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The Impact of Untreated Ankyloglossia on Feeding, Speech and the Psychosocial Domain: A Systematic Review and Meta-Analysis

Fariah I Gaba ^{1,*}, Chirag C Sheth ²

¹ Dental XL, Private Dental Practise. The Hague. The Netherlands.

² Department of Medicine, Faculty of Health Sciences, Universidad CEU Cardenal Herrera, CEU Universities, Alfara del Patriarca 46115, Valencia, Spain

* **Corresponding author:** Dr. Fariah Gaba (BMedSci. Hons, UK, DDS Valencia). Member of the ICOEV board of Registered Dentists in Spain. Registered Member on the Official List of Practising Dentists in The Netherlands (BIG). Registered Member of Quality Dentists in The Netherlands (KRT).

Email address: fariahgaba@gmail.com Contact Telephone Number: (+31) 649351157

ABSTRACT

Objectives: (1) To investigate and highlight the impact of untreated AG on feeding, speech and the psychosocial domain in children between 0-15 years. (2) determine surgical outcomes of corrective AG procedures and their influence on feeding, speech and the psychosocial domain (3) highlight AG as an important myofunctional and psychosocial disorder, (4) encourage further collaborative research from healthcare professionals.

Methods: The Central Register of Controlled Trials, EMBASE, EBSCO, NCBI and MEDLINE databases were searched, and a meta-analysis was conducted. Positive AG treatment outcomes over the past 33 years were also presented.

Results: Feeding Affectations: 49.76% of individuals received corrective AG surgery and 50.24% did not. Calculated risk ratio (RR) was 0.81 with a 95 % CI of 0.45 - 0.90. The GRADE quality assessment of evidence was “LOW”.

Speech Affectations: 60.20% of individuals never had corrective AG surgery whereas 39.80% did. Calculated risk ratio (RR) was 0.92 with a 95 % CI of 0.59 - 0.98 The GRADE quality assessment of evidence was “LOW”.

The Psychosocial Domain Affectations: Data availability was very limited and there was little acknowledgment of this topic from the included articles. Consequently, the quality of presented evidence was “LOW” as per the GRADE system.

Conclusions: Data depicting a significant link between AG and its level of influence over these issues between 0–15 year-olds is scarce, and our results were inconclusive. AG management requires more effective interdisciplinary communication to aid clinical decision making and further investigation is necessary.

Keywords: Meta-analysis; Ankyloglossia, Feeding, Speech, Psychosocial Domain, Surgical Outcomes

Abbreviations: AG – Ankyloglossia, WHO – World Health Organisation, UNICEF – United Nations Children’s Fund, HATLFF - Hazelbaker Assessment Tool for Lingual Frenulum Function, BTAT – Bristol Tongue-Tie Assessment Tool, PICOS Tool – Population, Intervention, Comparison, Outcomes and Study Design, ABM – Academy of Breastfeeding Medicine, GRADE - Grading of Recommendations, Assessment, Development and Evaluation, CASP - Critical Appraisal Skills Programme, PRISMA - Preferred Reporting System for Systematic Reviews and Meta-Analyses, IAR – Inferior Alveolar Ridge.

INTRODUCTION

Ankyloglossia (AG) possesses its etymological origins from the Greek “agkilos” (curved) and “glossa” (tongue) [49]. It is a congenital anomaly presenting clinically as an abnormally thickened, short or tight sublingual frenulum which significantly inhibits tongue movement. Over the last decade there has been a reported increase in AG prevalence within the infant

population (< 2 years) in Canada [28]. AG prevalence is variable and reported to be between 0.27 % - 10.69 % [2, 16, 18, 23, 40, 51, 52, 53]. To date there is no clearly defined, internationally standardised classification to aid health care professionals in AG detection and diagnosis [18, 25, 32, 46]

AG is a congenital defect that can cause difficulty with breastfeeding with untreated AG in infants being associated with a 25% to 60% prevalence of breastfeeding difficulties such as maternal nipple damage, poor milk supply and breast refusal. [2, 52] The incapability of infants to effectively latch onto the breast negatively affects infant development and could be a primary underlying cause [2, 11, 41, 52]. 3D Ultrasound image analysis has graphically illustrated how appropriate tongue mobility is imperative for breastfeeding efficacy [51] implying that depending upon the extent of severity of restrictive tongue mobility caused by AG, corrective procedures could help alleviate breastfeeding difficulties [37]. In 2003 the World health organisation (WHO) and the United Nations Children's Fund (UNICEF) initiated a programme highlighting the importance of breast-feeding during a child's first year of life thus reigniting worldwide interest in AG as a condition [57]. The campaign included a message stating that failure to do so could be the primary cause for complete loss of a child's protective immunity due to latching difficulties [2, 37, 38A, 58]. Nevertheless, a review published in Cochrane in 2017 [42] suggested that corrective procedures may not consistently improve infant feeding but may improve maternal nipple pain [42]. Many systematic reviews focus on the clinical importance of carefully selecting infants for AG corrective surgeries through specific and consistent criteria to diagnose AG and by assessing breastfeeding functionality using breastfeeding assessments [48, 56]. Though AG assessment tools are available, there are disparities between clinicians internationally with respect to AG diagnosis and treatment due to lack of acceptance and/or tool application [48]. One such tool is the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF) [20B] endorsed by the Academy of Breastfeeding Medicine (ABM) due to the tool's comprehensiveness. HATLFF is a quantitative assessment aid for AG diagnosis in infants from 0-6 months. It is categorised into 5 "appearance items" which refers to the visual examination of the tongue when lifted with detailed evaluation of the elasticity of the frenulum, length of the frenulum, location of the attachment of the lingual frenulum to the tongue, and finally the location of the attachment of the lingual frenulum to the inferior alveolar ridge (IAR). Scores of 2, 1 and 0 are given accordingly to the most appropriate descriptive match of the infant during clinical examination [20A, 20B, 45] Another widely utilised tool is the Bristol Tongue-tie Assessment

Tool (BTAT) [26] which provides good inter-person reliability and objectivity with more rapid clinical implementation. BTAT is an AG diagnostic aid for children. Based on 4 separate criteria (extent of tongue protrusion, extent of tongue lift, anatomical appearance of the tongue tip and location of the attachment of the lingual frenulum to the IAR), tongue features are scored from 0-2. Zero scores in each category implies great severity, whereas a score of 2 in each category is the least severe. Both BTAT and Hazelbaker Tools possess highly correlative findings when utilised [26]

Other notable concerns in children with AG are speech problems. It is reported that untreated AG negatively affects speech in children, [17] with concerns over articulation [31]. According to a study by Meissner et al., 71% of children with AG did have speech problems because of restricted tongue mobility, [38A] however few articles agree that AG does affect articulation by restricting tongue mobility. [27, 38A] Thus the relationship between AG and speech affectation remains unclear and rather than wide reports on detrimental effects, the evidence is sparse and poor with only 23% of paediatricians having clinically discerned a relationship. [38B] The effects of procedures to correct AG related speech deficits have not been properly investigated. In 2020, Khan et al. explored untreated AG and its relationship with speech articulation, stating that there was evident lack of appropriate statistical data comparisons and too much heterogeneity between the examined studies, to draw any definitive conclusions [29]. Equally a systematic review in 2015 by Chinnadurai et al. revealed low-quality evidence with an elevated risk of bias and disparities in outcome measurements. These all contribute to inconclusive results regarding the effectivity of surgical procedures on speech outcomes [7]. Furthermore, a study by Daggumati et al. highlighted the inadequacies of a universally accepted guideline on AG management, contributing to the controversiality of the topic [9].

Selection of an appropriate mode of action requires taking into consideration factors such as: (a) corrective procedures (frenectomy/frenotomy/frenuloplasty), (b) treatment timing (age appropriation) accounting for the extent of functional limitation and possible detriment to the child's speech development – presenting a very real dilemma [34A, 38B, 39]. Publications state that if surgical intervention is required for feeding and speech correction, then “Frenotomy” is the most advocated option. Frenotomy is classified into (i) “simple-release” (frenotomy, frenulotomy, frenectomy) and (ii) “suturing-followed-surgery” (frenuloplasty) [29]. The positive effects on breast feeding efficacy are described in many clinical studies and

have increased in emphasis in the 21st century, [17, 56],_with an elevated demand for frenotomies in the last decade [28, 55]

An additional effect of untreated AG is the potentiality to generate psychosocial problems. Despite research focussing on the physical restrictions caused by untreated AG on feeding and speech, little to no research has been conducted on the possible psychosocial impact in children, such as behavioural and self-esteem problems and much is still unknown in this respect [34A]. Restrictions in licking of the lips/ice-creams, cleaning food debris from dentition and the inability to play wind instruments are all additional issues which if extenuated further into adolescence, could be a catalyst for teasing from other individuals within a social setting [34A,37]. A publication in 2003 by Lalakea and Messner states that any observable psychosocial impact in young children with untreated AG may not necessarily present in an obvious manner until late childhood, and though young children may be symptomatic, they may not necessarily be aware of /recognise their symptoms. Additionally, as children mature both physically and mentally, their appreciation and awareness of socially embarrassing situations could be a deterrent for them to report their symptoms unless directly questioned. [34B]

Therefore, the objective of this systematic review and meta-analysis is to: (1) investigate and highlight the impact of untreated AG on feeding, speech and the psychosocial domain in children between 0-15 years. (2) determine surgical outcomes of corrective AG procedures and their influence on feeding, speech and the psychosocial domain (3) highlight AG as an important myofunctional and psychosocial disorder, (4) encourage further collaborative research from healthcare professionals.

Articles revealing a link between AG and feeding, speech and psychosocial deficits, and articles describing the effect of corrective AG procedures on these issues were analysed.

METHODS:

Protocols and Registration:

This systematic review and meta-analysis was created in accordance with the Preferred Reporting System for Systematic Reviews and Meta-Analyses (PRISMA).

Data Sourcing Methodology:

An initial generalised search was conducted online only evaluating and searching through a multitude of databases as recommended by COCHRANE - The Central Register of Controlled

Trials, EMBASE, EBSCO, NCBI and MEDLINE. Only literature in English was searched for and articles not in English were discarded. The period from which online databases were searched for by both authors was from March 1st, 2023 – March 28th, 2023.

A MeSH search string of studies involving the following key words were included: (ankyloglossia OR lingual tongue tie OR short lingual frenulum OR frenectomy OR frenulectomy OR frenulotomy OR frenuloplasty) AND language OR speech OR articulation OR disarticulation OR misarticulation OR phonological) AND breastfeeding OR breast AND feeding) AND psychological AND effects) AND social AND effects) OR psychosocial AND effects)

Study Selection & Data Extraction

The populations, interventions, comparisons outcomes and study design (PICOS) model of clinical questioning for evidence-based medicine was utilised as the defining criteria for article incorporation. The PICOS point system [Supplementary Material Section: Table 1], helped to create a summarisation of included articles analysing feeding capabilities speech and psychosocial outcomes [Table 2] [17].

TABLE 2: P.I.C.O.S Summary of article evaluation of Ankyloglossia - Best overall score of 44

CRITERIA	MESSNER ET AL.	HOGAN ET AL.	BALLARD ET AL.	MASAITIS & KAEMPF	MARME T ET AL.	NOTEST INE	LALAKEA & MESSNER
Study population							
- Study Dropouts	4	3	2	1	2	N/A	N/A
- Study losses to followup	2	3	3	0	N/A	N/A	N/A
Intervention							
- Description of surgery type	2	2	2	2	3	2	1
Measurement of effect							
- Outcome measures	2	2	2	4	2	2	2
- Duration of followups	2	3	1	2	2	N/A	N/A
Analysis & Results							
- Analytical adequacy	4	4	2	2	2	0	0

- Adequacy of data presentation	1	1	1	0	0	0	0
TOTAL SCORED	35	35	26	22	15	7	5
N/A: Not Applicable							

TABLE 10: Inclusion and Exclusion Criteria. AG = Ankyloglossia (“Tongue-Tie”), RCT =Randomised Controlled Trials, LC = Lactation Consultants, SLP = Speech Language Pathologists; **a** < 36 weeks gestational period; **b** with specification of potential harm caused to the infant/child during/post-operative tongue release surgeries including but not limited to frenotomies, frenuloplasties and Z -plasties.

Classification	Criteria	
	Inclusion	Exclusion
Publication Dates:	Articles published from 01/01/90 - 31/03/23	Publications > 30 years old;
Language:	Articles in English	Articles not in English
Study Population: Age/Gender/Ethnicities	Human subjects between the ages of 0-15 years, Male and female, All ethnicities AG symptomatology present	Neonates/infants/children/ adolescents with Down syndrome, Pierre Robin syndrome, Opitz syndrome or any inheritable developmental craniofacial abnormalities. Premature neonates. [16] a ,
Study Designs	RCT’s, prospective studies, cohort studies, comparative studies, clinical trials, literature reviews, case reports b	Inadequately defined inclusion/exclusion criteria. Inappropriate comparative groups. Large discrepancies in sample sizes. Insufficient data to carry out appropriate statistical analysis. More than 5% of patients lost to follow up Studies only stating the composition ratio of language symptoms in hospitalized patients with respect to speech analysis. Inappropriate assessment and/or diagnosis of the extent of AG symptomatology on the effects of feeding/speech/the psychosocial domain of affected individuals due to: (i) Inadequate expert examination (i.e., for breastfeeding efficiency analysis – no LC examination. For Speech issues – no SLP examination. For psychosocial domain aspects – lack of / unrecorded interdisciplinary discussions between medical specialists.

		Abstracts only, editorials, directories, cross-sectional retracted studies, lectures, biographies.
Surgical Procedures Implemented and Analysis of Corrective AG Surgery Efficacies	<p>Procedure/Intervention: Surgical – frenulotomies, frenuloplasties, frenectomies.</p> <p>Comparisons: Surgical OR pre-surgical and post-surgical examination on breastfeeding, speech and psychosocial status of individuals.</p> <p>Outcome measures: Objective/subjective examination of:</p> <p>(i) Feeding - breast-feeding difficulties (specifically attachment and nipple pain), bottle feeding, bolus formation and movement, deglutition.</p> <p>(ii) Speech – development, pronunciation, articulation</p> <p>(iii) (Psycho)social aspects - self-esteem issues, sialorrhea, etc.</p>	Any non-surgical pathways for AG management including complementary medicinal methodologies e.g. speech therapy, stretching exercises, myo-facial release therapies, physical therapy etc.

In total 2 separate phases were conducted for the purpose of this systematic review: phase 1 (low level search) independently undertaken by both the lead author (FIG) and second author (CCS) with the purpose of collating, reviewing and evaluating the abstracts of articles against pre-determined exclusion criteria and irrelevant articles were discarded [Table 10]. Phase 2 (high- level search): was undertaken by the lead author (FG) who manually screened appropriate full-text, English only publications (namely RCT's, prospective studies, cohort studies, comparative studies, clinical trials, literature reviews and case reports), evaluating the impact of untreated AG on feeding, speech and the psychosocial domain of children between the ages of 0-15 years. FG manually reviewed all reference lists in order to ensure that all relevant publications were included for the development of this systematic review, cross-matching against pre-determined inclusion and exclusion criteria [Table 10]. The data collected by the lead author (FG) from all included full text articles were; author(s) names, publication year, study design, study populations (with respect to the investigated population age, gender and ethnicities), treatment interventions (surgical only), treatment outcome (success/failure).

The 3 primary areas of interest were independently scrutinised by the lead author (FG) undergoing a more in-depth analysis with both authors (FG and CCS) concurring to focus on the following within each field:

- (1) Feeding - breast-feeding difficulties (specifically attachment and nipple pain), effects on bottle feeding, bolus formation and movement, deglutition.
- (2) Speech – impediments in development, pronunciation and articulation
- (3) (Psycho)social domain - self-esteem issues, sialorrhea, teasing by peers, capacity to play wind instruments etc.

Surgical outcomes of corrective AG procedures over the past 33 years were also explored and included. As an additional area of evaluation, the lead author (FIG) assessed risk-benefit measurements associating AG with speech deficits and feeding difficulties. All calculations were checked at least 3 times and validated by the second author (CCS) [**Supplemental Tables 3 and 5**]

Extracted data by the lead author (FG) (including references of selected articles) was cross-checked by the second author (CCS). In instances where author discrepancy was evidenced in phase 1, articles were reanalysed and progressed to phase 2, with conflicts being resolved by the lead author (FG). Inter-reviewer reliability was determined utilising Cohen's Kappa test which was calculated at 0.58 and thus presumed adequate.

Quality Assessment Measures

For result reliability AXIS, GRADE and CASP tools were used for the purpose of this systematic review:

- (A) AXIS Tool consisting of 20 questions scrutinized data acquisition procedures from all included articles by considering: (i) adequacy in representation of participant population, (ii) inclusion of appropriate sample sizes, (iii) non-responding participants and the effect of the acquired statistical data on study reliability (iv) the effect on study outcomes.[12] [**Figure 2**].

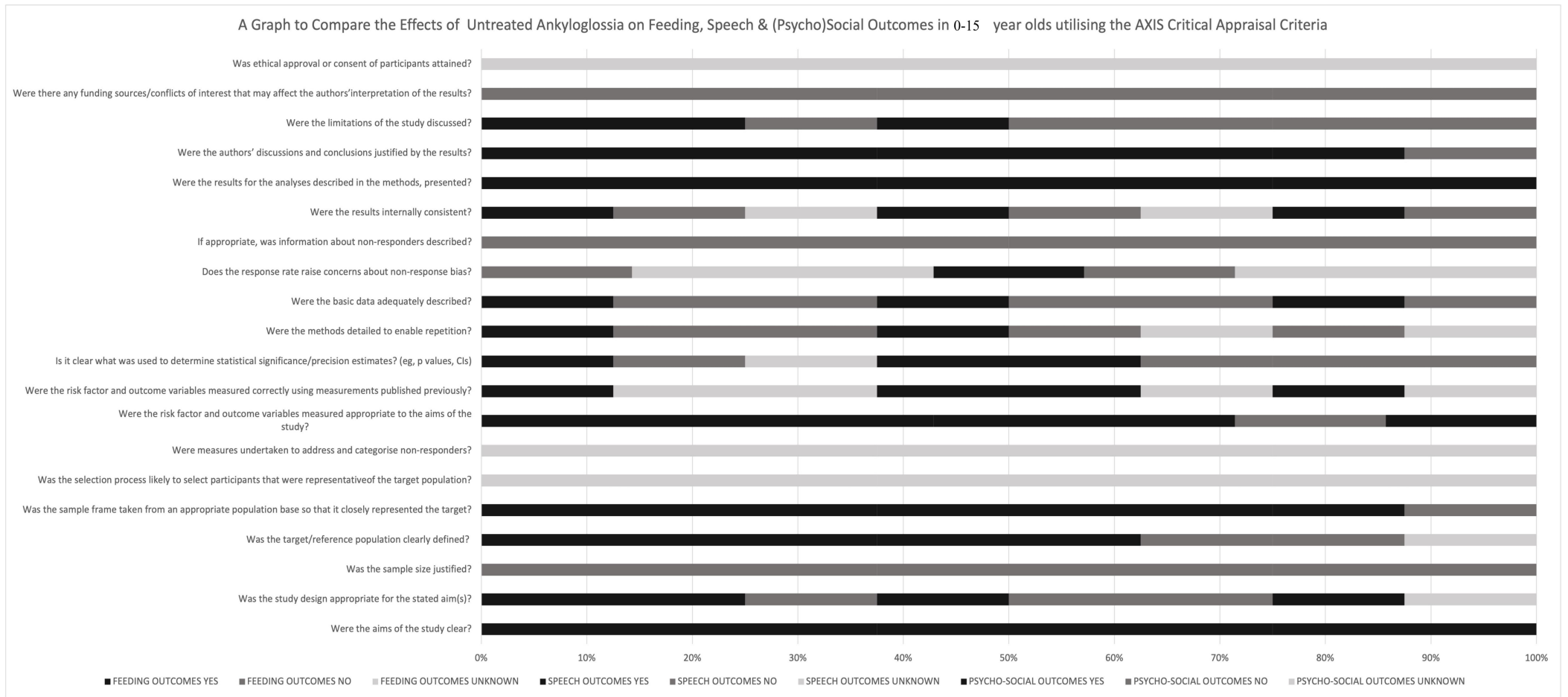


Figure 2: A Graph to Compare the Effects of Untreated Ankyloglossia on Feeding, Speech & (Psycho)Social Outcomes in 0 - 15-year-olds utilising the AXIS Critical Appraisal Criteria

(B) GRADE (Grading of Recommendations, Assessment, Development and Evaluation) system categorised into 4 levels of evidence: very low, low, moderate, and high, used to characterise the strength of evidence presented in the literature by evaluating scientific evidence based on intended outcomes, study limitations and study accuracy [Table 4].

TABLE 4: GRADE Evidence Profile Summarisation of the Quality of Evidence Gradations & Calculated Risk Ratios (RR) of negative symptomatology experienced by Untreated AG patients from the included Literature. RCT = Randomised Controlled Trials, AG = Ankyloglossia, RR = Risk Ratio, CI - Confidence Interval. *: Quality of evidence rating decreased from "high" to moderate" due to general concerns regarding the methodology of individual studies, validity of the outcome measures, and some variability in effects rather than a limitation in one category. **: Quality of evidence rating decreased from "high" to "moderate" and then to "low", due to elevated concerns sparsity of data availability, the use of trained specialists for objective assessment of the dependant variable, concerns regarding result bias, inadequate sample sizes. Contributes to an overall questioning of evidence strength. **a**: Surgical procedures - Frenotomy/Frenuloplasties/Frenectomies. **b** - Refers to reference articles: 27, 30, 37, 56, 59. **c** - Refers to reference articles: 2, 23, 36, 41, 56 **d** - Refers to reference articles: 33, 34A, 35

QUALITY ASSESSMENT						Summary of findings				
Number of Studies	Study Design	Study Limitations	Consistency	Directness	Other Considerations	Number of Patients	Effect	Relative 95% CI	Absolute 95% CI	QUALITY
SPEECH OUTCOMES										
5 b	Prospective Controlled Trials	Serious (-2) **	no important inconsistency	no uncertainty	-	Treated AG a 121/304 = 39.80%	Untreated AG 183/304 = 60.20%	RR 0.92	/1000	LOW (+++)
	RCT's, Prospective Case, Series,							0.59 - 0.98	(to)	
	Cohort studies, case control									

	studies, comparative studies									
FEEDING OUTCOMES										
5 c		Serious (-1) *	no important inconsistency	no uncertainty	-	Treated AG a 1363/2739 = 49.76%	Untreated AG 1376/2739 = 50.24%	RR 0.81	260/1000	
	RCT's, Prospective Controlled Trials,							0.45-0.90		LOW (+++)
	Cohort studies									
(PSYCHO)SOCIAL OUTCOMES										
		Serious (-2) **	no important inconsistency	no uncertainty	Sparse data availability	unclear	unclear			
3d	Case Series, comparative literature reviews									LOW(+)

(C) Critical Appraisal Skills Programme (CASP) TOOL, used to assess included review articles, questioning obtained and stated result validity, and examining the local applicability of result findings to the wider population [63] [Figure 3A and 3B].

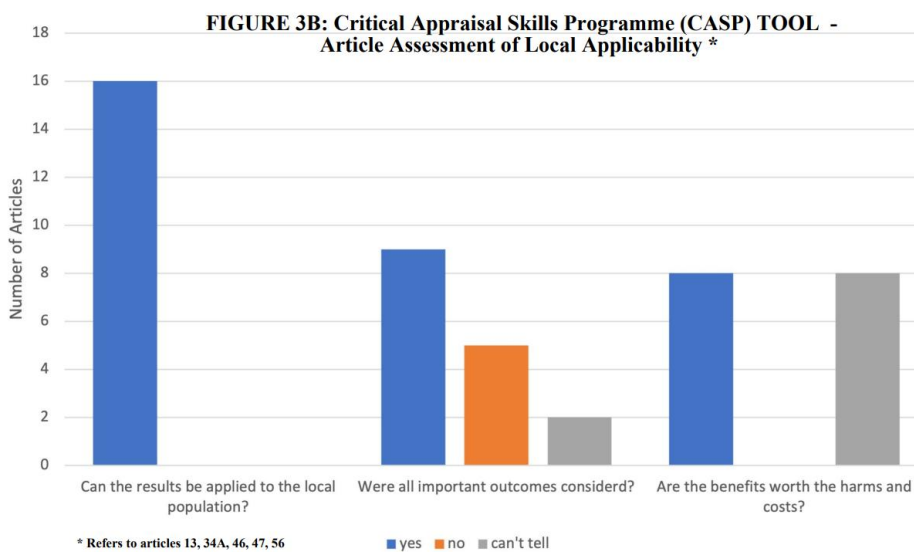
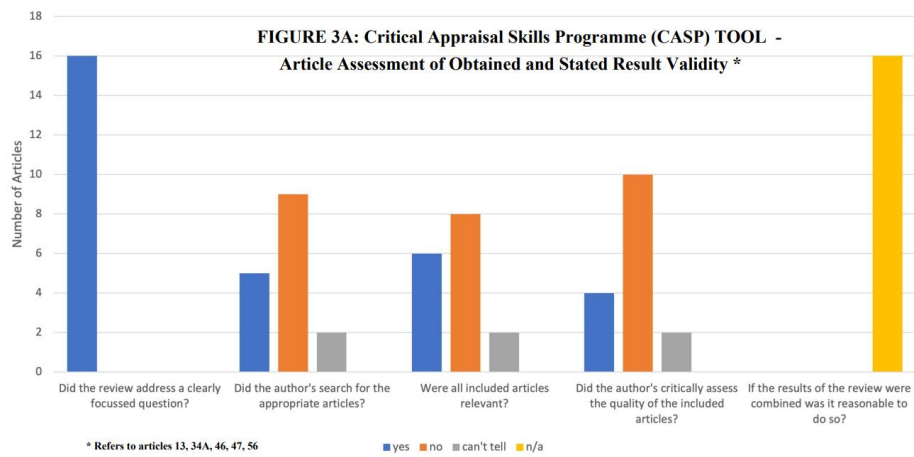


Figure 3: Summary of Critical Appraisal Skills Programme Tool (CASP) for the evaluation of Literature Reviews with respect to: (A) Result Validity & (B) Local Applicability

Outcome Measures

The primary evaluated outcomes are (1) to investigate and highlight the impact of untreated AG on feeding, speech and the psychosocial domain in children between 0-15 years. (2) determine surgical outcomes of corrective AG procedures and their influence on feeding, speech and the psychosocial domain (3) highlight AG as an important myofunctional and psychosocial disorder, (4) encourage further collaborative research from healthcare professionals.

Statistical Analysis

A meta-analysis was performed on the extracted data where appropriate (i.e., data collected from RCT's, prospective and cohort studies etc). The odds ratios and 95% confidence

intervals were calculated from these study types (Figures 4 and 5). Microsoft Excel and SPSS Software Programmes were utilised for data analysis and graphical representation. The primary author (FIG) evaluated risk-benefit assessments measuring associations between AG and feeding difficulties and AG and speech deficits were generated via calculation of measures of association involving: (1) absolute risk differences (ARD's), (2) numbers needed to treat (NNT's), (3) numbers needed to harm (NNH's), (4) relative risk reductions (RRR's) [Supplementary Material: Tables 2 – 5]. No risk –benefit assessment measurements for AG and psychosocial domain affectations were generated due to a lack of data availability. Data is summarised utilising a mixture of descriptive and illustrative statistics, with the results of the quality assessment of included articles being presented in tables or figures [Figures 3-5, Table 4].

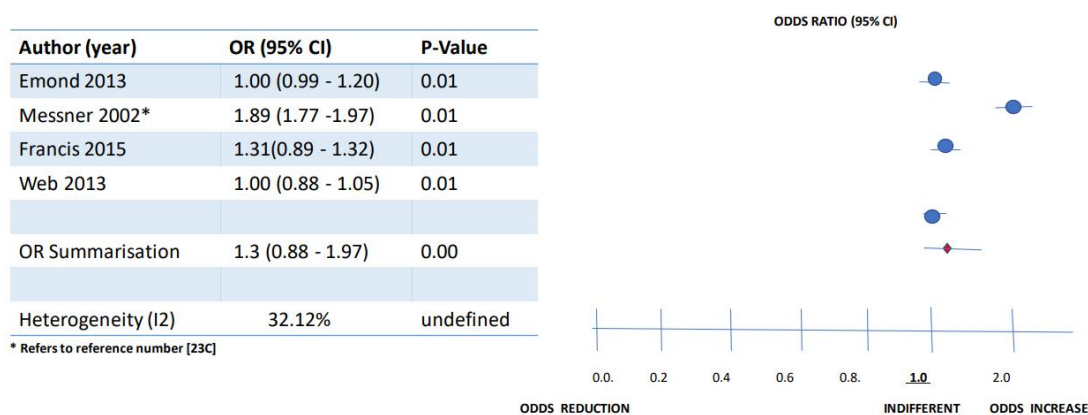


Figure 4: Results Summary of Articles Included in the AXIS Tool Analysis of Stated Feeding Difficulties Experienced by Untreated AG Patients

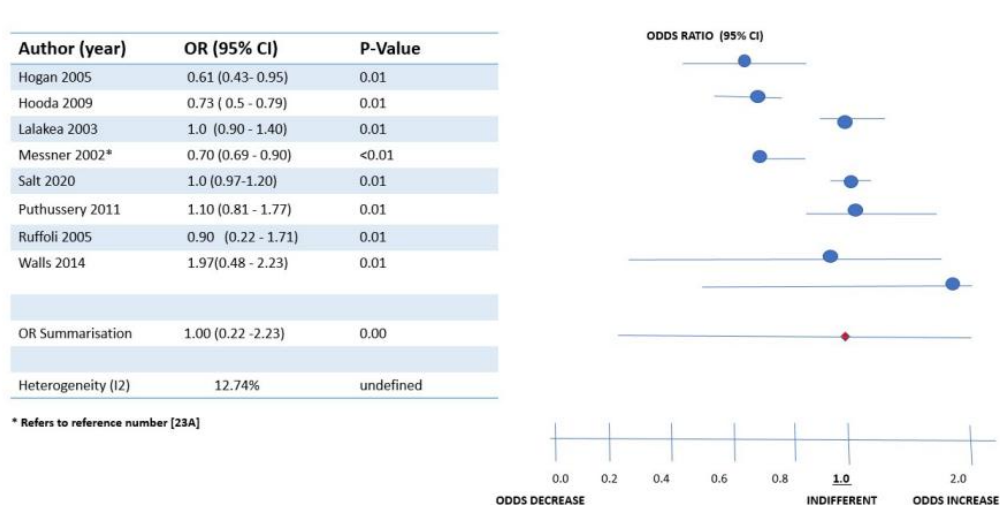


Figure 5: Results Summary of Articles Included in the AXIS Tool Analysis of Stated Speech Difficulties Experienced by Untreated AG Patients

RESULTS:

Search Results

Initially 1808 articles were retrieved including duplicates. 810 studies were excluded and following the elimination of duplicates, 998 relevant articles were obtained. 44 articles were eligible for inclusion; however, 14 articles were further excluded due to ambiguous data. 30 articles were included in our final review [Figure 1].

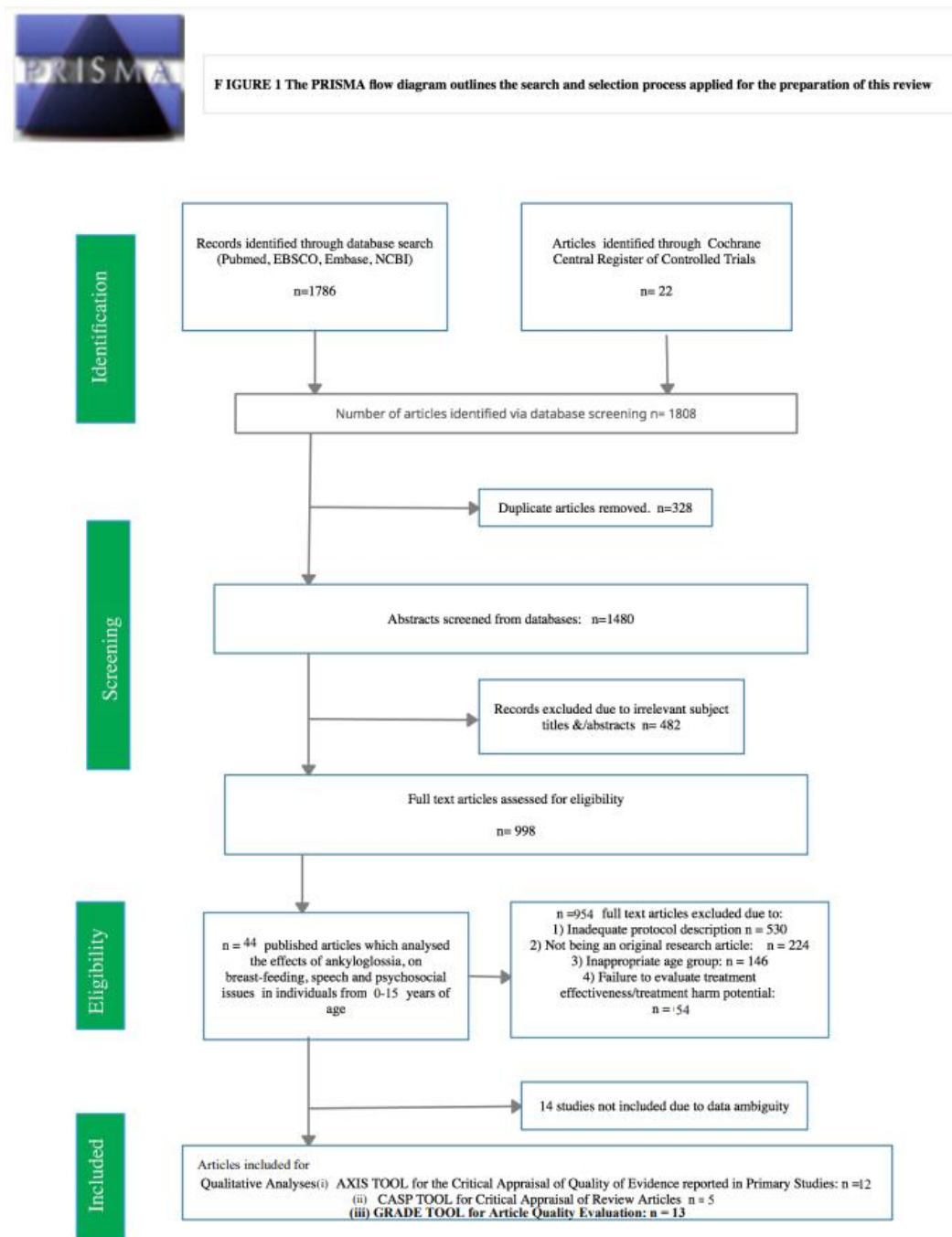


Figure 1: The PRISMA Flow diagram outlines the search and selection process applied for the preparation of this review.

Excluded articles were removed due to: (a) Poor protocol descriptions, (b) inadequate descriptions of the inclusion/exclusion criteria, (c) poor validation of assessment methods (d) inappropriate age group (e) the inclusion of ill-defined, non- consecutive samples of data (f) insufficient sample sizes/ power analysis. Refer to table 10 for further details.

A total of 15 publications were retrieved for AG association and feeding difficulties, 12 publications for AG association and speech difficulties and 3 publications for AG and its affectation of the psychosocial domain. From the 30 articles included, 12 articles underwent AXIS Tool and Meta-critical analysis - (13, 17, 23, 24, 34B, 38A, 38B, 43, 46, 47, 54, 56) [Figures 4 and 5] - 5 underwent CASP Tool analysis - (13, 34A, 46, 47, 56) [Figures 3A and 3B], and 13 underwent GRADE tool evaluation (27, 30, 37, 56, 59, 2, 23, 36, 41, 56, 33, 34A, 35) [Table 4]

Analysis of Study Quality

From the articles included, AXIS tool analysis revealed 45% of articles agreed untreated AG negatively affected feeding. Likewise, 43.3% and 27.5% of publications agreed that untreated AG negatively affected speech outcomes and psycho-social outcomes respectively. In comparison, 30.0% of articles stated that untreated AG did not influence feeding, and the same percentage of publications independently evaluating speech and psychosocial outcomes (35%) concurred that untreated AG did not influence these variables. A quarter of articles (25.0%) were undecided with respect to the influence of untreated AG on feeding capability. A further 15.0% and 25.0% of articles could not decide whether untreated AG affected speech and psychosocial outcomes respectively [Figure 2].

Commenting on result validity and local applicability of data collated by the studies, the CASP tool graphs demonstrate lack of result validity with respect to the author's manner of critical data assessment, search methodology for the inclusion of relevant data, and the proportion of relevant articles included [Figure 3A]. Comparatively, our investigation into overall applicability of local results demonstrated that the authors of more than half of the included articles considered all the important outcomes.

Furthermore, 50.0% of authors were unable to confidently state whether individuals who had corrective procedures benefitted more than the side-effects they experienced (such as excessive and prolonged post-operative bleeding etc) [Figure 3B]. The appraisal of evidence

strength as per the GRADE system revealed that articles evaluating feeding, speech and psychosocial outcomes of individuals with untreated AG was of “LOW quality”

Statistical Evaluations of Combined Results:

The included articles represent a total of 3,043 patients who were enrolled in studies investigating the effects of ankyloglossia in children between the ages of 0 - 15 years on feeding and speech. Due to sparse data availability and the nature of the studies included (i.e., descriptive studies), numerical data for the psychosocial domain were unable to be calculated.

Analysis of AG on Feeding Efficacies:

A total of 2,739 individuals were analysed, of which 1363 (49.76%) received corrective AG surgery in the form of frenotomies/frenectomies. 1376 (50.24%) never received AG treatments [Table 4]

The risk ratio (RR) was calculated as 0.81 [Table 4], implying that there is a 0.81 elevated risk of a poor consequence (i.e., difficulty in latching onto the nipple for breastfeeding infants resulting in inadequate nourishment) in infants with untreated AG thus affecting feeding ability. Comparatively, the group which had corrective procedures, tabulated 95 % CI were between 0.45 - 0.90 [Table 4]. GRADE Tool for study quality was “LOW” [Table 4]. Heterogeneity between studies was determined by the I² statistic and calculated using the test-based method of Higgins and Thompson 2002 [22B], to create a “fixed effects model” [Figure 4]. I² value was 32.12% classifying the heterogeneity between studies as “unimportant - moderate” [Figure 4].

The most investigated and reported negative effect of untreated AG is its impact on feeding ability, especially on breastfeeding in infants [2, 17, 23, 39, 54]. Our results demonstrate this trend.

Studies were carried out in a variety of geographical locations including the United States of America [54] and the United Kingdom [23, 39] with presented data comparisons [39, 54]. Articles examined the effects of frenotomy on post-operational and functional improvement of tongue-tie in neonates, relying on parent reported outcomes and followed their progression over an average period of 3 years [23, 39, 54]. Three principal functional tongue movements (i.e., ability to clean their teeth, licking the outside of the lips and eating ice- cream) were

objectively examined and rated. Results showed no significant difference between those who had corrective frenotomy procedures and those without AG. This implies that corrective procedures improved tongue mobility and function in AG patients [23, 35, 39, 46, 54].

Analysis of AG on Speech Affection:

A total of 304 individuals were recruited, of which 183 (60.20%) had not received corrective AG treatments. 121 (39.80%) had received corrective AG surgical treatments in the form of frenotomies/frenectomies [Table 4]. The calculated risk ratio (RR) was 0.92 [Table 4], implying that there is a 0.92 elevated risk of a poor consequence (i.e., speech deficits) in patients who have not had corrective procedures, in comparison to the group who have had corrective procedures. Tabulated 95 % CI were between 0.59 - 0.98. GRADE Tool for quality assessment was “LOW” [Table 4]. The calculated I2 value was 12.74% classifying the heterogeneity between studies as “unimportant” [Figure 5].

Literature suggests that the second most prevalent concern after feeding difficulties was speech. Several distinct outcome variables were measured across the included studies, with most studies including intelligibility and articulation assessments. Two studies used a speech-language pathologist to determine the extent of detriment to both intelligibility and articulation [11, 21], whilst another study used information provided by the parents [54].

Some articles compared surgical alternatives to frenotomy [11, 54], whilst others evaluated the efficacy of the frenotomy procedures, reporting significant improvement in intelligibility and articulation post- treatment of children affected with AG. However, findings relating to speech fluency, word formation and enunciation were not presented [11, 21, 55]. Though some studies agreed there were speech improvements post- surgery, these articles placed more influence upon the safety of corrective procedures, rather than on the extent of speech improvement as a key outcome [8,31,35, 41].

Despite articles agreeing that untreated AG contributes to speech difficulties, the data presented focussed more on the advantages of corrective procedures and were less focussed on evaluating the extent of speech difficulties experienced by untreated AG patients (e.g., the ability to pronounce “D”, “T”, “N” etc) - which was the purpose of our systematic review. Thus, the assessment of the quality of evidence was evaluated to be “LOW” as per the GRADE system [Table 4].

Analysis of Untreated AG on the Psychosocial Domain:

There was difficulty in locating literature focussed solely on the psychosocial effects of untreated AG on individuals between the ages of 0–15 years, that was considered of sufficient quality to be included in this systematic review.

Two review articles implied that there could be a link between untreated AG, and the accompaniment of social embarrassment with a possibility of attributing to low self-esteem experienced by children due to teasing from peers [24, 36].

Other included literature reported primarily upon the significant improvement in the ability of the children to lick outside the lips, improvement in the ability to lick ice-cream, and in the ability to clean the teeth utilising the tip of the tongue when compared with untreated individuals [11, 21, 54]. All these factors relate more towards the improvements of “social” aspects that corrective AG intervention has and failed to report on the possible psychological ramification that individuals with untreated AG had or could have in the future (such as self-esteem issues, behavioural problems etc).

Therefore, due to: (a) sparsity in available data (b) studies with small participant number and management of outcome limitations with respect to tongue mobility and (c) lack of unbiased methodological standardisation to assess and compare child behaviour, the quality of evidence was deemed “LOW” as per the GRADE system [Table 4]

Assessment of Harms: Feeding and Speech

The number needed to treat (NNT) and number needed to harm (NNH) values were used to examine the benefits-risk of surgery (frenectomy, frenotomy, frenuloplasty) on speech and feeding in subjects between the ages of 0-15 years with AG [Supplementary Material: Tables 3 and 5]. NNT/NNH analysis was conducted on feeding and speech affectations. The considered criteria evaluated for feeding were specifically improvements in attachment capabilities, reduced nipple pain, bottle feeding, bolus formation and movement, deglutition. For improvements in speech, development, pronunciation and articulation were examined. The risks considered were errors occurring during AG surgery such as excessive bleeding, Wharton duct injury, airway compromise, lingual dysfunction and infection. Calculations were done on all included articles reviewed by FIG and CCS, with no reports of harmful effects within the scope of treatments undertaken by older children.

DISCUSSION:

The objective of this systematic review and meta-analysis was to: (1) critically assess and highlight the impact of untreated AG on feeding, speech and the psychosocial domain in children between 0-15 years. (2) determine surgical outcomes of corrective AG procedures and their influence on feeding, speech and the psychosocial domain (3) emphasise AG as an important myofunctional and psychosocial disorder, (4) encourage further collaborative research from healthcare professionals.

Study Characteristics

Data was evaluated from an aggregate of 3,043 patients with average patient numbers at 31.6/study. AG prevalence reported in literature varies from 0.27 % - 10.69 % [2, 16, 18, 23, 40, 52, 53]. The findings reveal a lack of standardisation for AG classification and diagnosis, as there has yet to be a universally accepted tool for the identification and assessment of AG within a clinical setting. HATLFF is one of many diagnostic tools used by several studies due to its reliability [45], however, not all studies presented statistical data well, with included studies reporting a lower prevalence of AG identification and diagnosis when routine clinical examinations of oral mucosa were done as reported in epidemiological studies (0.27% to 4.24%) [18, 23, 40, 53] in comparison to studies exclusively reporting on AG prevalence - 4.14% and 10.69% respectively [2, 23]. The high diagnostic variability (mainly including tongue mobility or attachment point of the lingual frenulum) [48] was often demonstrable via absent p-values, confidence intervals and standard deviations – thereby complicating data calculations. Thus, in some instances the interpretation of figures presented within our findings is more descriptive.

In neonates, AG prevalence was reported to be between 2.38% and 10.69% [16, 23]. This was significantly higher than in studies investigating children or adolescents (0.27% - 4.24%) [18, 23, 40, 53]. These findings suggest that individuals suffering from the milder form of AG, may spontaneously resolve oral-functional muscular impediments as the maxillary and mandibular arches develop [16, 23, 18, 40, 53].

Numerous publications have evaluated the relationship between AG and breastfeeding difficulties, including the positive outcomes of surgery [17, 42]. One retrospective cohort study provided data focussing on feeding difficulties (other than breast-feeding complications) experienced by young individuals with AG [32]. The case group underwent AG surgery

(frenotomy), after its detection by specialists within the first month. The extracted data from the intervention group was compared with another group whereby the mothers were offered AG surgery with all other variables remaining constant (i.e., after detection of AG within the first month of life, and frenotomy as the surgical intervention being offered) yet rejected the proposal [32]. The article reported positive results in feeding when AG surgery was done [32]. Further review and clinical trial studies corroborate the increased efficacy of breastfeeding once surgery is completed, with positive effects such as nipple pain reduction [17,50]. Antithetically, an RCT by Emond et al. in 2014 [13], concluded that early intervention via surgery (namely frenotomy) did not improve breastfeeding in infants diagnosed with mild-moderate AG [13].

Currently surgery for AG is not standard practise unless congenital AG is diagnosed alongside strongly expressed maternal breast-feeding difficulties [2, 8, 17,21, 23, 31, 32, 37]. A systematic review in 2013 by Web, Hao and Hong [56] verified that AG surgeries are effective in providing objective and subjective benefits to AG patients. This was based on quality evidence in a small selection of studies. [56]. Five prospective RCT's conducted between 2004 – 2012 focussing on the benefits of frenotomies on feeding difficulties, demonstrated its effectivity in alleviating and improving AG symptomatology in patients with severe AG [3, 4, 5, 10]. However, a larger body of evidence is necessary before frenotomies are recommended as corrective procedures in patients with mild AG symptomatology. Our findings expose a gap for research into the effects of untreated AG on feeding outcomes in older children, specifically on mastication, bolus formation and deglutition.

Overall, the methodological limitations in measured outcomes of included articles were of “LOW” quality. Datasets were generally undersized, imposing limitations on the determination of medium-long-term benefits of procedures on feeding outcomes. There was unimportant – moderate heterogeneity between the included studies [Figures 4 and 5] and unvalidated assessment methods. Further research would generate impact and confidence in the estimate of effect, thus positively influencing re-grading scores [61].

In speech outcome data, 5 studies were discovered [11, 18, 21, 47, 54]. An RCT by Heller et.al in 2005, compared two varying techniques for a frenuloplasty procedure (“4 flap Z shaped” or “horizontal - vertical shaped”) in children with an approximate age of 6 years, whereby intelligibility and articulation were compromised [21]. The study revealed significant improvement in articulation as determined by specialised speech-language pathologists. However, when children’s speech with specific ratings for regularity were

evaluated, result outcomes were not as positive. Evident bias relating to: (a) which health care professional (HCP) carried out the procedure and (b) the location of surgeries in specialist tertiary care centres was apparent [11, 54].

One cohort study had a relatively small sample size of subjects ($n = 23$) with an approximate age of 6 years, grouping them into those who had AG surgery and those without surgery [11]. The control group consisted of children without AG. Surgeries were done at an Israeli tertiary urban centre, and a standardised speech assessment tool was implemented by speech pathologists to objectively assess subjects' post-surgical speech outcomes [11].

The second cohort study targeted 3-year-old children who had AG surgery as neonates and compared their speech outcomes with children who never had AG [54]. The validity of this study is questionable due to: (i) omitting reasons why infants were presented for treatment (ii) absence of objective assessment of subjects' speech via specialist speech and language pathologists. (iii) increased risk of bias as parents of AG sufferers would communicate greater concerns over speech deficits than those whose children with no AG diagnosis.

Three studies [9, 47, 54] compared speech disorders in both children and infants with and without AG. 1857 children of similar age were observed. Walls et al. [54] reported a significant difference ($P = 0.01$) between parental speech perceptions in 104 infants, all 3 years of age, split into 2 groups; those with AG with no surgical correction and those without AG, through a retrospective cohort study in 2014. Salt et al 2020, [47] did a preliminary study of 59 participants concluding that there was no significant difference in speech outcomes, intelligibility and tongue mobility in the 3 comparison groups; those without AG; those with AG and who have had surgery; those with AG and who have not had surgery.

In 2002, a cross-sectional study of 1694 children by Garcia et. al [18], investigated the effects of an anatomically 'short' lingual frenum on pathologies other than breast-feeding. The incorrect pronunciation of the "double r" phoneme was classified as dysglossia. [18]. The study found a statistically significantly difference between having a short lingual frenum and dysglossia ($OR = 0.02$; $95\%CI = 0.00 - 0.015$) [9]. Despite a large sample size, it scored "LOW" on quality as per GRADE analysis. A positive correlation between untreated AG and its negative effects on speech articulation was undetermined in our study due to a lack of high-quality evidence in literature.

For AG correction, surgery is generally recommended as the disorder is due to anatomical and structural abnormality [55]. The 2 primary surgical techniques advocated are: Frenuloplasty

or Z-Plasty (sutures after incision), purposefully used to release “tight lingual frenulum”, and frenulotomy, frenotomy or frenectomy (simple release of tight lingual frenulum without sutures).

A study by Lalakea et al [34A] discovered that despite half of patients having subjective complaints over speech, all individuals possessed comparatively normal speech when contrasted with patients without AG. In 2013, Camargo et.al demonstrated improvements in speech articulation post AG surgery as a result of tongue mobility improvements. However, improvement in speech after AG surgery was relatively unchanged due to incomplete re-establishment of temporal control [6].

Our investigation revealed 4 cohort studies with contradictory outcomes to those described previously. The studies compared speech between individuals who had AG surgery and AG patients who had no interventional procedures. Walls et. al [54] was the only study which denoted significant improvement in speech. However, the 3 other articles [9, 11, 47] differed in their outcomes as they all used expert medical specialists to judge speech outcomes of patients – all of whom stated that there was no significant difference in speech between AG treated and AG untreated cohorts.

The psychosocial effect of untreated AG in our age group compared to those who have undergone AG surgery was scrutinised. Our study showed that (i) articles were scarce (ii) articles were retrospective in nature (iii) investigations were of poor quality (iv) included studies focussed primarily on social impact of AG and paid little attention to the psychological detriment of untreated AG on subjects. One study considered the extent to which AG contributes to sialorrhoea and complicates maintenance of adequate oral hygiene and the negative social implications experienced by individuals [41] Another study based its assessment of social outcome on parental reports alone, creating result subjectivity thereby introducing significant levels of bias and unreliability [54]. Furthermore, assessment of social outcome inhibitions were limited to the ability to eat ice-cream, lick the outside of the lips, and teeth cleaning. Reasons were unclear as to the possible implications of these issues [54]. No other comparative study was found which included a section on the psychosocial impact of untreated AG on individuals from 0–18 years. Two further articles implied that there could be a link between untreated AG and social embarrassment with a possibility of attributing to low self-esteem due to teasing from peers [54 ,55].

Key strengths of our review included the spectra of databases searched, and the large number of included articles, thereby strengthening our statistical analysis. Additionally, all included

articles and their reference lists were thoroughly examined by both authors, and cross-matched against pre-determined inclusion and exclusion criteria. A rigorous evaluating tool (“AXIS”) to scrutinize information presented in primary research articles, permitted a more detailed assessment of individual aspects of study design. Subjective permission of greater flexibility by incorporating quality of reporting with risk of bias when assessing articles, gave our review a clear advantage over other appraisal tools such as the ‘Cochrane Risk of Bias Tool’ which fails to address poor quality reportage [22B]. Our use of the GRADE Tool for added critical evaluation of the authors’ presented evidence adds clarification to our findings. By using the NNH/NNT framework for quantitative benefit-risk assessment provided: (i) a simple and quantifiable approach for risk and benefits assessment (ii) a clear, transparent and comprehensible risk-benefit overview of our data [44].

Limitations of this review were evidenced by differences in the results and conclusions drawn by the included articles. The most critical limitation was that of a concrete diagnosis of AG, which was a result of deficient standardisable and validated criteria/tests ranging from evaluations of lingual frenulum functionality (as defined in the Hazelbaker’s tool, to frenulum length. Secondly, studies which included the results of subjective parental responses instead of speech assessment by medical professionals, introduced bias into the findings, thereby reducing the quality of evidence. Thirdly, included articles possessed reduced sample numbers and were of “low quality”. Fourthly, the majority of included studies were observational, increasing the probability of introducing bias in subject identification and selection in our 3 investigatory aspects; feeding, speech and the psychosocial domain. Finally, despite our study uncovering several articles on AG (especially its affectation upon feeding and speech), there is still deficient data availability strongly indicating a lack of sound knowledge base of clinicians [53], potentially influencing treatment decisions and information provided to the patient/ patient’s parents regarding AG and its management.

CONCLUSIONS:

Our findings were inconclusive with respect to the assessment of the effects of untreated AG in children between 0–15 years on feeding, speech and psychosocial outcomes. This review uncovered that AG surgery can be of some benefit to improve feeding and speech, although little is known about its psychosocial effects on individuals from 0-18 years. Many articles reported similar concerns by clinicians who agreed that untreated AG can present feeding and

speech difficulties later in life, with a chance of affecting sociability – however concrete data demonstrating this is questionable and scarce - this doubtfulness is reflected in our study.

The principal reason why the assessment of study quality of the included articles is challenging to clarify, is due to the lack of a universally accepted definition of AG. For appropriate management, effective interdisciplinary communication between specialists within the fields of dentistry and medicine is vital to aid clinical decision making. Our review outcomes rely on informed decisions sourced from specialists such as speech language pathologists, feeding specialists and lactation consultants to name but a few. Depending on the severity of AG and deciding if surgery is necessary, it is imperative for the treating clinician to carry out a risk-benefit evaluation prior to surgical referral.

Comparatively fewer investigations into alternative non-invasive treatments of young patients with AG have been done, to determine their effects on feeding, speech and the psychosocial domain. Our review opens a potential window for further research into this aspect, with the capability of revealing unexplored benefits. Information relating to possible psychosocial effects that patients with untreated AG may experience, could be a novel area for research. With this in mind, clinicians could provide an overall better quality of care as well as being better practised in informing and providing appeasement for anxious parents.

Authors contribution statement: FIG analysed and interpreted the patient data regarding the impact of untreated ankyloglossia on feeding, speech and the psychosocial domain in individuals between 0-15 years and was a major contributor in writing the manuscript. FIG and CCS reviewed included articles. FIG cross-checked extracted data and all presenting disputes were resolved by CCS. All authors read and approved the final manuscript.

Supplementary Tables

TABLE 1: P.I.C.O.S Point system criteria created for the purpose of assessing surgical intervention for AG correction on speech and feeding. **a** Tongue release surgery. **b** Interventional surgeries including but not limited to frenotomies, frenuloplasties and Z -plasties. **c** Development and pronunciation and articulation. **d** Breast-feeding, bottle feeding, bolus formation and movement, and deglutition. **e** inability to socially interact with individuals from the same/similar peer-group, evidence of bullying (past/present), self-esteem issues.

STUDY POPULATION		MEASUREMENT OF EFFECT
Patient Selection		Outcome measures. 1 point/criterion
0	Not mentioned	Speech complications c
1	Only exclusion criteria stated	Feeding complications d
2	Inclusion + Exclusion criteria stated	Psychosocial inhibitions e
Study Design		Objective assessment of tongue mobility
1	Case Reports	Speed of infant weight gain analysis
2	Cohort Studies	Duration of followup
3	Retrospective case- control studies	1 < 1 month
4	Prospective case-control studies	2 1 - 3 months
5	Randomised controlled trials	3 > 3 months
Sample Size		ANALYSIS & RESULTS
1	<100	Analytical adequacy
2	100-1000	0 None
3	>1000	1 Verbally recorded
Patients with Untreated AG (N)		2 Presented in tabulated format
1	<50	3 Basic statistics performed
2	50 - 100	4 Systematical analysis
3	>100	Adequacy of data presentation
Patients with Treated AG (N) a		0 No data presentation
1	0-10	1 Some data presentation
2	10-100	2 All data was presented
3	>100	

Stated Patient Observations prior to interventions. 1 point/criterion

Age

Sex

AG severity

Feeding issues:

Severity of nipple pain and trauma

Latch/suck evaluation of breastfeeding

Assessment/mention of infant weight

Difficulty transitioning to solid foods

Evidence of being a “picky eater”

Speech issues:

Evident frustration when communicating

Difficulty in getting words out/trouble with sound formations “m”, “t”, “th”,
“l”, “b”, “s”

Evidence of a stammer/stutter

Mumbling/speaking softly

Psychosocial issues:

Low self esteem

Bullying/teasing by other peers (past/present)

Timid/shy/quiet behavioural characteristics

Observable lack of/difficulty with interacting socially amongst peers of similar age groups

Study Dropouts

0	Not mentioned
1	Stated + >10
2	Stated + <10
3	Stated + = 0
4	Stated + reason

Losses to followup

0	Not mentioned
1	Stated + > 25%
2	Stated + <25%

INTERVENTION

Description of surgery type. 1 point/criterion b

Post-interventional examination on breastfeeding

TABLE 2 - Data amalgamation from information presented from the included Literature of patients with reported speech deficits with and without a poor outcome, and person-years of untreated AG and treated AG groups. *: Referring to reported harmful effects of post-surgical procedures, problems with speech associated with restriction in tongue function and mobility.

Reported Poor outcome *				
	Yes	No	Total	IR (2 d.p)
CONTROLES (no surgery)	71	50	121	71/121 = 0.59
CASES (surgery)	117	66	183	117/183 = 0.64

TABLE 3: Risk-Benefit Assessments measuring associations between AG and Speech Deficits*

Measure of Association	Calculation	Estimate
ARD	$(71/121) - (117/183)$	-0.053 = -5.30%^a
NNT	$1 / (71/121) - (117/183)$	19 persons/1000 subject years^c
NNH	$1 / (0.64 - 0.59)$.	20 patients/1000 subject years^d
RRR	$(1 - 0.92) \times 100$	8.0%^b

Abbreviations: ARD - Absolute Risk Difference, NNT - Number Needed to Treat, NNH - Number Needed to Harm RRR - Relative Risk Reduction.

***a:** The probability of acquiring speech difficulties in symptomatic AG patients after surgery, **b:** The likelihood of severe harm/ a potentially life-threatening event occurring in symptomatic AG patients whilst undergoing Surgery, **c:** A measurement of the number of AG patients which would need to undergo surgery in order to determine the positive/negative/unaffected impact on speech post-surgery, **d:** The average number of patients required to be exposed to possible risks of AG corrective surgery in an average of 1 person who would not have been harmed otherwise.

TABLE 4 - Data amalgamation from information presented from the included Literature of patients with reported feeding problems with and without a poor outcome, and person-years of untreated AG and treated AG groups. *: Referring to reported harmful effects of post surgical procedures, problems with feeding associated with restriction in tongue function and mobility.

Reported Poor Outcome *	Yes	No	Total	IR (2 d.p)
	CONTROLS (no-surgery)	173	1160	1376
CASES (Surgery)	216	1190	1363	216/1363 = 0.16
NNH (1/(It-Ic))	1/(0.16-0.13) = 33 patients			

TABLE 5: Risk-Benefit Assessments measuring associations between AG and Feeding Difficulties*

Measure of Association	Calculation	Estimate
ARD	$(173/1363) - (216/1376)$	-0.030 = -3.0%^a
NNH	$1 / ((173/1363) - (216/1376))$	34 persons/1000 subject years^c
NNT	$1 / (0.16 - 0.13)$	33 patients/1000 subject years^d
RRR	$(1-0.81) \times 100 =$	19.0%^b

Abbreviations: **ARD** - Absolute Risk Difference, **NNT** - Number Needed to Treat, **NNH** - Number Needed to Harm, **RRR** - Relative Risk Reduction.

***a:** The probability of acquiring speech difficulties in symptomatic AG patients after surgery, **b:** The likelihood of severe harm/ a potentially life-threatening event occurring in symptomatic AG patients whilst undergoing Surgery, **d:** A measurement of the number of AG patients which would need to undergo surgery in order to determine the positive/negative/unaffected impact on speech post-surgery, **c:** The average number of patients required to be exposed to possible risks of AG corrective surgery in an average of 1 person who would not have been harmed otherwise.

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