34], identified through discussions with

192 review colleagues, was included in the screening process and subsequently retained. A total of four
193 papers were included in the full review: two identified from traditional database searches, one from
194 Google and one unpublished paper.

195

196 [Figure 2 about here]

197 **Figure 2. PRISMA Flow Chart for Google Search Engine**

198

199 **2.6. Search Validation**

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

205 **2.7. Data Extraction**

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

210 **2.8. Quality Appraisal**

211 Selection of the quality appraisal tool was undertaken once the final list of included papers had been
212 obtained and reviewed for their methodology. All four papers used a cross-sectional study design and
213 subsequently the Appraisal Tool for Cross-Sectional Studies (AXIS) was used by SB and SH [36]. This
214 tool was selected as the most appropriate for assessing the quality of the included papers because it has
215 been specifically designed for the critical and quality appraisal of cross-sectional studies. The AXIS
216 comprises 20 questions to appraise each paper's introduction, methods, results, discussion and other

217 issues related to bias and ethical conduct. The authors assigned a score to each of the categories. 2 for
218 papers that clearly provide the information required by the AXIS tool, 1 if this information is partially
219 present, but not clearly stated, and 0 if it is not present at all. This led to a maximum possible score of
220 40 on the AXIS. The quality appraisal of the included papers was completed separately by SH and SB
221 and scoring consensus was reached following discussion.

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

225 [Table 2 about here]

226

227 **2.8. Data Synthesis**

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

230 **3. Results**

231 **3.1. Review of the Data**

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

233 **3.1.1. Statistical Techniques**

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

241 **3.1.2. Methodological Approaches**

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

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

257 **3.1.3. Sample Populations**

258 Details of the population samples for each study are provided in Table 3. Only two of the four papers
259 [34, 38] reported any use of exclusion criteria in their sample definitions, and only one of these, hearing
260 loss, was common to both studies (see Table 4). Baker *et al* [34] reported the most comprehensive
261 exclusion criteria, including genetic, medical and developmental factors known to have some
262 association with SSD.

263 **Table 3. Summary Table of Included Studies.**

264 *[Table 3 about here]*

265

266 *[Table 4 about here]*

267

268 **3.1.4. Definition of SSD**

269 A key challenge for this review was the disparity in what is meant by the term ‘Speech Sound Disorder’
270 between papers. Barbosa *et al* [39] used the terms ‘speech disorder(s)’ and ‘speech processing’, the
271 former of which they broadly describe as having the potential to “*impair communication and*
272 *literacy*”^{[39](p2)}. Specific reference to distinct types of SSD was not made; however through their use of
273 the Brazilian speech sound assessment TEPROSIF [40] to “*determine the type and number of errors in*
274 *the child-age related phonological processes*”^{[39](p3)}, the implication was to focus on phonological
275 impairment (PI). Baker *et al* [34] were more explicit in stating their specific focus on children with
276 diagnosed PI, and defined the group as presenting with “*one or more age-inappropriate common*
277 *phonological error patterns [...] with no evidence of motor speech involvement*”^{[34](p7)}. As Baker *et al* [34]
278 themselves acknowledged, “*PI is presumed to be a cognitive-linguistic difficulty involving a difficulty*
279 *abstracting rules about the phonological system, and the abstract phonological representation of speech*
280 *rather than an articulation difficulty. As such, it is reasonable to suggest that non-nutritive sucking habits*
281 *would be unrelated*”^{[34](p11)}. Pereira *et al* [37] made reference to both ‘speech disorder(s)’ and ‘speech
282 changes’ and acknowledged that they did not distinguish between types of SSD. They provided some
283 definition of their application of the term ‘speech disorders’ as “*those reported by the parents and/or*
284 *guardians with respect to the production of the phonemes /t/, /d/, /n/, /l/, /r/, /s/, and /z/, considered*
285 *comprehensively as they are associated with alterations in the SS [stomatognathic system]*”^{[37](p2)}. The
286 repeated emphasis within this paper on the structures and functions of the stomatognathic system,
287 defined by the authors as comprising the functions of suction, swallowing, mastication, respiration and
288 speech^{[37](p.2)}, indicated the author’s intention to explore ‘speech disorders’ relating to articulation,
289 rather than those that are cognitive-linguistic in nature. Vieira *et al* [38] also referred to ‘speech
290 disorders’, ‘speech changes’ and the SS, as well as ‘speech impairment’. They defined their case group
291 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
344 [38]. As scoring information was not presented for the case or control groups, it is not possible to
345 determine or assess the severity of children’s speech sound errors within the case group. Baker *et al*
346 [34] provided a clear description and explanation of their selected published assessment tool, the
347 Diagnostic Evaluation of Articulation and Phonology (DEAP) [48]. Following administration of the

348 Phonology Assessment single word naming test, children were assigned to one of four groups based on
349 their obtained DEAP standard score, percentage of consonants correct (PCC) score and error patterns.
350 Only data from the PI group were included in the study. Children assigned to the PI group obtained a
351 DEAP standard score of 6 or less based on their PCC score. A score of 7-13 is understood to fall within
352 the normal range [48].

353 **3.3. Managing Bias**

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

355 **3.3.1. Sample Baseline Imbalances**

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

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

360 In Vieira *et al* [38] there is a reporting error in the paper. They reported equal overall sample sizes for
361 the case and control groups; however, manual calculations of the group totals from the data presented
362 in their analysis^{[38](p.1362)} indicate a marked group imbalance (see Table 1). There is also a significant
363 gender imbalance within the total sample, which contains 73% more males than females. Vieira *et al*
364 [38] briefly acknowledged this imbalance in their discussion. Baker *et al* [34] also reported a sample
365 gender imbalance, with 55% more males than females in their PI group. The SSD prevalence figures in
366 the wider literature also show a tendency for more boys than girls [17, 49-52].

367 Barbosa *et al* [39] included in their sample children born prematurely (n=19) and, as acknowledged by
368 the authors, this population are significantly more likely to present with “increased risk of
369 developmental problems with speech”^{[39](p4)}. The inclusion of this population, which constitutes 15% of
370 the total study sample, may have some impact on the results as they potentially comprise almost 1/3 of
371 the reported ‘below normal’ group. Prematurity is often cited in the wider literature as being
372 associated with speech sound difficulties in later development [53-55].

373 **3.3.2. Recall Bias**

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

381 **3.4. Summary of Findings from Included Papers**

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

385 **3.4.1. Feeding Type and Speech Sound Development**

386 Barbosa *et al* [39] suggested an association between bottle feeding and SSD in preschool children, such
387 that delaying bottle use until after age nine months appeared to show some small protective effect (OR:
388 0.32, 95% CI: 0.10-0.98). Pereira *et al* [37] also reported a significant correlation between speech
389 sound difficulties and bottle feeding ($p=0.056$). This may indicate a liberal application of their reported
390 adopted 5% significance level^{[37](p.2)}. Vieira *et al* [38] found no significant association between feeding
391 type and SSD. Baker *et al* [34] similarly found no association between feeding type and the presence or
392 absence of SSD (specifically PI).

393 **3.4.2. Duration of Feeding Type and Speech Sound Development**

394 Pereira *et al* [37] and Vieira *et al* [38] both collected data on duration of feeding method and speech
395 sound development but did not report on these data within their papers. Baker *et al* [34] suggested a
396 trend whereby longer breastfeeding duration is associated with higher percentage consonants correct
397 (PCC) scores, resulting in more accurate speech sound production for spoken words. Barbosa *et al* [39]
398 reported that children scoring as normal or 1 standard deviation above normal on the “Test para

399 evaluar los procesos fonológicos de simplificación” (TEPROSIF) speech sound assessment tended to
400 have been breastfed for longer than those scoring below expectation for their age [40]. They asserted
401 that delaying bottle feeding until after age 9 months may be to some extent a protective factor against
402 subsequent SSD (OR: 0.32, 95% CI: 0.10-0.98).

403 **3.4.3. Non-nutritive Sucking and Speech Sound Development**

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

410 **3.4.4. Duration of Non-nutritive Sucking and Speech Sound Development**

411 Baker *et al* [34] reported that, while the relationship between NNS and presence of SSD was non-
412 significant, they did identify a trend between longer pacifier use and lower PCC scores. Barbosa *et al*
413 [39] reported that children who used a pacifier for more than three years were much more likely to
414 present with below normal speech sound development (OR: 3.4, 95% CI: 1.08-10.81). Pereira *et al* [37]
415 suggested that using a pacifier for less than one year was not associated with speech sound difficulties,
416 whereas digit sucking persisting for up to four years was positively correlated with the presence of SSD
417 (p= 0.012). Vieira *et al* [38] found no association between NNS and SSD.

418 **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
444 development, which, as Baker *et al* [34] acknowledged, is perhaps an unlikely association. Vieira *et al*
445 [38] and Pereira *et al* [37] attempted to explore a possible relationship between physical sucking and
446 the physical act of speech articulation, which may perhaps present a more probable association, and
447 therefore should be the focus of further research in this area. However, it is important to consider that
448 the nature of the chosen speech sound assessment method does not determine the type of SSD a child
449 may have [57]. For example, children with phonological impairments, which may be identified using the
450 phonology subtest of the DEAP [48] can also present with speech motor difficulties and vice versa.

451 Therefore, while the four included studies report findings of atypical speech sound development, these
452 cannot reliably be interpreted as identifying specific types of SSD.

453 **4.2.2 Definition of Population**

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

460 **4.2.3 Confounding Factors**

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

466 **4.2.4. Missing Data**

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

470 **4.2.5. Exposure Measures – Nutritive and Non-nutritive Sucking**

471 The nature of NNS behaviours vary significantly within and across cultures, with some children
472 engaging only in these behaviours before sleep, while others show persistent behaviours throughout
473 the day [59]. It is surprising that NNS sucking frequency was not reported in more of the papers. The
474 authors of the current review would suggest that future research in this area include information on
475 behaviour frequency as well as duration and causation (e.g., self-soothing behaviour at certain times of

476 the day) in order to provide a comprehensive account of sucking behaviours, with which to then
477 explore speech sound development outcomes in relation to early feeding methods.

478 **4.2.6. Outcome Measures – Speech Sound Disorder (SSD)**

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

493 **4.2.7. Managing Bias**

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

500 Recall bias is inherent in studies reliant on participant questionnaires for data collection, and applies to
501 each of the four studies included in this review. A way to address this would be to carry out a

502 prospective study, such as the Avon Longitudinal Study of Parents and Children [65].

503 **4.3. Conclusions**

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

512 **4.4. Potential Impact of Review Findings**

513 The studies included in this review explore two distinct types of SSD: PI and articulation disorder.
514 Several different classifications of SSD are presented in the literature [52, 66-67]. It has been suggested
515 that an association between physical sucking and physical speech articulation may present a more
516 logical relationship than that between physical sucking and cognitive speech sound processing [34].
517 The potential impact of the findings of this review is that further research is required to explore the
518 relationship between the physical aspects of sucking and speech sound development. This work should
519 use more precise and detailed measures for sucking behaviours and speech sound development with
520 explicit consideration of the different classifications of SSD. Fundamental to this is the careful
521 consideration of the many documented confounding variables involved in this proposed association
522 [17]. Future research should aim to provide clinically relevant findings that might be easily and
523 usefully applied to the clinical settings where these populations receive support. An optimal outcome
524 measurement approach would include detailed speech sound assessment from single sound imitation
525 through to connected speech samples [60]. Ideally, these data would be captured through video
526 recording in order to facilitate precise and accurate transcription by a qualified Speech and Language
527 Pathologist (SLP). Audio recording of the data with the assessment administration and transcription

528 completed by a qualified SLP is recommended as a minimum requirement for future research in this
529 area.

530 **5. Appendix**

531 Appendix 1. PRISMA Flow Chart – Search Engine Searches
532

533 **6. Supplementary Material**

534

535 **7. Statements**

536 **7.1 Acknowledgments**

537 Not applicable.

538 **7.2. Statement of Ethics**

539 The authors have no ethical conflicts to enclose.

540 **7.3. Disclosure Statement**

541 The authors have no conflicts of interest to declare.

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545 **7.5. Author Contributions**

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

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779 **9. Figure Legends**

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

781 Fig 2. PRISMA Flow Chart for Google Search

782 Table 1. Table 1. Inclusion and Exclusion Criteria

783 Table 2. Table 2. Quality Assessment Criteria and Scoring

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

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

786 **Table 1. Inclusion and Exclusion Criteria**

INCLUSION CRITERIA	EXCLUSION CRITERIA
<ul style="list-style-type: none"> • Children aged from birth onwards, with or without identified SSD • Report of infant feeding method AND presence or absence of non-nutritive sucking behaviours • Report of speech sound development outcome • A maximum of 15% of sample population born prematurely¹ • All methodologies and settings • International papers • Published in English Language 	<ul style="list-style-type: none"> • Children diagnosed with: <ul style="list-style-type: none"> ○ congenital disorders ○ identified learning difficulties, or ○ sensorineural hearing loss • Populations that had received speech pathology prior to or as part of the reported study

787 ¹ Preterm birth classified as before 37 weeks completed gestation. World Health Organisation (WHO).
 788 <https://www.who.int/news-room/fact-sheets/detail/preterm-birth> Accessed 16/10/2019.
 789

790

791 **Table 2. Quality Assessment Criteria and Scoring**

AXIS Quality Assessment Criteria		Barbosa et al (2009)	Vieira et al (2016)	Pereira et al (2017)	Baker et al (2018)
INTRODUCTION					
1	Were the aims/objectives of the study clear?	2	2	2	2
METHODS					
2	Was the study design appropriate for the stated aim(s)?	2	2	2	2
3	Was the sample size justified?	0	0	0	0
4	Was the target/reference population clearly defined? (Is it clear who the research was about?)	0	2	0	2
5	Was the sample frame taken from an appropriate population base so that it closely represented the target/reference population under investigation?	2	2	2	2
6	Was the selection process likely to select subjects/participants that were representative of the target/reference population under investigation?	2	2	2	2
7	Were measures undertaken to address and categorise non-responders?	0	0	0	0
8	Were the risk factor and outcome variables measures appropriate to the aims of the study?	2	1	1	2
9	Were the risk factor and outcome variables measured correctly using instruments/measurements that had been trialled, piloted or published previously?	1	1	0	1
10	Is it clear what was used to determine statistical significance and/or precision estimates? (e.g. p-values, confidence intervals)	2	2	2	2
11	Were the methods (including statistical methods) sufficiently described to enable them to be repeated?	2	2	2	2
RESULTS					
12	Were the basic data adequately described?	2	2	1	2
13	Does the response rate raise concerns about non-response bias?	1	1	1	0
14	If appropriate, was information about non-responders described?	N/A	N/A	N/A	N/A
15	Were the results internally consistent?	2	0	0	2
16	Were the results presented for all the analyses described in the methods?	2	2	2	2
DISCUSSION					
17	Were the authors' discussions and conclusions justified by the results?	1	0	1	2
18	Were the limitations of the study discussed?	2	0	2	2
OTHER					
19	Were there any funding sources or conflicts of interest that may affect the authors' interpretation of the results?	0	0	0	0
20	Was ethical approval or consent of participants obtained?	2	2	2	2
TOTAL (max 40)		27	23	22	29

792

793 **Table 3. Summary Table of Included Studies**

References	Study Type	Participants / Setting	Measures / Outcomes	Statistical Analysis	Key Findings
Barbosa et al 2009 ^[39]	Cross-sectional	128 children aged 37-70 months 59 females 69 males (3 years (n=58); 4 years (n=49); 5 years (n=21)). From three public kindergartens in Punta Arenas (Patagonia), Chile. Feeding method and non-nutritive sucking behaviours reported for all participants.	Self-administered parent questionnaire to provide information on feeding and pacifier/digit sucking history. TEPROSIF ¹ standardised phonological assessment for children aged 3-7 years.	SPSS ² 13.0 Chi-square tests compared age and categorical characteristics, and TEPROSIF classifications and categorical characteristics. Two-sided p-values of association between these variables. Multivariable logistic regression of associations between potential risk factors and SSD ³ . Adjusted and unadjusted models (age and gender) for OR ⁴ and 95% CI ⁵ .	1) Pacifier use, finger sucking and bottle feeding are associated with SSD in preschool children. 2) Delaying bottle feeding until 9 months may be protective from subsequent SSD (OR: 0.32, 95% CI ⁴ : 0.10-0.98). 3) Children who sucked their fingers were 3x more likely to have SSD (OR: 2.99, 95% CI: 1.10-8.00, p = 0.02). 4) Using a pacifier for >3years increased likelihood of SSD threefold (OR: 3.4, 95% CI: 1.08-10.81).
Vieira et al 2016 ^[38]	Case-Control	273 children aged 36 -71 months and enrolled at one of 15 state preschools	Self-administered parent questionnaire to provide information on feeding and pacifier/digit sucking	STATA/SE ⁶ 9.0 ORs with 95% CIs and p values.	1) Only gender was significantly associated with SSD (OR = 1.79; CI 95% = 1.03-3.10; p =

		<p>in Recife, Brazil.</p> <p>Males (n=173), Females (n=100).</p> <p><i>Case Group</i> (n=108): presented with speech alterations, as reported by parents.</p> <p><i>Control Group</i> (n=165): no speech alterations reported by parents.</p> <p>Feeding method and non-nutritive sucking behaviours reported for all participants.</p>	<p>history.</p> <p>ABFW Children's Language Test Phonological Evaluation Protocol Subtest. Standardised for Brazilian children aged 2-12 years.</p>		<p>0.038).</p> <p>2) No significant association between feeding, NNS⁷ and SSD.</p> <p>3) No significant association between SES⁸ and SSD (due to authors' claimed homogeneity of sample).</p> <p>4) No significant association between age and "speech alterations", although most of Case group were age 3 years.</p>
Pereira et al 2017 ^[37]	Cross-sectional	<p>(Parents of) 289 children aged 1-12 years assisted at a family health strategy unit in northern district of Porto Alegre, Brazil.</p> <p>Male (n=145), Female (n=144).</p> <p>Feeding method and non-nutritive sucking behaviours reported for all participants.</p>	Self-administered parent questionnaire.	SPSS 19.0 Chi-square for p values.	<p>1) Correlation between SSD and bottle-feeding (p = 0.056).</p> <p>2) Correlation between pacifier use and SSD (p= 0.046).</p> <p>3) Pacifier use <1 year not associated with SSD.</p> <p>4) Correlation between thumb sucking for up to 4 years duration and SSD (p= 0.012).</p>
Baker et al	Cross-	199 Australian-English speaking	Self-administered	Statistical package not	1) Duration of breastfeeding and PI not

2018 ^[34]	sectional	<p>children aged 48-66 months with and without PI.</p> <p>Males (n=121) females (n=78).</p> <p>Children enrolled on the Sound Start study (an RCT⁹ for children with PI¹⁰).</p> <p>Meeting criteria following DEAP assessment for PI only or 'No Impairment'</p> <p>Feeding method and non-nutritive sucking behaviours reported for all participants.</p>	<p>parent questionnaire.</p> <p>DEAP¹¹ phonology assessment. PCC¹² score.</p>	<p>reported.</p> <p>Chi-square for p values.</p>	<p>associated (p=0.055), nor severity (p=0.396).</p> <p>2) Longer breastfeeding duration showed higher PCC scores in PI group.</p> <p>3) Duration of pacifier use and PI not associated (p=0.745), nor severity (p=0.106).</p> <p>4) Longer pacifier duration showed lower PCC scores.</p>
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794 ¹TEPROSIF = Test para evaluar los procesos fonológicos de simplificación; ²SPSS = Statistical Package for
795 the Social Science; ³SSD = Speech Sound Disorder; ⁴OR = Odds Ratio; ⁵CI = Confidence Interval; ⁶STATA =
796 general purpose statistical software package; ⁷NNS = Non-nutritive sucking; ⁸SES = socioeconomic status;
797 ⁹RCT = randomised control trial; ¹⁰PI = phonological impairment; ¹¹DEAP = Diagnostic Evaluation of
798 Articulation and Phonology; ¹²PCC = Percentage Consonants Correct.

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

Exclusion Criteria Reported by Included Papers	Barbosa et al (2009)	Vieira et al (2016)	Pereira et al (2017)	Baker et al (2018)
No parent/carer concern	-	-	-	✓
Diagnosed Developmental Delay	-	-	-	✓
Congenital Malformations	-	✓	-	-
Physical or mental disability impacting speech development	-	✓	-	-
Hearing loss	-	✓	-	✓
Cleft lip and/or palate	-	-	-	✓
Articulation impairment only	-	-	-	✓
Childhood Apraxia of Speech	-	-	-	✓
Diagnosed Childhood Dysarthria	-	-	-	✓

800

Figure 1: PRISMA Flow Chart Traditional Database Searches



PRISMA 2009 Flow Diagram: Database Searches

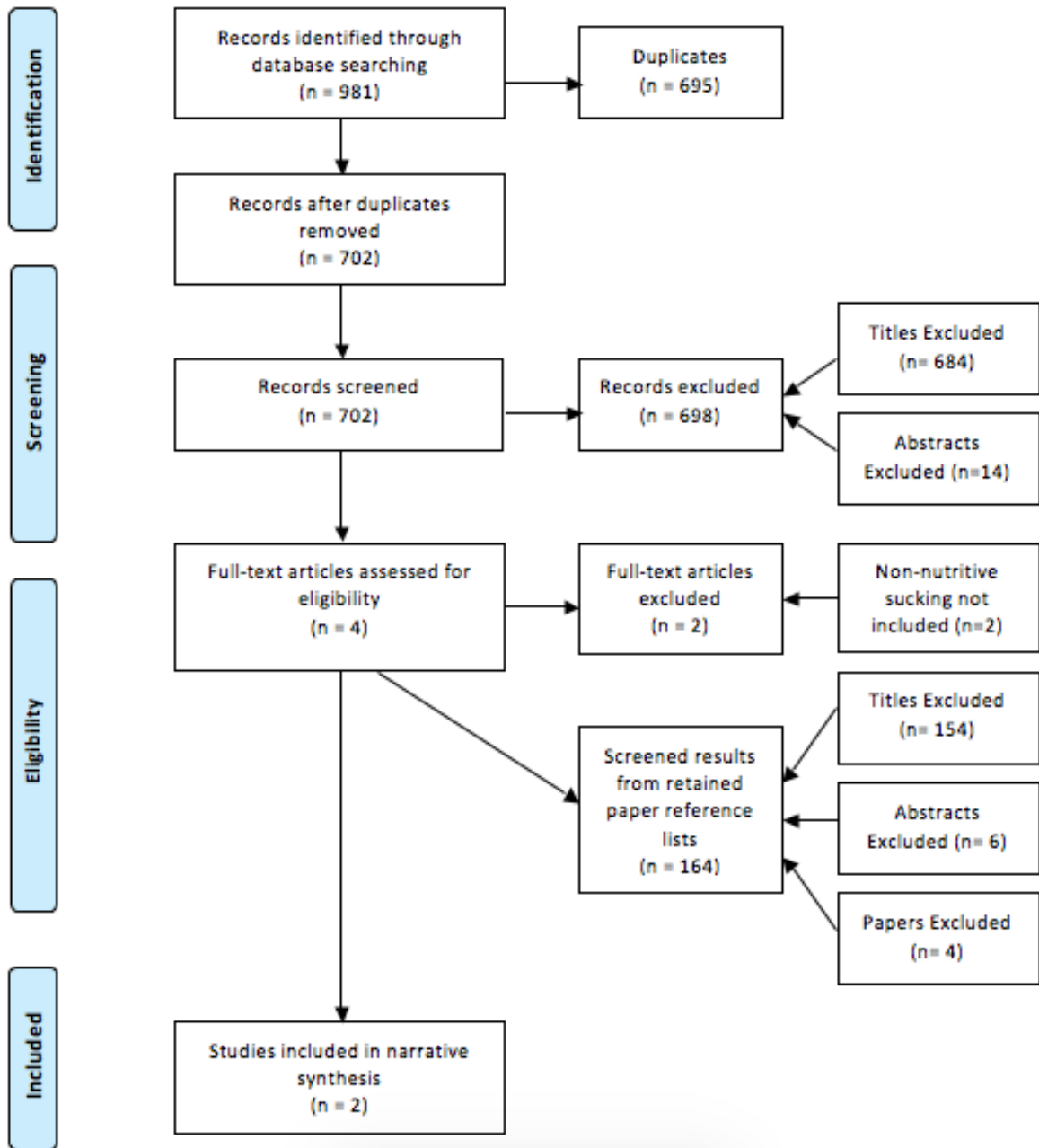
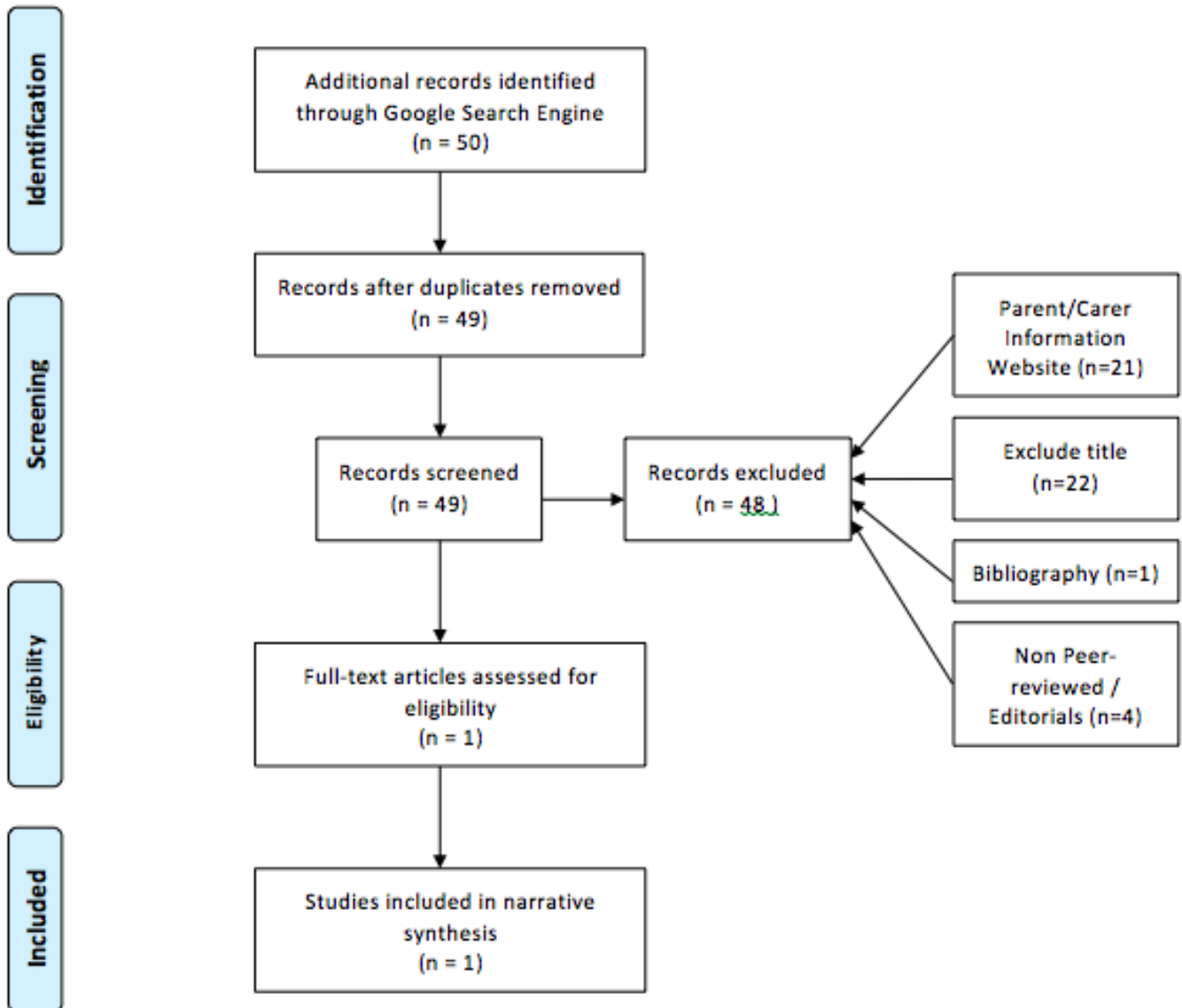


Figure 2: PRISMA Flow Chart Google Search Engine



PRISMA 2009 Flow Diagram: Search Engine Results



Appendix B. ALSPAC proposal approval

Email received 19/04/2016 from ALSPAC Committee (alspac-exec@bristol.ac.uk)

Dear Mrs Samantha Burr,

The Executive Committee are pleased to approve your proposal entitled 'What is the effect of different feeding methods and non-nutritive sucking behaviours on child speech development?' subject to the appropriate costs being covered – you may wish to refer to section 1.3 of the ALSPAC access policy (<http://www.bristol.ac.uk/alspac/researchers/data-access/>) for further information on possible costs. Please note this approval is also subject to ethical approval and possible input from our participant advisory panel. The reference number for your proposal is B2658 (please quote this number on all future correspondence).

The University of Bristol finance department need at least one month to agree and sign off costs - you may wish to refer to the access policy for further information. I have copied in Ross Robinson (Deputy Executive Director) and Melanie Lewcock (COCO90s project manager) who will be able to assist you with providing exact costs for ALSPAC.

Please note that ALSPAC fully supports the Wellcome Trust and the RCUK policies on open access. It is your responsibility to ensure that any papers you publish resulting from this project comply with these. Please see section 6.6.1.1 of the access policy for further details (<http://www.bristol.ac.uk/alspac/researchers/data-access/>).

Can you please ensure you send me the final grant proposal as submitted for our records. I will be monitoring the proposals process and I would therefore appreciate any updates regarding this project.

The approved project summary will be listed on ALSPAC's website and your name and details of your proposal may be tweeted.

Regards,

Becky Allen

On behalf of the ALSPAC Executive

Please note as of 1st April 2015 we have updated our costs, please see our ALSPAC access policy for more details (<http://www.bristol.ac.uk/alspac/researchers/data-access/>)

Appendix C. . List of objects for age 25 month speech assessment taken from Roulstone et al (2010)

Object naming assessment, adapted from Pagel Paden *et al*, (1987)

<i>Target</i>	<i>Response</i>	<i>Velar</i>	<i>Consonant cluster</i>	<i>Liquid</i>	<i>Fricative</i>	<i>Postvocalic consonant</i>
Brick		1	1	1		1
Brush			1	1	1	1
Clock		2	1	1		1
Cup		1				1
Fish					2	1
Flag		1	1	1	1	1
Flower		1	1	1	1	1
Fork		1			1	1
Glasses		1	1	1	2	1
Light				1		1
Plate			1	1		1
Quack		2	1	1		1
Sock		1			1	1
Slide			1	1	1	1
Snake		1	1		1	1
Spoon			1		1	1
Potential occurrences		11	11	10	12	15

Appendix D. Strand One Parts B & C: ALSPAC 61 month clinical speech assessment single word target list (*n*=20)

butterfly	calculator	chips	clocks
glasses	helicopter	hippopotamus	photograph
present	spaghetti	string	Three
pyjamas	squirrel	telephone	Toothbrush
skirt	starfish	television	yellow

Appendix E. Strand One Part B: bar graphs for consonant error frequencies at age 25 months

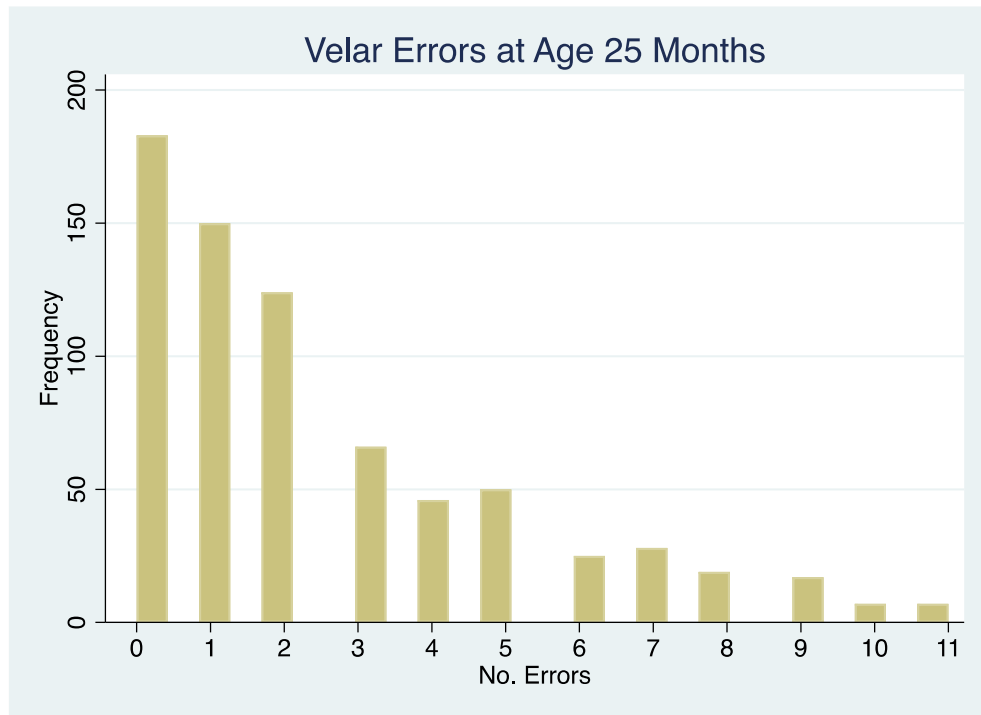


Figure 1. Bar graph of velar errors at age 25 months

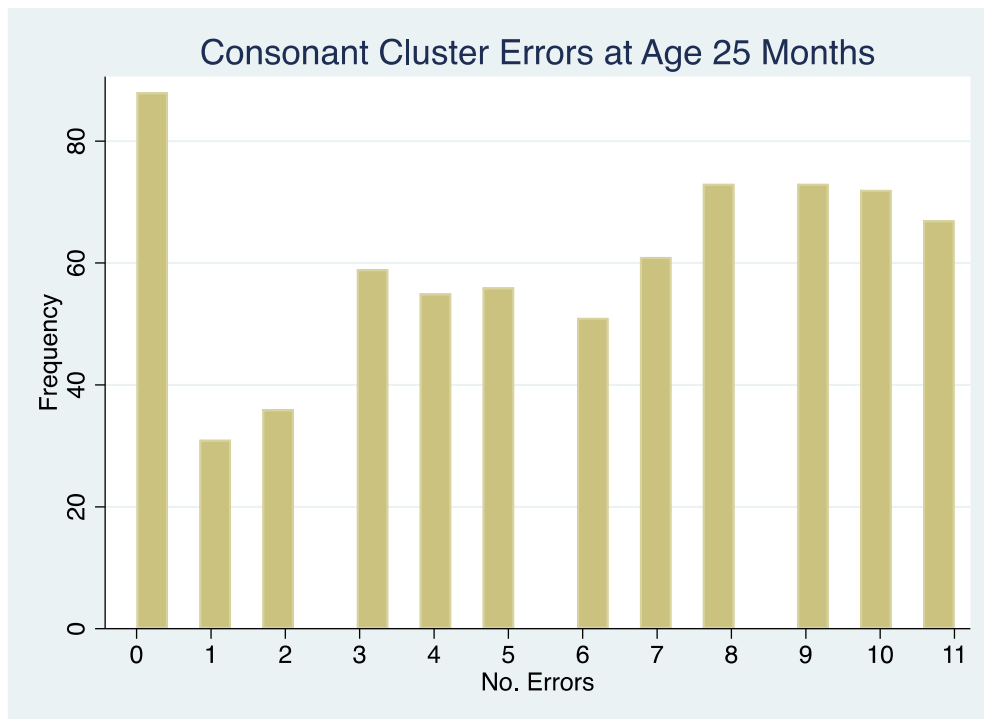


Figure 2. Bar graph of consonant cluster errors at age 25 months

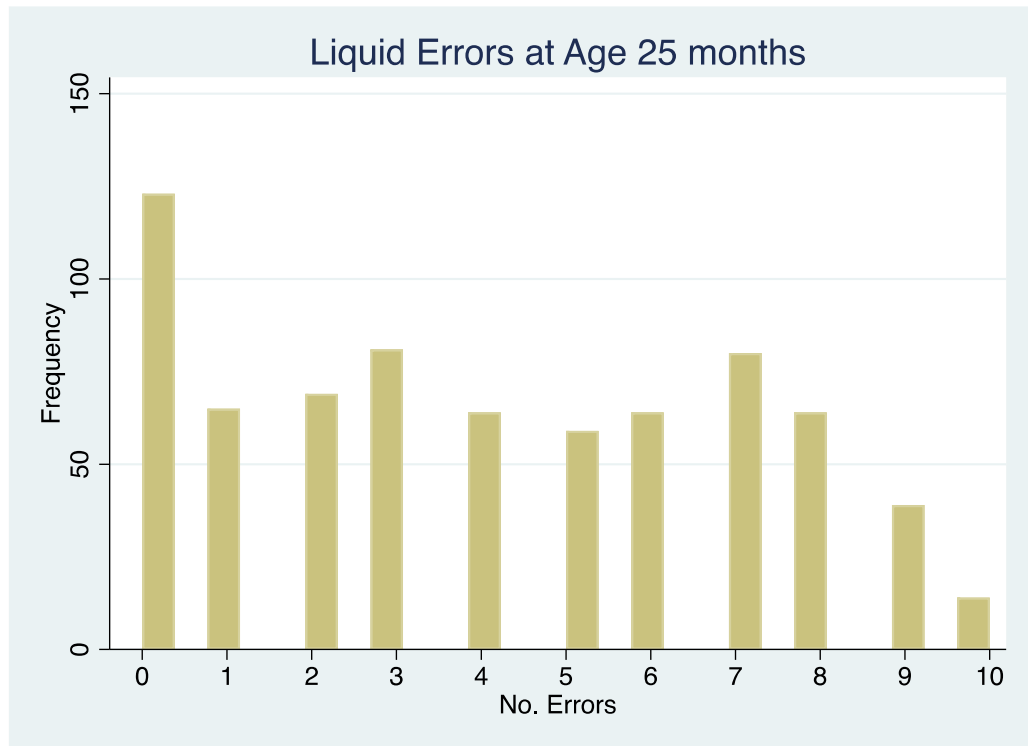


Figure 3. Bar graph of liquid errors at age 25 months

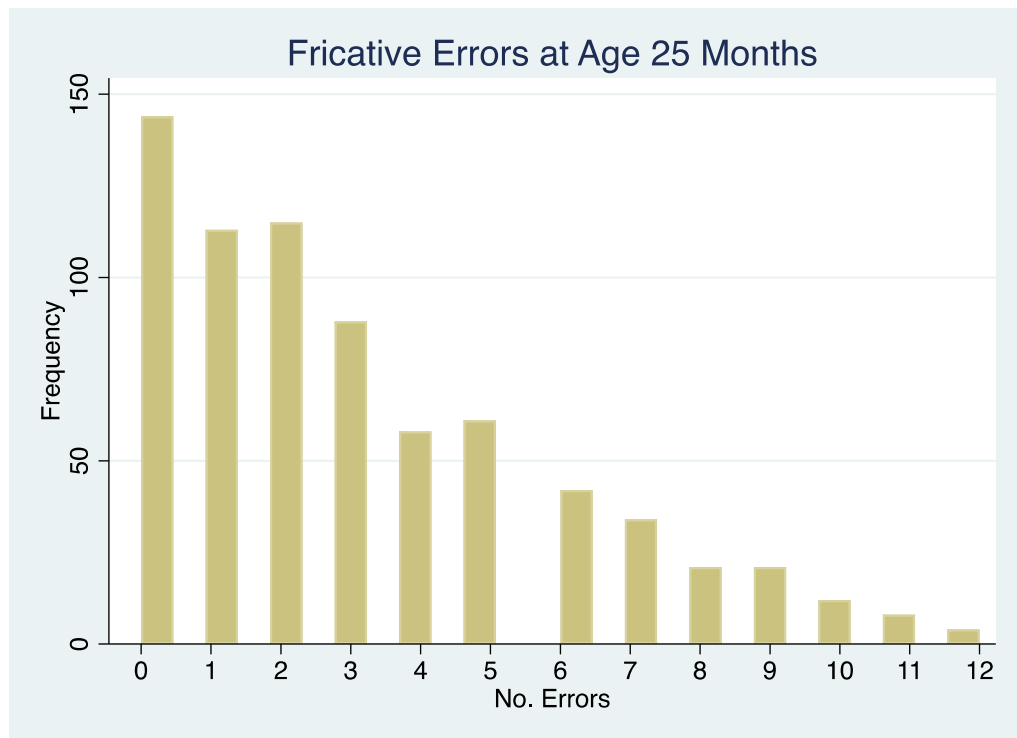


Figure 4. Bar graph of fricative errors at age 25 months

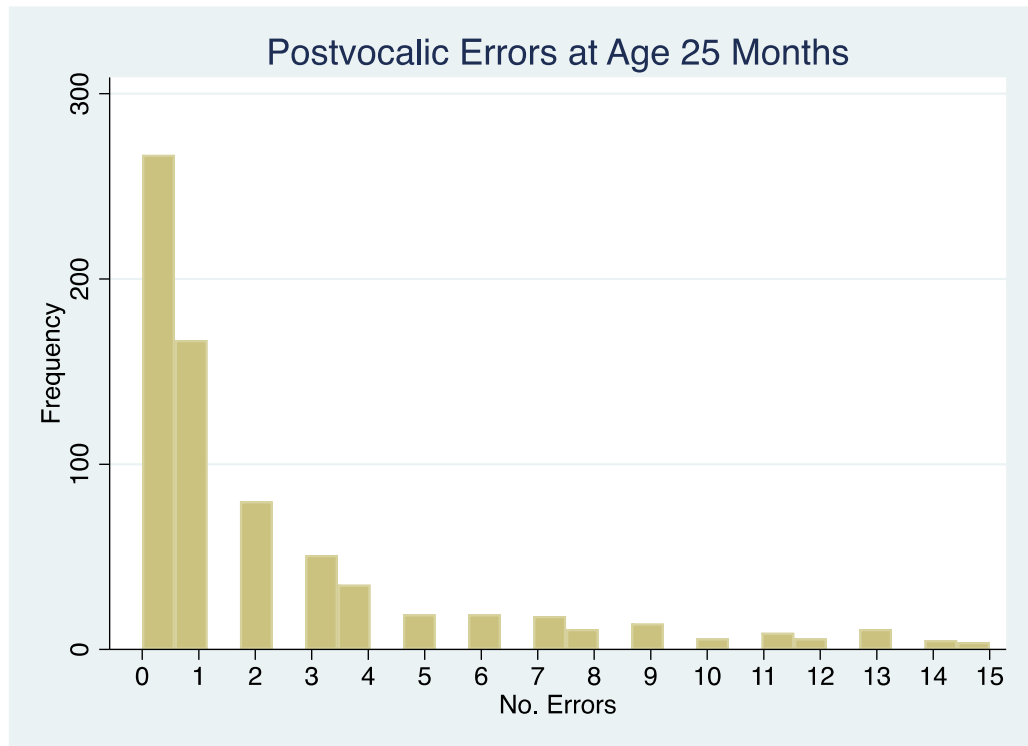


Figure 5. Bar graph of postvocalic errors at age 25 months

Appendix F. Strand One Part B: bar graphs for speech sound error types at age 61 months

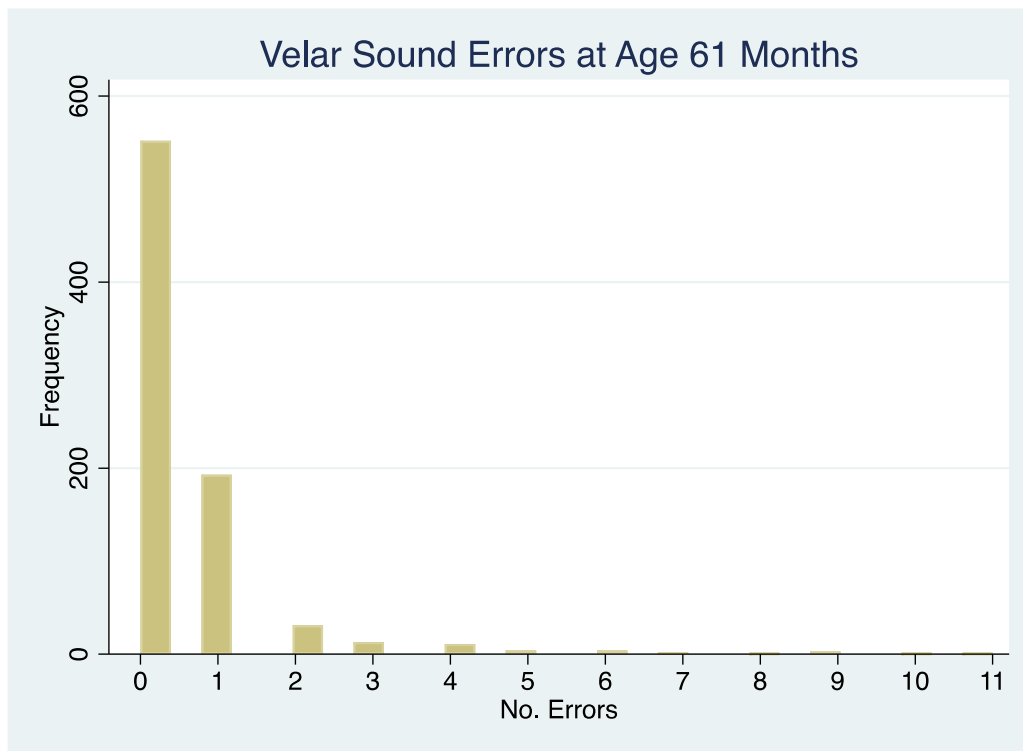


Figure 1. Bar graph of velar errors at age 61 months

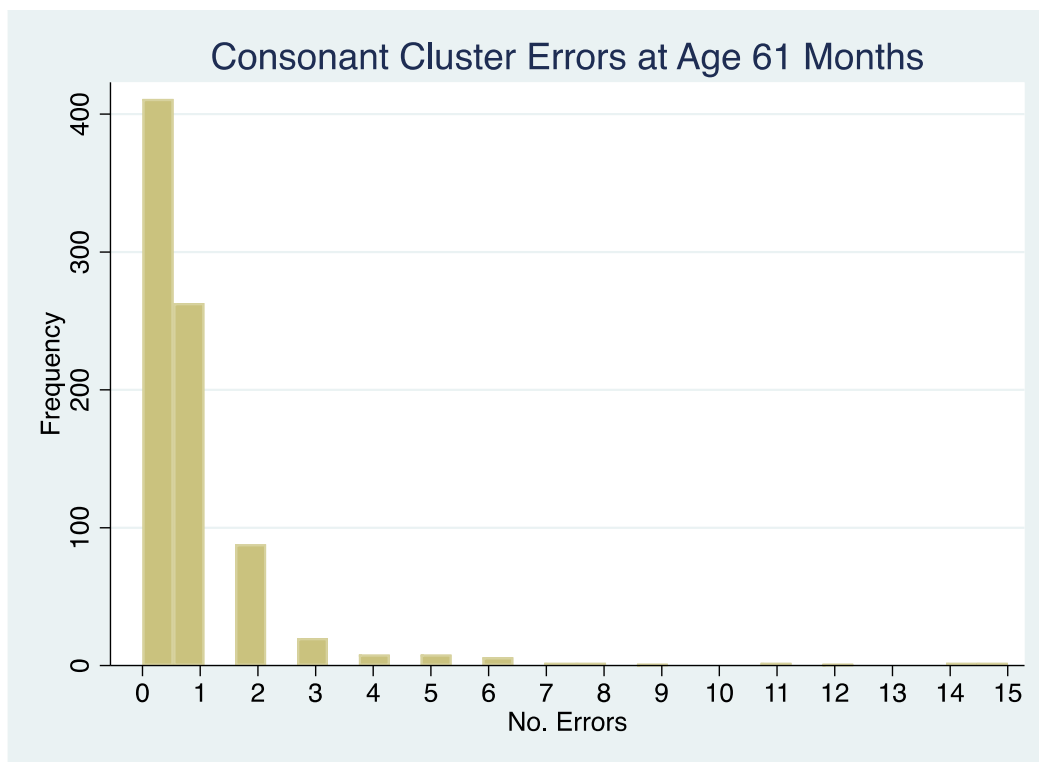


Figure 2. Bar graph of consonant cluster errors at age 61 months

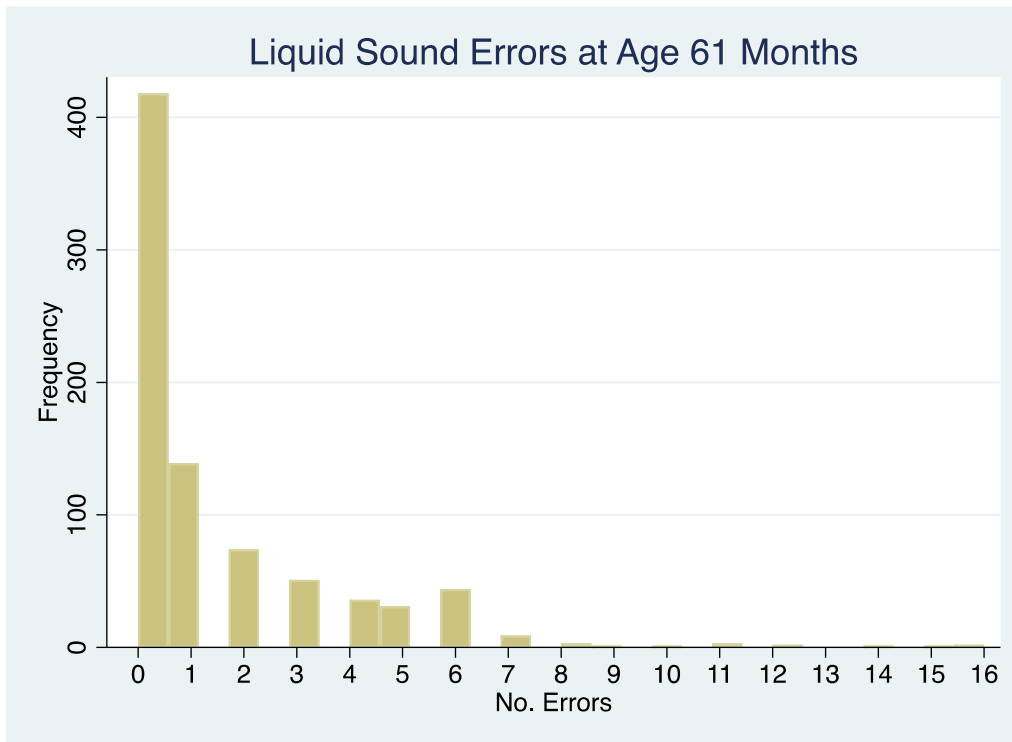


Figure 3. Bar graph of liquid errors at age 61 months

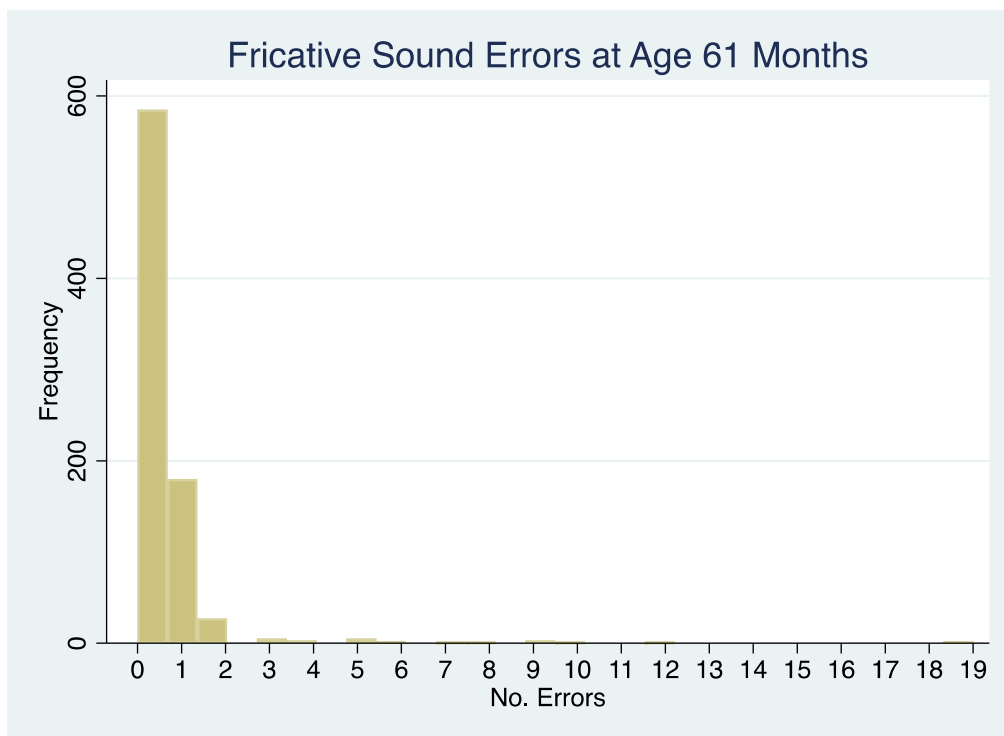


Figure 4. Bar graph of fricative errors at age 61 months

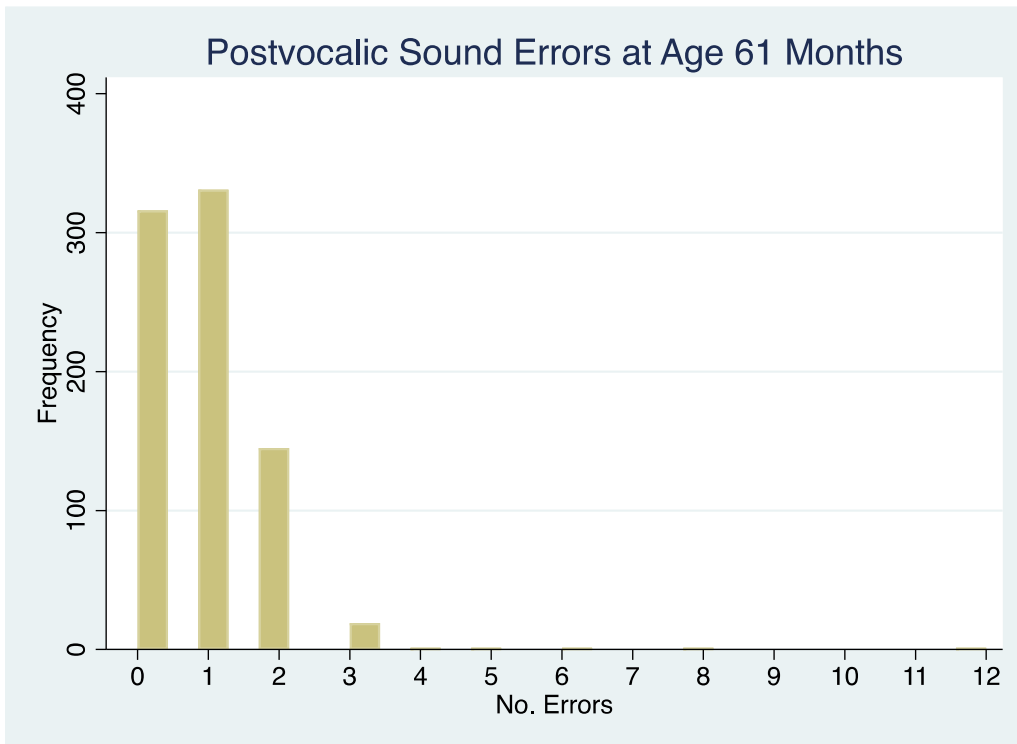


Figure 5. Bar graph of postvocalic errors at age 61 months

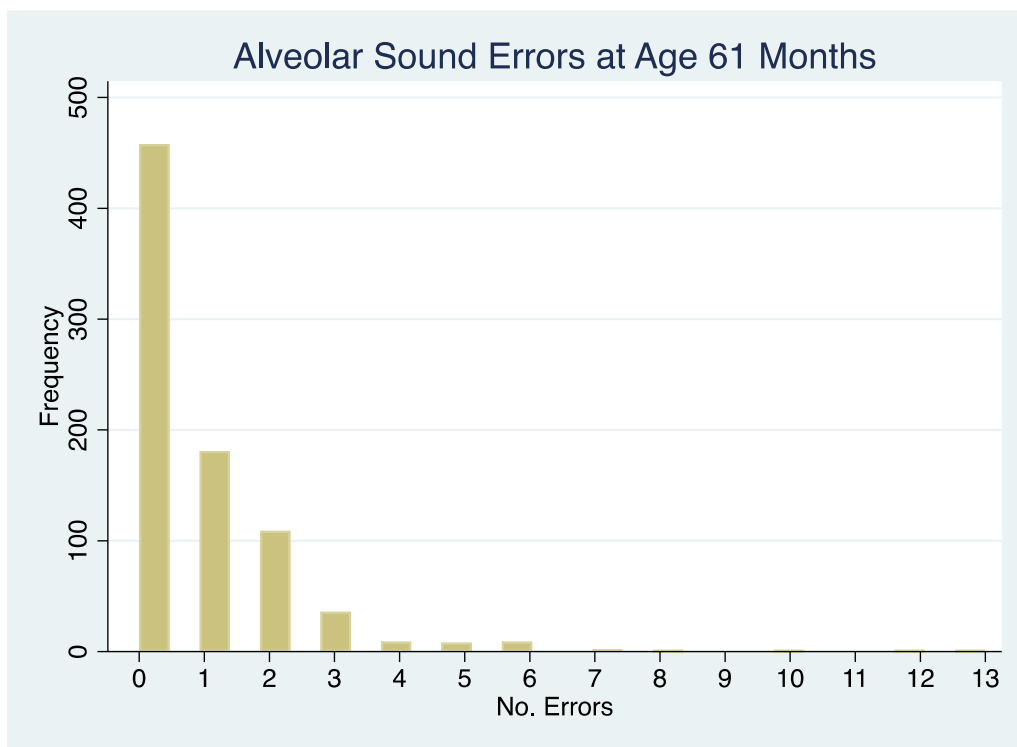


Figure 6. Bar graph of alveolar errors at age 61 months

Appendix G. Strand Two: standard operating procedure (SOP) ALSPAC-G2

Open



STANDARD OPERATING PROCEDURE FOR:

G2 Speech Assessment

SOP Details:

Number: SOP-COCO-0750	Version: 2
Author(s): Samantha Burr	
Authorised by: Melanie Lewcock	
Date Published: XX/06/2017	
Date to be reviewed: XX/06/2018	

Review History:

Review Date	Reviewed By	Section(s) Amended	Authorised By
27/03/2018	Samantha Burr	4.2.2.1	

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1	Purpose/Scope	Error! Bookmark not defined.
2	Definitions & Abbreviations	Error! Bookmark not defined.
2.1	Pre-Requisite Knowledge, Training, Equipment & Systems	Error! Bookmark not defined.
3	Roles & Responsibilities (Actors)	Error! Bookmark not defined.
4	Procedure	Error! Bookmark not defined.
4.1	Prior to the visit	Error! Bookmark not defined.

4.2	During the visit.....	Error! Bookmark not defined.
4.2.1	Administer the parent questionnaire....	Error! Bookmark not defined.
4.2.2	Administer the TPT speech assessment (Go to 4.2.3 if child is age 3;0+)	Error! Bookmark not defined.
4.2.3	Administer the DEAP speech assessment	Error! Bookmark not defined.
4.3	At the end of the visit	Error! Bookmark not defined.
5	Saving and Storage of Data Recordings.....	Error! Bookmark not defined.
5.1	After the visit	Error! Bookmark not defined.
5.2	Download of recordings	Error! Bookmark not defined.
6	Quality Control Measures	Error! Bookmark not defined.
7	Information Security	Error! Bookmark not defined.
8	Related Documents and Appendix	Error! Bookmark not defined.
9	Fieldworker competency criteria	Error! Bookmark not defined.

NOTE: Passwords should be stored in the password bank held by Data Linkage and Information Security Manager, not in SOPs

1 Purpose/Scope

Recruiting the next generation of ALSPAC (the offspring of the children in ALSPAC) will provide a three generational cohort with data, available for answering research questions relating to life course and genetic epidemiology and providing insights into biological and causal epidemiological mechanisms.

The speech assessment will be carried out with the child (G2) at the 24 month, 36 month and 48 month clinic appointments. The child's responses will be audio recorded for precise phonetic transcription by a qualified Speech and Language Therapist at a later date. The purpose for doing this measure is to provide detailed speech sound data, available for providing insights into causal epidemiological mechanisms.

2 Definitions & Abbreviations

SOP: Standard Operating Procedure

FW: Fieldworker

SFW: Senior Fieldworker

SP: Study participant

COCO90s: Children of the children of the 90s

PIS: Patient information sheet

PI: Principle investigator

DEAP: Diagnostic Evaluation of Articulation and Phonology (speech assessment)

TPT: Toddler Phonology Test (speech assessment)

2.1 Pre-Requisite Knowledge, Training, Equipment & Systems

- Access to COCO90s REDCap Database
- Access to R:Drive
- Knowledge of how to administer the speech assessment
- Knowledge of how to use the audio recording equipment

The following equipment should be prepared:

- Microphone (including USB lead, desktop stand and pop filter)
- Laptop/PC (running software programme Audacity)
- Parent questionnaire (labelled with participant ID)
- Speech assessment recording sheet (labelled with participant ID)
- Consent 6 (labelled with participant ID)
- DEAP assessment stimuli picture book and manual for reference
- TPT assessment stimuli picture book and manual for reference
- A selection of simple toys to provide a brief break during assessment where needed (bubbles, click-clack track, blocks etc).

3 Roles & Responsibilities (Actors)

Who	What & Why
COCO90s project manager	To ensure fieldwork team have received full training. To ensure that all data is collected and processed according to the protocol and ALSPAC information security guidelines.

COCO90s senior fieldworker	To ensure equipment is available and maintained and to liaise with the study PI if there are any problems with data collection
COCO90s fieldworkers	To obtain informed consent from study participants. To administer the parent questionnaire at the beginning of the session. To set up the audio recording equipment and to administer the speech assessment.

4 Procedure

4.1. Prior to the visit

- Ensure that the child meets the inclusion criteria for the study and does not meet the pre-defined exclusion criteria.
- Ensure you have the correct assessment and assessment form appropriate to the child's age:

Assessment	Age
TPT	2;0 – 2;11
DEAP	3;0 – 6;11

- Ensure the picture stimulus flip-book is positioned up-right on the table. If you are using the DEAP, turn the pages until the 'Articulation' title page is facing the child's position. Position the book away from the microphone to one side, to avoid interference.
- **TPT**: Ensure you are familiar with the '*Notes for Administration*' and '*Tips for Testing Two-year-olds*' printed inside the assessment booklet.
- **DEAP**: Ensure you are familiar with the assessment instructions printed inside the blue Articulation and Oro-motor assessment booklet. These are under the bold text subheadings of Picture Naming, Speech Sound Stimulability, Diadochokinetic, Isolated Movement and Sequenced Movements.
- Ensure the microphone is positioned on the table in front of where the child will be positioned, between 60cm and 1 metre away. The logo and voice grill must be facing the child.
- Ensure the pop filter is clamped securely to the microphone stand and that the disc is positioned in front of the microphone, like a shield.
- Ensure the microphone is connected to the PC/laptop by the USB cable. There is no 'on' switch on the microphone. When it is 'on' and has a power feed the red LED will show.
- If using a laptop, please ensure the power supply is connected to ensure sufficient battery for recording.

Complete a test recording with the microphone (see below) and record for 10-20 seconds. Log to record the microphone is working.

- Open Audacity.
- Press the 'record' button (red circle) when you are ready to record. Ensure you leave a 1-2 second pause before speaking to give the programme time to run.
- Say the date, Child ID and FW name
- Use the pause button if a break is required during the assessment
- Press the 'stop' button (yellow square) to stop recording.

- Press the 'play' button (green triangle) to playback the test.
- Ensure the computer is recognising the microphone and that the recorded file can be replayed, if the file cannot be replayed, repeat the test recording. If this does not work close and reopen Audacity and repeat the steps above. If problems continue, consult the SFW.
- A pack should be set up containing the items listed in section 2.1, using a microphone that has been successfully tested.

4.2 During the visit

- Give the SP the opportunity to read the patient information booklet and to ask any further questions.
- Ensure the SP correctly completes **Consent 6** – the adult will need to consent to their completing of the questionnaire, and to the child completing the speech assessment.

4.2.1. Administer the parent questionnaire.

- Go through the questions on the questionnaire with the G1 and record on Redcap

4.2.2. Administer the TPT speech assessment (Go to 4.2.3 if child is age 3:0+)

Note: there is no specific oro-motor element to the assessment for 2-year olds, only a picture-naming task.

- Cue child in to assessment activity using 'Tips for Testing Two-Year-Olds' information top-right inside assessment booklet as guidance.
- Please note the instructions for prompting in the top left of the booklet.
- As you turn the picture, you also need to turn the book. To show the appropriate picture.
- Try to turn each page carefully, trying to ensure that the child answers once the page has been turned, rather than during, to minimise noise interference with the speech recording. If the child is responding too quickly, gently remind them to wait for you to turn the page, before responding.
- Some pictures trigger more than 1 target word – the item numbers of the target words are indicated in white in the bottom right of each picture.
- Many children will respond better to an approach of 'reading' the book together as a shared activity, rather than repeatedly asking 'what's this?'.
- There is no prescribed way of cueing the child in to naming the items, ***however*** it is important that if you say the word for the child to repeat, you clearly mark the item with 'i' (imitated).
- Avoid speaking over the child to ensure clarity of recording. If this happens prompt them to repeat.

4.2.3. Administer the DEAP speech assessment

Articulation Assessment – Picture Naming

- On the DEAP form add child ID, gender and date of birth DD/MM/YY and date of clinic visit.
- Open the blue Articulation and Oro-motor assessment booklet and refer to the left-hand side of the booklet (blue page).
- Cue the child in to the task by saying, "now [child's name], we are going to look at some pictures!".
- Refer to and follow the instructions under the subheading 'Picture naming' at the top left of the page.
- Turn each page carefully, trying to ensure that the child answers once the page has been turned, rather than during, to minimise noise interference with the speech recording. If

the child is responding too quickly, gently remind them to wait for you to turn the page, before responding.

- Many children will respond better to an approach of 'reading' the book together as a shared activity, rather than repeatedly asking 'what's this?'
- Avoid speaking over the child to ensure clarity of recording. If this happens prompt them to repeat.
- If you need to use cues to prompt the child to name the picture, use the following strategies (e.g. for 'pig'):
 - *Function*: it lives on a farm
 - *Sentence completion*: Peppa ____
 - *First sound/syllable*: It starts with "puh..."
 - *Modelling*: Say 'pig'
- There is no prescribed way of cueing the child in to naming the items, **however** it is important that if you say the word for the child to repeat, you clearly mark the item with 'i' (imitated).
- For the DEAP assessment (children aged 3+ years), at the end of the 30 pictures of the picture-naming task, administer the 3 'complex' pictures immediately following on in the flip book (frog in web / sheep with pram / monkey on ball). For each picture ask the child "*can you tell me all about this picture?*". There is no need to make any written notes as the response will be audio recorded for later transcription.
- Once you have administered all pictures, remove the book from the table to reduce distractions.

Articulation Assessment – Speech Sound Stimulability

- On the DEAP paper form add the child ID, gender and date of birth DD/MM/YY
- Refer to the right half of the blue table on the left-hand page and follow the directions under the bold text heading 'Speech Sound Stimulability' but elicit ALL the items in the list.
- In each instance, it is the first sound in the word that is being assessed. Tick the box if you feel the response was accurate for each repetition or note the sound they used if it was different.
- After the word, elicit the single sound on its own (right-hand side of the column). For example, "pie – pie – pie – p".
- Ensure that you model a clear single sound, rather than adding an extra vowel sound at the end (say 'p' rather than 'puh'). Tick the box if you feel the response was accurate for each repetition or note the sound they used if it was different.

Articulation Assessment – Diadochokinetic

- Move on to the right-hand side of the assessment booklet (red page).
- Cue the child in to the task as per the instructions under the bold text heading Diadochokinetic.
- Note the age of the child and the number of repetitions required for the task.
- Try not to clap too loudly yourself, to avoid interference with the audio recording. Model 'gentle' clapping for the child to copy. Alternatively use each finger to distinguish number of repetitions.
- Note the score for correct sequence, intelligibility and fluency in the right-hand column.

Articulation Assessment – Isolated Movement

- Cue the child in to the task as per the instructions under the bold text heading Isolated Movement.
- Score the 2nd attempt, unless the child performs the full action the first time, in which case score the first attempt.
- Score the child for each exercise, using the following as a guide (see also bottom of red page on assessment booklet):

Score	Performance
0	An important part of the gesture is missing / other oral gestures are used / a speech sound is made / no oral movement is produced.
1	The overall pattern of gesture is acceptable but defective (e.g. accuracy, force, speed).
2	Accurate performance after some protracted pauses during which unsuccessful movements might be present.
3	Accurate performance after verbal instruction.

Articulation Assessment – Sequenced Movements

- Cue the child in to the task as per the instructions under the bold text heading Sequenced Movements.
- Score the child for each exercise, following the instructions in 4.2.2.4 above.

5. Saving and Storage of Data Recordings

5.1. After the visit

- Update the REDCap record to complete
- Give the paperwork to the line manager for scanning purposes

5.2. Download of recordings by Fieldworker

- Open Audacity
- A box will appear, click File
- Scroll down and Click Export
- A box will appear
- Copy the files to the relevant (child folder at [\[R:\Studies\T23POC Focus on COCO 90s\Raw data\Bxxx Burr\Raw recordings\]](#))
- Rename the file with the participant visit ID (will be the child ID in all cases) and participant type (child)
- Ensure file has appeared on the R:Drive before going back to Audacity software and then clear recording by clicking on Edit “Undo record”

Encrypting files by SFW or Project manager

- Select files to be sent via Fluff from raw data on R:Drive.
- R click and select “7 Zip”
- Select “Add to archive”
- Choose “7z archive” format
- Options – tick “Create SFX archive” and encryption method AES -256.
- Use previously confirmed password created with collaborator
- This will create “Test.exe tmp” file for Fluff file on R: Drive

Fluffing Files

- <https://www.bristol.ac.uk/it-services/applications/fluff/>
- Click on “Facility for the upload of large files”
- Click on “I want to upload a file”
- Click on “File to upload - choose file
- Select file from R:Drive
- Choose: “Retain file until end of: select a date”
- This will generate an email to your university address which you can forward to the required recipient of the files
- NB Please note that you should ensure that the expected recipient will be expecting the file within the timeframe specified.

6. Quality Control Measures

- Ensure accurate entry of the participant ID when recording and saving data
- 3 monthly observation sessions of FW’s to be incorporated.

7. Information Security

All data should be entered directly into Redcap

<https://alspacredcap.epi.bris.ac.uk/redcap/> choose **My Projects**. This system ensures accurate data by having upper and lower limits for data entered and will notify the fieldworker if the measurement is out of range. FW will log in to the REDCap data collection system using unique identifiers (individual usernames and passwords). FW will always log out completely both from Redcap and the computer at the end of the session.

On occasion, there may be an IT problem that impedes use of the computer system and the data collection system. The default position is that if a participant is already at the workstation when the problem occurs, then data collection should be made via the paper data collection form. Copies of these are made available in each room. The fieldworker (once finished with a participant) should then refer to SOP-FWK-0617 for trouble shooting REDCap. If the issue remains unsolved then the issue should be reported by raising a ticket with the IT helpdesk.

REDCap is housed on a secure server and can only be accessed by those with a REDCap user name and password. Access to data collection instruments is restricted to relevant users and different rights of access can be set for each user.

8. Related Documents and Appendix

9. Fieldworker competency criteria

To be able to complete this measure unsupervised fieldworkers must demonstrate the following:

- FW is able to log on to REDCap with their own user name and ID, locate the correct participant and select the relevant session
- FW was able to explain the procedure accurately and appropriately for the participant/parent and obtain informed consent
- FW demonstrated functioning of microphone, assessment materials and audacity software
- FW can complete a test recording
- FW is able to correctly save and store audio recordings
- FW can clear recording on Audacity

Appendix H. Clinical decision making guideline for scoring phonological errors on TPT and DEAP

- If child refuses an item, score the omitted no.V and no.C and mark all errors as 0. The refused items can be removed prior to analysis as they are highlighted as refused on the cue/response tab of the data entry form.
- Unrecognisable responses (e.g. bearing no obvious match to the target) will be scored for V/C errors and 'other' only.
- If "yes" is pronounced "yeah" the WF 's' will be omitted for PCC scoring but not scored as a FCD.
- /θ/ to /d/ scored as stopping only, not backing.
- /w/, /l/, /r/ changed to /d/ = mark as 'other'.
- 'Zebra' pronounced by some as 'zeebra' – ignore as vowel change acceptable (dialect). Similarly, 'strawberry' pronounced as 2 or 3 syllable accepted – not marked as WSD if 2 syllable (strawbri).
- If item missed (by fieldworker) and therefore not presented to child, leave no.V and no.C blank and mark all errors as 0 so that they come up as 'missing' when database exported to STATA. The missed items can be removed prior to analysis as they are highlighted as missing on the cue/response tab of the data entry form.
- Dark /l/ change to vocalised 'l' [o] in WF position (e.g. *girl*). Rationale: dialect/regional. Mark as C error but no specific process.
- WM/WF glottal replacement of /t/ will be scored as consonant error for the purposes of PCC calculation, but will not be considered a clinically relevant (i.e. pathological) error.
 - Glottal replacement of other sounds (e.g. /p/ or /m/ > [ʔ]) will be scored as backing and other error, specified as glottalisation.
- Consonant deletion does not count as stopping.
- Nasalisation of oral consonants will be marked as 'other – nasalisation'.
- /l/ and /j/ to /r/ mark as 'other'.
- /sp/ to /b/ mark as voicing and cluster reduction.
- Lateralised fricatives (e.g. /f/ or /s/ > [ɸ]) will be scored as "other error" and specified with note (e.g. lateralised / affrication) – not backing.
- Gliding is gliding only, nor fronting or backing.
- Errors on sounds NOT scored by DEAP are to be recorded as "other error" (e.g. errors on WM consonants).
- Cluster reductions will not also be scored as instances of consonant deletion, unless both consonants in the cluster are omitted.
- DEAP A12 – disregard all inaccurate responses (i.e. 'that').
 - Inaccurate response cue type = Sp + response = 'missing'. Leave error form blank.
- Simplification of compound words (e.g. *toothbrush* > "brush"): See p16 of TPT manual. Leave out of PCC as per manual. Score only errors, not PCC.
- Class assimilation as 'other' and then specify (e.g., "gloves" as "glug" or [d̥zʌd̥z]).
- Coalescence (e.g., /sw/ > [ʃ]): mark as "other" and then specify.
- Affrication (e.g., /tr/ > [tʃ]): mark as "other" and then specify.
- Immature tokens e.g., [fɪʃi] – ignore WF [i] and score main form as correct.
- A binary scoring system for errors, such that the child either has the error type or not for each target word, rather than more than one occurrence of the same process in a single word.

Appendix I. Strand Three: parent questionnaire

PARENT QUESTIONNAIRE FOR CLINIC SESSION PRIOR TO DEAP SPEECH ASSESSMENT

Form to be filled in by the Speech & Language Therapist administering the assessment.

SECTION 1: ELIGIBILITY FOR INCLUSION	
Do any of the following apply to your child? (PLEASE TICK ALL APPLICABLE)	
A) Genetic Disorder (including Downs Syndrome and other identified syndromes) <input type="checkbox"/>	
B) Other congenital anomaly (e.g Cerebral Palsy, Global Developmental Delay) <input type="checkbox"/>	
C) Diagnosed Learning Disability <input type="checkbox"/>	
D) Permanent Hearing Loss (Sensorineural) <input type="checkbox"/>	
E) Cleft lip and/or palate and/or submucous cleft palate <input type="checkbox"/>	
F) English as second or additional language <input type="checkbox"/>	
SECTION 2: PREVIOUS SPEECH AND LANGUAGE THERAPY	
1) Has your child ever attended regular appointments with a speech therapist to work specifically on their sounds?	YES NO
SECTION 3: TONGUE TIE	
2) Was your child born with a tongue-tie?	YES NO
a. Did your child have the tongue-tie cut?	YES NO
i. How old was your child when the tongue-tie was cut?	Age __ (weeks)
SECTION 4: DUMMY / DIGIT SUCKING	
1) Has your child ever used a dummy?	YES NO
2) Has your child ever sucked their finger/thumb?	YES NO
3) At 6 months	
a. Dummy - mostly / sometimes / never	
b. Finger/Thumb - mostly / sometimes / never	
4) At 12 months	
a. Dummy - mostly / sometimes / never	
b. Finger/Thumb - mostly / sometimes / never	
5) At 24 months	

- a. Dummy - mostly / sometimes / never
- b. Finger/Thumb - mostly / sometimes / never

6) At **36 months**

- a. Dummy - mostly / sometimes / never
- b. Finger/Thumb - mostly / sometimes / never

7) At **48 months**

- a. Dummy - mostly / sometimes / never
- b. Finger/Thumb - mostly / sometimes / never

SECTION 5: FEEDING HISTORY

- 1) In the first **4 weeks**, did you
- a. Exclusively breastfeed
 - b. Exclusively bottle-feed
 - c. Mixed feeding (breast and bottle)

- 2) At age **2-3 months**, did you
- a. Exclusively breastfeed
 - b. Exclusively bottle-feed
 - c. Mixed feeding (breast and bottle)

- 3) At age **4-6 months**, did you
- a. Exclusively breastfeed
 - b. Exclusively bottle-feed
 - c. Mixed feeding (breast and bottle)

- 4) At age **7-9 months**, did you
- a. Exclusively breastfeed
 - b. Exclusively bottle-feed
 - c. Mixed feeding (breast and bottle)

5) Other feeding method (please give details including ages and duration).

6) At what age did you introduce solids (e.g. soft, pureed foods)? (___ months)

Appendix J. Health Research Authority (HRA) approval 2017

This appendix has been removed as it contains personal information.

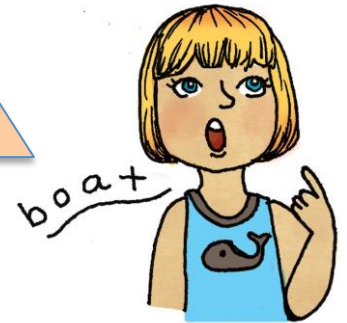
Appendix K. Strand Three: participant information sheet (child)

Patient Information Sheet (4-5 years)

Samantha Burr



Baby Feeding and Sounds



We are looking at how babies are fed and how children use sounds in their talking.

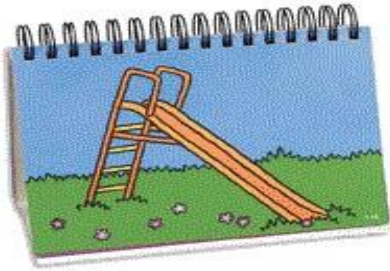
You can come to clinic with

_____ (Mummy/Daddy/Carer's name)

**and meet a Speech Therapist,
who helps children with their
talking.**



The therapist will ask _____ (as above) some questions.



The therapist will show you some pictures for you to name.

Samantha.burr@nhs.net

01.10.2017 Patient Information Sheet (4-5 years)

PIS_Child_ Version No.01/Date:

Samantha Burr

The therapist will use a special microphone to record your talking. This helps her to hear the sounds better.



At the end of the session, you will get a little toy and a certificate for taking part.

If you would like to take part, you can write your name here:

My name is

Appendix L. Strand Three: participant information sheet (accessible information version for adults)

This appendix has been removed as it contains personal information

Appendix M - Strand Three: participant information sheet (adults)

This appendix has been removed as it contains personal information

Appendix N. Strand Three: consent to participate form

IRAS ID: 230190

Study Number: ICA-CDRF-2016-02-053

Participant Identification Number for this trial:



CONSENT TO PARTICIPATE FORM

Title of Project: The effect of different feeding methods and non-nutritive sucking behaviours on child speech development

Name of Researcher: Samantha Burr

Please **initial** box

1. I confirm that I have read the information sheet dated..... (version.....) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.
2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.
3. I understand that relevant sections of my and my child's medical notes and data collected during the study, may be looked at by individuals from the University of the West of England, from regulatory authorities or from Solent NHS Trust, where it is relevant to my and my child's taking part in this research. I give permission for these individuals to have access to my and my child's records.
4. I understand that the information collected about me will be used to support other research in the future, and may be shared anonymously with other researchers.
5. I agree to my General Practitioner being informed of my participation in the study.
6. I agree to the assessment of my child's speech being audio recorded. I understand that video recording will **not** be made. I understand the recording will be anonymous and may be listened to by individuals from the University of the West of England.

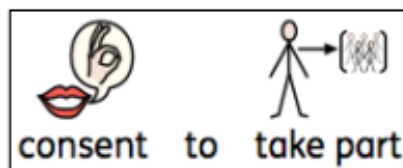
- 7. I agree to participating via video platform if it is not possible to attend a face-to-face clinic appointment.
- 8. I agree to take part in the above study.
- 9. I am happy to be contacted by the research team after the appointment for the purpose of Patient and Public Involvement and Engagement (PPIE) (optional).

Name of Participant	Date	Signature
Name of Person taking consent	Date	Signature

When completed: 1 for participant; 1 for researcher site file; 1 to be kept in clinical notes.
 Version No.03 / Date: 21.07.2020

Appendix O. Strand Three: consent to participate form (accessible information version)

CONSENT TO PARTICIPATE FORM



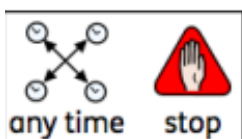
Title of Project: The effect of different feeding methods and non-nutritive sucking behaviours on child speech development

Name of Researcher: Samantha Burr

Please initial box



1. I confirm that I have read the information sheet dated.....
(version.....) for this study. I have had the opportunity to think about the information, ask questions and I am happy with the answers I have been given.



2. I understand that my participation is voluntary and that I am free
to withdraw at any time without giving any reason, without my medical care or legal rights being affected.



3. I understand that relevant sections of my and my child's medical
notes and data collected during the study, may be looked at by individuals from the University of the West of England, from regulatory authorities or from Solent NHS Trust, where it is relevant to my and my child's taking part in this research. I give permission for these individuals to have access to my and my child's records.

4. I understand that the information collected about me will be used to support other research in the future, and may be shared anonymously with other researchers.



5. I agree to my General Practitioner being informed of my participation in the study.

6. I agree to the assessment of my child's speech being audio recorded. I understand that the recording will be anonymous and may be listened to by individuals from the University of the West of England.

7. I agree to take part in the above study.

Name of Participant

Date

Signature

Name of Person taking consent

Date

Signature

Appendix P. Strand Three: clinic SLT information sheet

This appendix has been removed as it contains personal information

Appendix Q. PPIE activity poster advert

**BE A PART
OF SOMETHING
BIGGER**

Do you have a child aged 5 years and under?

You could help us learn how to better support children with Speech Sound Disorders.

(We are particularly interested in parents of children with speech sound difficulties; however this is not a requirement for attending)

Solent NHS Trust, the University of Bristol and the University of the West of England are planning a research project, which will help us identify which children are most likely to need help with speech sound difficulties. We hope that this will help us offer the right support to the right children at the right time. We need parents to help us design our research. Your input can make sure we ask the right questions.

If you would like to help, we are holding a focus group:

Friday 29th April from 10:30am – 12:00pm

**The Multi-Use Room, Spring Meadow Children's Centre, Smannell Road,
Andover, SP11 6JP**

Tea and cakes will be provided!

Babes in arms are welcome to come to the meeting.

If you are interested and would like to be involved, please contact me,
Samantha Burr at samantha.burr@solent.nhs.uk,
or you can leave a message for me on 0300 300 2019

Join the discussion online at www.afasic.org.uk/parentforum



Appendix R. PPIE parent forum aims and questions

PPIE Event – 29th April 2016
10:30am – 12pm @ Spring Meadow Children’s Centre

Materials: Pens, paper, notebook, feedback sheets, cake, biscuits etc.

Housekeeping

- Fire exits / assembly point
- Refreshments – help yourselves at any time
- Loos / baby change / breastfeeding
- Group confidentiality / mutual respect of views
- Plan for today: intros, discussion, feedback
- Finish time

Introduction to Me

- Qualified children’s SLT with Solent NHS, based in Andover/Winchester, specialising in SSD, interest in research – recently had baby, HV sowed the seed for the project....

What is the purpose of the parent group?

- o To get parent’s ideas and input for the study to make sure that the research we do is relevant and meaningful to parents.
- o Parents will be directly involved in the study so we want to ensure that:
 - the information we give to parents is accessible and useful to them
 - we collect the data in a way that works for them
 - the way we feedback to them is useful
 - the way we share our findings and results have a real impact for parents
- o Having your say throughout the life of the project.....

Introduction to Study

What is it about?

- o Looking at the relationship between feeding, sucking and speech development.
- o What we mean by ‘speech’.
- o Joint project between Solent NHS Trust, UoB and UWE – the team is made up of SLT research specialists, lecturers in child and family health, qualified SLTs, psychologists.

What are we hoping to achieve?

- o To describe the different effects that breast and bottle-feeding have on speech development.
- o To use this information to help midwives, HVs, childcare workers, SLTs and parents to pick up children who are at risk of SSD (e.g. risk checklist).
- o To help parents make informed decisions about caring for their children.
- o To help SLTs choose the best assessment methods for the child.
- o To work with bottle/dummy companies to develop teats that reduce impact on speech.

Questions/Discussion Points
1) The study looks at feeding, sucking and speech development.
<i>a. What does this mean to you? Is it important? Why?</i>
<i>b. What do you want this study to tell you?</i>

Discuss study aims (useful? wording? more?):

To use this information to help midwives, HVs, childcare workers, SLTs and parents to pick up children who are at risk of SSD (e.g. risk checklist).

To help parents make informed decisions about caring for their children.

To help SLTs choose the best assessment methods

To work with bottle/dummy companies to develop teats that reduce impact on speech.

2) We will be recruiting children to our study using information from current SLT caseloads. We will contact families who have a child under the age of 5 years who has been diagnosed with speech difficulties to invite them to take part in the study.

a. How would you want us to contact you?

b. What information would you want to be given?

c. How would you want to contact us to find out more?

3) Your child already has appointments booked with the speech therapist as part of their NHS care plan. Would you be willing to attend an extra appointment to have the assessment for the research project?

a. Would you be willing to attend an extra appointment to have the assessment for the research project?

4) The assessment for the research project will take about 60-90 minutes to complete. Because we have to record your child talking, we need to ensure there is no background noise, and so it has to be done in clinic. We would reimburse parents travel costs.

a. How far would you be willing to travel to come to the appointment?

The assessment will involve bringing your child to an appointment with a researcher (SLT). They will ask you questions about your child's development since birth (case history). They will ask about how your child was fed from birth (bottle, breast, mixed) and whether they like to have a dummy or suck their hand/finger. They will also carry out a speech assessment with your child to collect information on the sounds they use and any difficulties they might have.

a. What are your concerns/thoughts on this?

From the assessment results, it may be the case that information/difficulties come to light that may not have previously been addressed/recognised. This scenario would be clearly outlined in the information we give to parents when we invite them to participate in the study.

a. As a parent of a child who has been assessed for the project, how would you want us to share this information with you?

As your child will already be under the care of an NHS Speech Therapist, we are able to share the results of the research assessment with them as this may help them to provide the right care/support for your child.

During the speech assessment, we will need to record your child's talking to make sure we can accurately collect the data. This will be audio (sound) recording, not video. We will provide you with full information on how we will keep this recording (data) safe.

1. What specific information do you want to know about our data storage and use?

- a. How will the data be stored?**
- b. Who will have access to it?**
- c. What will it be used for?**
- d. When will the data recording be destroyed?**

Looking to the future – would you be interested in being a Parent Forum member for the study long-term? Parent Representative?

Appendix S. PPIE parent forum feedback sheet

NHS Speech Sound Research – Parent Forum Feedback Sheet

We'd like to collect some information about the parents who attend the Parent Forum so that we can understand more about your views, experience and perspectives.

Please circle your age range:			
Under 25	26-35	36-45	45+
Do you have children under 5 years old?	YES / NO	If yes, how many?	
Has your child ever seen a Speech & Language Therapist?			
Has your child been diagnosed with Speech Sound Difficulties?			
Have you ever taken part in health research? (e.g.)			

Please tell us what you thought about today's session:

(e.g. Do you feel you have contributed to the study? Were you able to have your say? What did you like about today? What could we have done differently to improve the session for you?)

<i>Comments:</i>

If you are interested in staying in touch with the study as part of the Parent Forum, please fill in your contact details so that we can keep you up to date.

Your details will be stored securely and will only be accessible to the Lead Researcher, Sam Burr.
Your details will NOT be passed to any other 3rd party.

Name: _____ **Email address:**

Appendix T. PPIE poll online parent forum questions

1. **Imagine you are out and about during the day.**
 - a. Where would you expect to see a poster/advert for this study?
2. **Imagine you've seen the poster and you want to know more about the study.**
 - a. How do you want to contact us?
3. **Imagine your child meets the criteria for our study.**
 - a. How would you want us to invite you to participate?
4. **Imagine your child already has appointments booked with the speech therapist as part of their NHS care plan (e.g. therapy, review).**
 - a. Would you be willing to attend an extra appointment to have the assessment for the research project?
5. **The assessment session will last 60-90 minutes. Because we have to record your child talking, we need to ensure there is no background noise, and so the room environment has to be controlled. This means we need to see your child in clinic, rather than at your home. We will reimburse parents travel costs.**
 - a. How far would you be willing to travel to come to the appointment?
6. **Imagine you have agreed to participate in the study. The assessment will involve bringing your child to an appointment with a researcher. The researcher will also be a qualified Speech and Language Therapist. They will ask you questions about your child's development since birth (case history). They will ask about how your child was fed from birth (bottle, breast, mixed) and whether they like to have a dummy or suck their hand/finger. They will also carry out a 20-30 minute speech assessment with your child to collect information on the sounds they use and any difficulties they might have. We will provide feedback on the results of the research assessment.**
 - a. How would you like to receive the feedback?
7. **During the speech assessment, we will need to record your child's talking to make sure we can accurately collect the data. This will be audio (sound) recording, not video. We will provide you with full information on how we will keep this recording (data) safe.**
 - a. What specific information do you want to know about our data storage and use?
8. **If your child were a participant in the study, would you want to be informed of the progress and findings of the study via regular updates?**
9. **Now we'd like you to think about getting more involved with the research. When families are invited to take part in the study, we will be giving them information about the project and the assessment session.**

- a. Would you be willing to help design the information sheets to make sure they are relevant and useful for parents?

10. During the 4 year project, we will be keeping in touch with Parent Representatives throughout the project as part of a Steering Group aimed at providing input and feedback to guide the research as it progresses.

- a. If you would like to be one of our Parent Representatives, how would you like us to keep in touch with you?

11. 2-3 Parent Representatives will be invited to attend Steering Group Meetings twice a year with the researchers as well as members of the University and NHS support teams. These will be opportunities to discuss the progress and updates on the study and contribute to the next phase. Travel to these meetings will be reimbursed.

- a. Would you be willing to attend some of these meetings?

Appendix U. PPIE poster RCLT conference (September 2017)

The Benefits of Using Social Media for PPI Activities

Samantha Burr, Paediatric Speech & Language Therapist, Children's Therapy Service
samantha.burr@solent.nhs.uk

BACKGROUND

A Patient & Public Involvement (PPI) activity was carried out as part of the preparation of a research application for a National Institute of Health Research (NIHR) Clinical Doctoral Research Fellowship (CDRF).

AIM: To maximise public involvement (parents of children under 5 years) in research design in a short timeframe.



METHOD	Face-2-Face	Online Web Forum	Social Media
	90 minute meeting at a local children's centre, advertised with posters, Twitter and Facebook.	Discussion board hosted by parent-led children's charity Afasic, advertised on Twitter and Facebook.	Facebook Poll App used to create an online survey, advertised on Twitter and Facebook.
RESULTS	6 attendees from within 5 mile radius of centre. 5 registered for continued involvement.	129 forum views. NO posts or replies.	1190 visitors in <1 month. 149 poll responses. 61 parents registered for continued involvement. Participants from across UK.
CONCLUSIONS	<ul style="list-style-type: none"> ✓ Personal interaction and discussion. X Travel (geography) X Time (setup, participant) X £££ (booking, travel, expenses) X Limits group size X Publicity issues X Cancellations / no-shows 	<ul style="list-style-type: none"> ✓ Established platform ✓ Recognised organisation ✓ IT support ✓ Live visit/reply counters ✓ Instant live discussion X Sign-up / email verification required X Visit VS engagement X IT skills / familiarisation required to navigate forum threads 	<ul style="list-style-type: none"> ✓ Easy access (smartphone/ computer) ✓ Instant responses ✓ Reduced time burden for parent ✓ Easy intuitive setup (poll app) ✓ Engage huge national/ international audience ✓ Instant publicity via hash tags (#parents) / handles (@JoeBloggs) ✓ Instant data download (excel) X Impact of 'facelessness' on response quality?

NEXT STEPS:

Explore participant experience of using social media to participate in health research.

REFERENCES: www.afasic.org.uk www.facebook.com www.twitter.com

Appendix V. Strand One Part A: unadjusted and adjusted logistics regression models (rerun with bottle feeding as the reference group).

Table 1. Logistic regression of parental speech concern at age 18 months and feeding method at ages 4 weeks and 15 months (bottle feeding as reference group)

Feeding Method		Model 0: Unadjusted			Model 1: Adjusted for biological sex and home ownership			Model 2a: Adjusted for biological sex, home ownership and weak sucking at age 4 weeks		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.89 [0.71,1.12]	-0.99	.322	0.90 [0.71,1.13]	-0.92	.356	0.91 [0.72,1.14]	-0.82	.411
	Breast fed	0.82 [0.69,0.98]	-2.23	.026	0.83 [0.69,0.99]	-2.09	.037	0.86 [0.72,1.02]	-1.71	.087
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	1.04 [0.85,1.28]	0.40	.692	1.07 [0.86,1.32]	0.60	.548	1.09 [0.88,1.36]	0.83	.407
	Breast fed	0.52 [0.30,0.89]	-2.37	.018	0.53 [0.31,0.92]	-2.26	.024	0.52 [0.30,0.92]	-2.25	.024

Table 1. (Continued)

Feeding Method		Model 2b: Adjusted for biological sex, home ownership and maternal age			Model 3: Adjusted for biological sex, home ownership, weak sucking at age 4 weeks and maternal age		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.91 [0.73,1.15]	-0.75	.453	0.93 [0.73,1.17]	-0.65	.518
	Breast fed	0.85 [0.71,1.02]	-1.77	.077	0.88 [0.73,1.05]	-1.40	.161
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-
	Mixed fed	1.08 [0.87,1.34]	0.74	.462	1.12 [0.90,1.39]	1.01	.311
	Breast fed	0.55 [0.32,0.95]	-2.13	.034	0.55 [0.31 0.96]	-2.09	.037

Note: *N* for Age 4 Weeks: model 0 *n*=8134; model 1 *n*=7969; model 2a *n*=7969; model 2b *n*=7969; model 3 *n*=7969.

Appendix W. Strand One Part B: univariable analysis of feeding and speech error frequency at age 25 months

Table 2. Univariable negative binomial regression results for feeding and speech sound error frequency at age 25 months (bottle feeding as reference group)

Exposure Variable: Feeding Method		Consonant Type Error Frequency at Age 25 Months								
		Velar			Consonant Cluster			Liquid		
		IRR [95% CI]	<i>z</i>	<i>p</i>	IRR [95% CI]	<i>z</i>	<i>p</i>	IRR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	1.02 [0.80,1.29]	0.14	.890	0.95 [0.81,1.11]	-0.69	.488	0.98 [0.81,1.17]	-0.27	.788
	Breast fed	1.03 [0.86,1.24]	0.35	.728	1.14 [1.00,1.29]	2.09	.037	1.17 [1.01,1.34]	2.12	.034
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	1.11 [0.89,1.40]	0.93	.353	1.18 [1.01,1.38]	2.15	.031	1.22 [1.02,1.46]	2.16	.031
	Breast fed	1.38 [0.89,2.15]	1.42	.155	1.20 [0.88,1.62]	1.16	.244	1.08 [0.75,1.56]	0.44	.663

Table 1. (Continued)

Exposure Variable: Feeding Method		Consonant Type Error Frequency at Age 25 Months					
		Fricative			Postvocalic		
		IRR [95% CI]	<i>z</i>	<i>p</i>	IRR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-
	Mixed fed	1.03 [0.83,1.28]	0.29	.769	0.94 [0.69,1.28]	-0.38	.703
	Breast fed	1.06 [0.89,1.25]	0.64	.521	1.01 [0.79,1.28]	0.05	.962
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-
	Mixed fed	1.10 [0.90,1.35]	0.93	.352	1.03 [0.76,1.40]	0.21	.833
	Breast fed	1.11 [0.74,1.67]	0.51	.613	0.93 [0.50,1.74]	-0.22	.823

Note: *N* for all age 4 week models =694 except fricative (*n*=693).
N for all age 15 month models *n*=437.

Appendix X. Strand One Part B: unadjusted and adjusted logistics regression models for feeding and speech error frequencies at age 25 months (rerun with bottle feeding as the reference group).

Table 1. Negative binomial unadjusted and adjusted regression models for feeding method and consonant cluster speech sound error frequency at age 25 months (bottle feeding as reference group)

Feeding Method		Model 0: Unadjusted			Model 1: Adjusted for biological sex and home ownership			Model 2a: Adjusted for biological sex, home ownership and weak sucking at age 4 weeks			Model 2b: Adjusted for biological sex, home ownership and maternal age		
		IRR [95% CI]	<i>z</i>	<i>p</i>	IRR [95% CI]	<i>z</i>	<i>p</i>	IRR [95% CI]	<i>z</i>	<i>p</i>	IRR [95% CI]	<i>z</i>	<i>p</i>
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	1.18 [1.01,1.38]	2.15	.031	1.17 [1.01,1.37]	2.09	.036	1.19 [1.02,1.38]	2.24	.025	1.24 [1.06,1.44]	2.71	.007
	Breast fed	1.20 [0.88,1.62]	1.16	.244	1.22 [0.90,1.64]	1.29	.197	1.25 [0.92,1.69]	1.45	.147	1.33 [0.98,1.80]	1.85	.064

Table 1. (Continued)

Feeding Method		Model 3a: Adjusted for biological sex, home ownership and word combination at age 25 months			Model 3b: Adjusted for biological sex, home ownership and RDLS comprehension standardised score at age 25 months			Model 4: Adjusted for biological sex, home ownership, weak sucking at age 4 weeks, maternal age, word combination at age 25 months and RDLS comprehension standardised score at age 25 months		
		IRR [95% CI]	<i>z</i>	<i>p</i>	IRR [95% CI]	<i>z</i>	<i>p</i>	IRR [95% CI]	<i>z</i>	<i>p</i>
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	1.15 [1.02,1.38]	1.87	.062	1.10 [0.95,1.28]	1.32	.187	1.15 [0.99,1.33]	1.88	.060
	Breast fed	1.20 [0.90,1.60]	1.23	.219	1.08 [0.81,1.45]	0.55	.585	1.21 [0.91,1.61]	1.32	.188

Note: *N* for Age 4 Weeks: model 0 *n*=694; model 1 *n*=686; model 2a *n*=686; model 2b *n*=686; model 3a *n*=681; model 3b *n*=667; model 4 *n*=662.

Note: *N* for Age 15 Months: model 0 *n*=437; model 1 *n*=432; model 2a *n*=427; model 2b *n*=432; model 3a *n*=430; model 3b *n*=417; model 4 *n*=410.

Table 2. Negative binomial unadjusted and adjusted regression models for feeding method and liquid speech sound error frequency at age 25 months (bottle feeding as reference group)

Feeding Method		Model 0: Unadjusted			Model 1: Adjusted for biological sex and home ownership			Model 2a: Adjusted for biological sex, home ownership and weak sucking at age 4 weeks			Model 2b: Adjusted for biological sex, home ownership and maternal age		
		IRR [95% CI]	<i>z</i>	<i>p</i>	IRR [95% CI]	<i>z</i>	<i>p</i>	IRR [95% CI]	<i>z</i>	<i>p</i>	IRR [95% CI]	<i>z</i>	<i>p</i>
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	1.22 [1.02,1.46]	2.16	.031	1.22 [1.02 1.45]	2.17	.030	1.24 [1.04,1.48]	2.35	.019	1.28 [1.02,1.46]	2.71	.007
	Breast fed	1.08 [0.75,1.56]	0.44	.663	1.12 [0.98,1.60]	0.60	.546	1.17 [0.82,1.68]	0.87	.387	1.22 [0.85,1.75]	1.08	.278

Table 2. (Continued)

Feeding Method		Model 3a: Adjusted for biological sex, home ownership and word combination at age 25 months			Model 3b: Adjusted for biological sex, home ownership and RDLS comprehension standardised score at age 25 months			Model 4: Adjusted for biological sex, home ownership, weak sucking at age 4 weeks, maternal age, word combination at age 25 months and RDLS comprehension standardised score at age 25 months		
		IRR [95% CI]	<i>z</i>	<i>p</i>	IRR [95% CI]	<i>z</i>	<i>p</i>	IRR [95% CI]	<i>z</i>	<i>p</i>
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	1.19 [1.02,1.46]	1.97	.048	1.15 [0.97,1.38]	1.57	.116	1.21 [1.01,1.44]	2.12	.034
	Breast fed	1.10 [0.77,1.56]	0.52	.601	1.01 [0.71,1.43]	0.04	.966	1.15 [0.81,1.64]	0.79	.432

Note: *N* for Age 15 Months: model 0 *n*=437; model 1 *n*=432; model 2a *n*=427; model 2b *n*=432; model 3a *n*=430; model 3b *n*=417; model 4 *n*=410.

Appendix Y. Strand One Part B: univariable logistic regression models for feeding and speech error frequencies at age 61 months (rerun with bottle feeding as the reference group).

Table 3. Univariable logistic regression model for feeding and speech sound error frequency at age 61 months (bottle feeding as reference group)

Exposure Variable: Feeding Method		Consonant Type Error Frequency at Age 61 Months								
		Velar			CC			Liquid		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.64 [0.40,1.01]	-1.91	.056	0.77 [0.50,1.19]	-1.16	.247	0.78 [0.50,1.20]	-1.15	.251
	Breast fed	0.60 [0.42,0.85]	-2.83	.005	0.72 [0.52,1.01]	-1.90	.057	0.93 [0.67,1.29]	-0.44	.660
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.67 [0.45,1.00]	-1.94	.052	0.72 [0.49,1.05]	-1.71	.088	1.03 [0.70,1.51]	0.14	.890
	Breast fed	0.77 [0.34,1.74]	0.62	.538	0.62 [0.29,1.32]	-1.24	.215	1.71 [0.79,3.69]	1.37	.171

Table 1. (Continued)

Exposure Variable: Feeding Method		Consonant Type Error Frequency at Age 61 Months								
		Fricative			Postvocalic			Alveolar		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.68 [0.41,1.11]	-1.55	.122	0.54 [0.35,0.84]	-2.71	.007	0.50 [0.32,0.78]	-3.07	.002
	Breast-fed	0.82 [0.57,1.17]	-1.10	.272	0.67 [0.47,0.95]	-2.24	.025	0.50 [0.35,0.69]	-4.07	<.001
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.89 [0.59,1.34]	-0.57	.567	0.84 [0.57,1.26]	-0.84	.402	0.59 [0.40,0.87]	-2.66	.008
	Breast-fed	0.59 [0.24,1.45]	-1.14	.253	0.60 [0.28,1.29]	-1.32	.188	0.34 [0.15,0.77]	-2.59	.010

Note: *N* for all age 4 week models *n*=709.

N for all age 15 month models *n*=488.

Appendix Z. Strand One Part B: unadjusted and adjusted logistic regression models for feeding and speech error frequencies at age 61 months (rerun with bottle feeding as the reference group).

Table 1. Logistic unadjusted and adjusted regression models for feeding at age 4 weeks and velar speech sound error frequency at age 61 months (bottle feeding as reference group)

Feeding Method		Model 0: Unadjusted			Model 1: Adjusted for biological sex and home ownership			Model 2a: Adjusted for biological sex, home ownership and weak sucking at age 4 weeks		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.64 [0.40,1.01]	-1.91	.056	0.65 [0.40,1.04]	-1.82	.069	0.64 [0.40,1.03]	-1.82	.069
	Breast fed	0.60 [0.42,0.85]	-2.83	.005	0.61 [0.43,0.88]	-2.67	.008	0.61 [0.43,0.88]	-2.66	.008
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.67 [0.45,1.00]	-1.94	.052	0.61 [0.40,0.93]	-2.31	.021	0.59 [0.39,0.91]	-2.41	.016
	Breast fed	0.77 [0.34,1.74]	0.62	.538	0.68 [0.30,1.56]	-0.90	.366	0.71 [0.31,1.61]	-0.82	.409

Table 1. (Continued)

Feeding Method		Model 2b: Adjusted for biological sex, home ownership and maternal age			Model 2c: Adjusted for biological sex, home ownership and OME age 61 months			Model 3a: Adjusted for biological sex, home ownership and language score at age 38 months		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.64 [0.40,1.03]	-1.82	.068	0.62 [0.38,1.01]	-1.94	.052	0.70 [0.43,1.14]	-1.44	.150
	Breast fed	0.61 [0.42,0.88]	-2.65	.008	0.62 [0.43,0.90]	-2.51	.012	0.65 [0.44,0.94]	-2.28	.022
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.63 [0.41,0.96]	-2.12	.034	0.59 [0.39,0.92]	-2.34	.019	0.67 [0.43,1.04]	-1.77	.077
	Breast fed	0.73 [0.32,1.67]	-0.75	.454	0.70 [0.30,1.62]	-0.83	.405	0.71 [0.30,1.69]	-0.77	.441

Table 1. (Continued)

Feeding Method		Model 3b: Adjusted for biological sex, home ownership and RDLS comprehension standardised score at age 61 months			Model 4: Adjusted for biological sex, home ownership, weak sucking at age 4 weeks, maternal age, OME age 61 months, language score at age 38 months and RDLS comprehension standardised score at age 61 months		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.81 [0.49,1.33]	-0.84	.399	0.78 [0.46,1.34]	-0.89	.375
	Breast fed	0.84 [0.56,1.25]	-0.87	.383	0.81 [0.52,1.24]	-0.97	.330
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.82 [0.51,1.29]	-0.86	.388	0.85 [0.51,1.42]	-0.63	.530
	Breast fed	0.89 [0.37,2.18]	-0.25	.804	0.95 [0.36,2.51]	-0.10	.924

Note. *N* for Age 4 weeks: model 0 *n*=709; model 1 *n*=701; model 2a *n*=701; model 2b *n*=701; model 2c *n*=668; model 3a *n*=675; model 3b *n*=601; model 4 *n*=554.

N for Age 15 months: model 0 *n*=488; model 1 *n*=483; model 2a *n*=475; model 2b *n*=483; model 2c *n*=459; model 3a *n*=460; model 3b *n*=416; model 4 *n*=37

Table 2. Logistic unadjusted and adjusted regression models for feeding at age 4 weeks and consonant cluster speech sound error frequency at age 61 months (bottle feeding as reference group)

Feeding Method		Model 0: Unadjusted			Model 1: Adjusted for biological sex and home ownership			Model 2a: Adjusted for biological sex, home ownership and weak sucking at age 4 weeks		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.72 [0.49,1.05]	-1.71	.088	0.79 [0.53,1.18]	-1.16	.247	0.78 [0.52,1.16]	-1.22	.222
	Breast fed	0.62 [0.29,1.32]	-1.24	.215	0.62 [0.28,1.36]	-1.19	.235	0.65 [0.29,1.45]	-1.05	.296

Table 2. (Continued)

Feeding Method		Model 2b: Adjusted for biological sex, home ownership and maternal age			Model 2c: Adjusted for biological sex, home ownership and OME age 61 months			Model 3a: Adjusted for biological sex, home ownership and language score at age 38 months		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.82 [0.55,1.23]	-0.94	.346	0.78 [0.52,1.18]	-1.17	.244	0.87 [0.57,1.32]	-0.67	.506
	Breast fed	0.67 [0.30,1.48]	-1.00	.318	0.55 [0.24,1.24]	-1.45	.148	0.68 [0.30,1.54]	-0.92	.359

Table 2. (Continued)

Feeding Method		Model 3b: Adjusted for biological sex, home ownership and RDLS comprehension standardised score at age 61 months			Model 4: Adjusted for biological sex, home ownership, weak sucking at age 4 weeks, maternal age, OME age 61 months, language score at age 38 months and RDLS comprehension standardised score at age 61 months		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.96 [0.62,1.49]	-0.16	.869	1.10 [0.68,1.80]	0.40	.691
	Breast fed	0.74 [0.32,1.72]	-0.70	.485	0.79 [0.31,2.00]	-0.51	.613

Note: *N* for Age 15 months: model 0 *n*=488; model 1 *n*=483; model 2a *n*=475; model 2b *n*=483; model 2c *n*=459; model 3a *n*=460; model 3b *n*=416; model 4 *n*=375.

Table 3. Logistic unadjusted and adjusted regression models for feeding at age 4 weeks and postvocalic speech sound error frequency at age 61 months (bottle feeding as reference group)

Feeding Method		Model 0: Unadjusted			Model 1: Adjusted for biological sex and home ownership			Model 2a: Adjusted for biological sex, home ownership and weak sucking at age 4 weeks		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.54 [0.35,0.84]	-2.71	.007	0.57 [0.36,0.89]	-2.46	.014	0.56 [0.36,0.89]	-2.48	.013
	Breast fed	0.67 [0.47,0.95]	-2.24	.025	0.71 [0.49,1.01]	-1.93	.054	0.71 [0.50,1.01]	-1.91	.057

Table 3. (Continued)

Feeding Method		Model 2b: Adjusted for biological sex, home ownership and maternal age			Model 2c: Adjusted for biological sex, home ownership and OME age 61 months			Model 3a: Adjusted for biological sex, home ownership and language score at age 38 months		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.59 [0.38,0.94]	-2.24	.025	0.54 [0.34,0.87]	-2.57	.010	0.61 [0.38,0.97]	-2.10	.036
	Breast fed	0.75 [0.53,1.08]	-1.53	.127	0.72 [0.50,1.05]	-1.71	.088	0.78 [0.54,1.13]	-1.32	.186

Table 3. (Continued)

Feeding Method		Model 3b: Adjusted for biological sex, home ownership and RDLS comprehension standardised score at age 61 months			Model 4: Adjusted for biological sex, home ownership, weak sucking at age 4 weeks, maternal age, OME age 61 months, language score at age 38 months and RDLS comprehension standardised score at age 61 months		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.75 [0.46,1.21]	-1.18	.236	0.78 [0.47,1.31]	-0.94	.347
	Breast fed	0.89 [0.60,1.30]	-0.60	.545	1.08 [0.70,1.64]	0.34	.735

Note: *N* for Age 4 weeks: model 0 *n*=709; model 1 *n*=701; model 2a *n*=701; model 2b *n*=701; model 2c *n*=668; model 3a *n*=675; model 3b *n*=601; model 4 *n*=554.

Table 4. Logistic unadjusted and adjusted regression models for feeding and alveolar speech sound error frequency at age 61 months (bottle-feeding as reference group)

Outcome Variable: Feeding Method		Model 0: Unadjusted			Model 1: Adjusted for biological sex and home ownership			Model 2a: Adjusted for biological sex, home ownership and weak sucking at age 4 weeks		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.50 [0.32,0.78]	-3.07	.002	0.55 [0.35,0.87]	-2.56	.010	0.55 [0.35,0.86]	-2.62	.009
	Breast fed	0.50 [0.35,0.69]	-4.07	<.001	0.53 [0.38,0.76]	-3.53	<.001	0.54 [0.38,0.76]	-3.48	<.001
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.59 [0.40,0.87]	-2.66	.008	0.67 [0.45,1.00]	-1.97	.049	0.66 [0.44,0.99]	-2.00	.046
	Breast fed	0.34 [0.15,0.77]	-2.59	.010	0.34 [0.15,0.78]	-2.52	.012	0.35 [0.15,0.82]	-2.41	.016

Table 4. (Continued)

Outcome Variable: Feeding Method		Model 2b: Adjusted for biological sex, home ownership and maternal age			Model 2c: Adjusted for biological sex, home ownership and OME age 61 months			Model 3a: Adjusted for biological sex, home ownership and language score at age 38 months		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.57 [0.36,0.90]	-2.42	.016	0.55 [0.34,0.87]	-2.54	.011	0.63 [0.39,1.00]	-1.96	.050
	Breast fed	0.56 [0.39,0.79]	-3.24	.001	0.55 [0.38,0.78]	-3.30	.001	0.58 [0.41,0.83]	-2.96	.003
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.70 [0.47,1.06]	-1.70	.089	0.68 [0.45,1.03]	-1.84	.066	0.72 [0.47,1.09]	-1.55	.121
	Breast fed	0.37 [0.16,0.86]	-2.30	.021	0.27 [0.11,0.66]	-2.87	.004	0.35 [0.14,0.84]	-2.35	.019

Table 4. (Continued)

Feeding Method		Model 3b: Adjusted for biological sex, home ownership and RDLS comprehension standardised score at age 61 months			Model 4: Adjusted for biological sex, home ownership, weak sucking at age 4 weeks, maternal age, OME age 61 months, language score at age 38 months and RDLS comprehension standardised score at age 61 months		
		OR [95% CI]	<i>z</i>	<i>p</i>	OR [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.69 [0.43,1.13]	-1.48	.139	0.74 [0.44,1.23]	-1.16	.245
	Breast fed	0.66 [0.45,0.97]	-2.14	.033	0.73 [0.48,1.11]	-1.49	.137
Age 15 Months	Bottle fed	1 [-]	-	-	1 [-]	-	-
	Mixed fed	0.87 [0.56,1.36]	-0.59	.554	1.00 [0.61,1.63]	-0.01	.995
	Breast fed	0.39 [0.16,0.98]	-1.99	.046	0.34 [0.13,0.95]	-2.07	.039

Note. *N* for Age 4 weeks: model 0 *n*=709; model 1 *n*=701; model 2a *n*=701; model 2b *n*=701; model 2c *n*=668; model 3a *n*=675; model 3b *n*=601; model 4 *n*=554.

N for Age 15 months: model 0 *n*=488; model 1 *n*=483; model 2a *n*=475; model 2b *n*=483; model 2c *n*=459; model 3a *n*=460; model 3b *n*=416; model 4 *n*=375.

Appendix AA. Strand Two Part A: univariable regression model *n* values for potential confounders associated with SwPCC score

Potential Confounding Variable	SwPCC Score <i>N</i>
Age	122
Biological Sex	122
Home Ownership Status	74
Maternal Age	122
Maternal Education	96
Ear Infection 12m	76
Ear Infection 48m	50
Syllable Combination 12 Months	84
Word Combination 24 Months	109

Appendix AB. Strand Two Part A: univariable logistic regression models for feeding and SwPCC scores (rerun with bottle feeding as the reference group).

Table 4. Univariable regression model results for feeding groups and overall SwPCC score (bottle feeding as reference group)

Exposure Variable: Feeding Method		Outcome Variable: SwPCC Score		
		Coef. [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-
	Mixed fed	3.56 [-7.59,14.71]	0.63	.528
	Breast fed	11.30 [0.82,21.78]	2.14	.035
Age 12 Weeks	Bottle fed	1 [-]	-	-
	Mixed fed	3.89 [-7.25,15.03]	0.69	.491
	Breast fed	3.25 [-6.76,13.26]	0.64	.522
Age 6 Months	Bottle fed	1 [-]	-	-
	Mixed fed	9.73 [-22.33,2.86]	-1.53	.129
	Breast fed	1.09 [-9.57,11.76]	0.20	.840
Age 9 Months	Bottle fed	1 [-]	-	-
	Mixed fed	-9.73 [-22.33,2.86]	-1.53	.129
	Breast fed	1.09 [-9.57,11.56]	0.20	.840

Note: *N* for age 4 weeks and 12 weeks models *n*=121.
N for age 6 month and 9 month models *n*=120.

Appendix AC. Strand Two Part A: univariable regression model *n* values for feeding groups and overall SwPCC score by age group

Exposure Variable: Feeding Group	Outcome Variable: SwPCC Score <i>n</i>		
	Age 24 Months	Age 36 Months	Age 48 Months
Age 4 weeks	41	36	44
Age 12 weeks	41	36	44
Age 6 months	40	37	43
Age 9 months	41	36	43
Age 12 months	25	25	26

Appendix AD. Strand Two Part A: univariable negative binomial regression model *n* values for potential confounders associated with CsPCC score

Potential Confounding Variable	Outcome variable: CsPCC Score <i>n</i>
Age	38
Biological Sex	38
Home Ownership Status	23
Maternal Age	38
Maternal Education	27
Ear Infection 12m	24
Ear Infection 48m	20
Syllable Combination 12 Months	26
Word Combination 24 Months	33

Appendix AE. Strand Two Part A: univariable logistic regression models for feeding and CsPCC scores (rerun with bottle feeding as the reference group).

Table 5. Univariable regression model results for feeding groups and overall CsPCC score

Exposure Variable: Feeding Method		Outcome Variable: Overall CsPCC Score		
		Coef. [95% CI]	<i>z</i>	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-
	Mixed fed	-4.84 [-25.83,16.16]	-0.47	.643
	Breast fed	-20.83 [-41.70,0.05]	-2.03	.051
Age 12 Weeks	Bottle fed	1 [-]	-	-
	Mixed fed	-7.43 [-21.86,6.99]	-1.05	.302
	Breast fed	-14.93 [-34.14,4.29]	-1.58	.124
Age 6 Months	Bottle fed	1 [-]	-	-
	Mixed fed	-9.81 [-25.87,6.24]	-1.24	.223
	Breast fed	-14.20 [-35.53,7.13]	-1.35	.185
Age 9 Months	Bottle fed	1 [-]	-	-
	Mixed fed	-9.81 [-25.87,6.24]	-1.24	.223
	Breast fed	-14.20 [-35.53,7.13]	-1.35	.185

Note: *N* for age 4 weeks and 12 weeks models *n*=37.

N for age 6 month and 9 month models *n*=38.

Appendix AF. Strand Three Part A: univariable regression models for feeding and overall SwPCC score with bottle feeding as reference group

Table 6. Univariable regression model results for feeding groups and overall SwPCC score (bottle feeding as reference group)

Exposure Variable: Feeding Method		Outcome Variable: Overall SwPCC Score		
		Coef. [95% CI]	<i>t</i> [df]	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-
	Mixed fed	9.08 [-6.71,24.87]	1.16 [49]	.254
	Breast fed	9.14 [-0.96,19.24]	1.82 [49]	.075
Age 12 Weeks	Bottle fed	1 [-]	-	-
	Mixed fed	-3.54 [-21.80,14.72]	-0.39 [49]	.698
	Breast fed	7.12 [-2.71,16.97]	1.45 [49]	.153
Age 6 Months	Bottle fed	1 [-]	-	-
	Mixed fed	-1.49 [-18.00,15.01]	-0.18 [49]	.857
	Breast fed	8.08 [-1.92,18.08]	1.62 [49]	.111
Age 9 Months	Bottle fed	1 [-]	-	-
	Mixed fed	-0.53 [-14.71,13.64]	-0.08 [48]	.940
	Breast fed	9.68 [-1.82,21.18]	1.69 [48]	.097

Note: *N* for age 4 week, 12 weeks and 6 month models (*n*=52).
N for age 9 month models (*n*=51).

Appendix AG. Strand Three Part A: univariable logistic regression models for feeding and CsPCC Scores (rerun with bottle feeding as the reference group)

Table 7. Univariable regression model results for feeding groups and overall CsPCC score (bottle-feeding as reference group)

Exposure Variable: Feeding Method		Outcome Variable: Overall CsPCC Score		
		Coef. [95% CI]	<i>t</i> [df]	<i>p</i>
Age 4 Weeks	Bottle fed	1 [-]	-	-
	Mixed fed	12.35 [-3.55,28.25]	1.60 [24]	.122
	Breast fed	19.04 [6.68,31.40]	3.18 [24]	.004
Age 12 Weeks	Bottle fed	1 [-]	-	-
	Mixed fed	2.90 [-17.88,23.68]	0.29 [24]	.776
	Breast fed	17.68 [6.61,28.76]	3.30 [24]	.003
Age 6 Months	Bottle fed	1 [-]	-	-
	Mixed fed	3.49 [-11.24,18.23]	0.49 [24]	.629
	Breast fed	20.71 [9.60,31.81]	3.85 [24]	.001
Age 9 Months	Bottle fed	1 [-]	-	-
	Mixed fed	5.68 [-7.73,19.09]	0.87 [24]	.390
	Breast fed	23.30 [10.77,35.83]	3.84 [24]	.001

Note: *N* for all models (*n*=27).

N for age 9 month models (*n*=51).

Appendix AH. HRA substantial amendment (July 2020)

This appendix has been removed as it contains personal information.