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CLINICOEPIDEMIOLOGICAL FINDINGS AMONG CHILDREN AND ADOLESCENT PRESENTING WITH SUSPECTED ANKYLOGLOSSIA IN OTORHINOLARYNGOLOGY PRACTICE AT A TERTIARY INSTITUTION

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ABSTRACT

Background: **Ankyloglossia is a congenital oral anomaly with varying timing and modality of presentation. This study aimed to determine the epidemiology, clinical presentations, and management of children and adolescents with ankyloglossia at an otorhinolaryngological practice at a tertiary institution in Nigeria.**

Methods: **This was a cross-sectional, hospital-based study of all children and adolescent patients who presented with tongue-tie in the Ear, Nose and Throat Department of Ekiti State University Teaching Hospital, Ado Ekiti, Nigeria. The study was carried out over a period of 10 years. Structured interviewer-administered questionnaires were administered to consenting patients to obtain data. Data obtained were collated and analysed using SPSS version 23. Data were expressed using frequency tables, percentages, bar and pie charts.**

Results: **A total of 18,431 patients were seen over the study period (2014 to 2023) out of which 94 (0.51%) presented with complaints of tongue-tie and 43 (0.23%) were diagnosed with ankyloglossia. Among the patients who presented with tongue tie, the peak age group was 1-5 years. There was male preponderance with a male-to-female ratio of 2.8:1.0. Urban dwellers accounted for 59.6%. There was no family history of ankyloglossia in the majority of the cases. Parents brought the patients in 45(47.9%) of the cases. The commonest presenting complaint was challenge with verbal communication among the patients as seen in 51(54.3%) of the patients presenting with tongue tie. No prior intervention was instituted in 8.5% while family physicians treated 43.6%. Normal tongue was recorded in 51(54.3%) cases. Ankyloglossia was diagnosed in 43(45.7%) cases, and other diagnoses included delayed developmental milestones in 23.4% and articulation disorder in 19.1%. In those with confirmed ankyloglossia, the commonest degree of ankyloglossia was mild (class 1) ankyloglossia. The commonest surgical treatment offered to patients diagnosed with tongue- tie was frenulotomy in 33(76.7%) cases**

Conclusion: **Understanding the anatomical definition of ankyloglossia is significant in the diagnosis and management of patients because many patients were wrongly diagnosed and treated for ankyloglossia. Hearing assessment should be done on children with suspected ankyloglossia because of possible articulation problems. Some of the patients that could not communicate properly were found to have hearing problems and not ankyloglossia.**

Keywords: Ankyloglossia, Frenulum, Speech disorder, Tongue-tie, Frenulotomy

INTRODUCTION

Ankyloglossia, or tongue-tie is defined as a clinical disorder where the tip of the tongue cannot be protruded beyond the lower incisor teeth because of a short lingual frenulum.[1] Lingual frenulum is an anatomical fibromucosal fold which connects the undersurface of the tongue to the floor of the mouth.[2] In this condition, the tongue is tethered or fused to the floor of the mouth. This occurs because the lingual frenulum is short and thickened, which restricts the movement of the affected tongue. The free mobility

of the tongue in its full range of mobility is impaired in such patients. Ankyloglossia occurs worldwide with varied prevalence and associated gender preponderance. It was reported that the risk of congenital malformation of the digestive system was significantly higher in males compared to females and the most common anomaly was ankyloglossia.[3] Documented prevalence of ankyloglossia from the literature varies between 0.1 and 10.7% worldwide.[4,5] The variability in incidence reported from literature is due to factors such as ages of the studied population, instrument used, and geographical location or degree of specialisation of the study centre.[6,7]

Ankyloglossia is a congenital anomaly.[8,9] The exact cause of ankyloglossia remains unclear, although it has been linked to certain syndromes such as pediatric obstructive sleep apnea syndrome.[10] It varies in degree, from mild ankyloglossia (some mobility) to complete ankyloglossia (tethered). There are different classifications of degrees of ankyloglossia. Kotlow classification of Ankyloglossia is based on measurements of the length of the free tongue (length of tongue from the insertion of the lingual frenulum into the base of the tongue to the tip of tongue). Normal: >16 mm, Mild (class 1): 12 to 16 mm, Moderate (class 2): 8 to 11 mm, Severe (class 3): 3 to 7 mm, and Complete (class 4): <3 mm.[11]

Clinically, ankyloglossia may be asymptomatic or symptomatic, depending on the degree of the disorder.[12-15] Symptomatic ankyloglossia usually presents with challenges in breastfeeding, such as poor sucking, latching problems, and problems with coordination of swallowing. There is an associated speech disorder (poor crying, articulation problems). In ankyloglossia, tongue mobility disorders lead to poor licking and kissing, and their associated complications. Treatment for symptomatic ankyloglossia typically involves surgical intervention, with the specific procedure determined by the severity of the condition. Frenulotomy is preferred in mild cases of ankyloglossia.[16-18] Frenulectomy is performed for moderate to severe ankyloglossia and frenuloplasty is carried out in cases of complete ankyloglossia.

There is a paucity of documented literature on ankyloglossia, or tongue-tie, in Nigeria. This study aimed to determine the epidemiology, clinical presentations, and management of children and adolescents with ankyloglossia in otorhinolaryngological practice at a tertiary institution of a sub-Saharan country.

MATERIALS AND METHODS

This was a cross-sectional hospital-based study of all the children and adolescent patients who presented with complaints of tongue tie in the Ear, Nose and Throat department of Ekiti State University Teaching Hospital, Ado-Ekiti, Nigeria. The study was carried out over a period of 10 years (January 2014 to December 2023). Informed consent was obtained from patients' parents and guardians, and the patients also gave their assent. Confidentiality of the obtained data was assured.

Structured questionnaires were administered to obtain data from patients and their parents by a research assistant and resident doctors. Detailed history on tongue-tie, feeding, speech, including ear, nose and throat symptoms (poor sucking, swallowing, poor licking), were obtained and documented in the profoma for each patient. Detailed oral, throat, ear, nose and neck examinations were carried out by the consultant otorhinolaryngologist and resident doctors in otorhinolaryngology clinic, and the findings were documented. Hearing assessment was done by the audiologist. The criteria for diagnosis included a short, thick, or stiff lingual frenulum, attached close to the tongue's tip, limited tongue movement, poor latch, frequent loss of latch, lengthened feeding times, and poor weight gain. These were confirmed on examination. Ankyloglossia was confirmed if tongue protrusion was not beyond the Vermillion border, and Pure-tone audiometry was done for adults. Patients with hearing impairment had audiological assessment done and treated accordingly. Data obtained were collated and analysed using SPSS version 23. Data were expressed using frequency tables, percentages, bar and pie charts.

Ethical Approval

Ethical clearance was sought and obtained before the research work was commenced from the ethical review committee of Ekiti State University Teaching Hospital, Ado Ekiti, Nigeria.

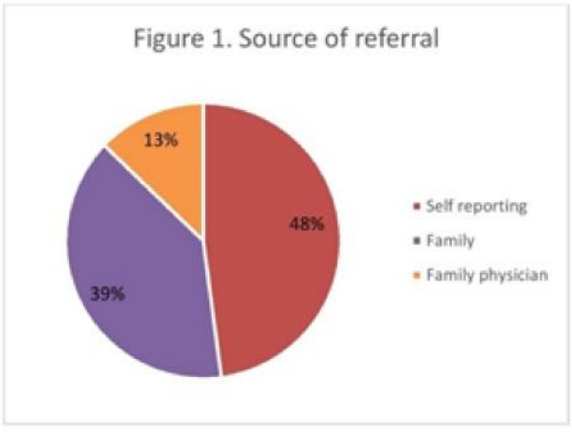
RESULTS

The age range of patients recruited was from less than one year to nineteen years. The total number of patients seen over the study period was 18,431 out of which 94 (0.51%) presented with complaints of tongue-tie and 43 (0.23%) were diagnosed with ankyloglossia. The prevalence of ankyloglossia in this study was 0.23%. The majority of the complaints of tongue tie were observed in children aged 1-5 years(48.9%, n=46) This was followed by infants who were 25 (26.6%). This is shown in table 1. In this study, there were 69 (73.4%) males and 25 (26.6%) females. The male-to-

Table 1: Sociodemographic features

Variables	Number	Percentage
Gender		
Male	69	73.4
Female	25	26.6
Religion		
Christian	87	92.6
Muslim	7	7.4
Domicile		
Rural	38	40.4
Urban	56	59.6
Parental education level		
No formal	5	5.3
Primary	31	33.0
Secondary	46	48.9
Graduate	12	12.8
Parental occupation		
Trading	19	20.2
Artisan	36	38.3
Civil servant	39	41.5
Family history		
Present	3	3.2
Absent	91	96.8
Age (year)		
≤1	25	26.6
1-5	46	48.9
6-10	15	16.0
11-15	7	7.4
≥16	1	1.1
Total	94	100.0

female ratio was 2.8:1.0. The commonest parental education was secondary in 46 (48.9%)cases followed by primary school in 31 (33.0%). The majority of the parents were civil servants in 39 (41.5%) cases followed by artisans in 36 (38.3%) cases respectively. The family history of tongue-tie was recorded in 3 (3.2%) cases. The commonest sources of referral were the parents in 45 (47.9%) cases. This was followed by the families and family physicians in 37 (39.3%) and 12 (12.8%) cases respectively. This is illustrated in figure 1.



The commonest presenting feature were problems with verbal communication by the patients in 51 (54.3%) cases. This was followed by poor articulation, poor licking and poor sucking in 43 (45.7%), 26 (27.7%) and 23 (24.5%) patients respectively. A patient presented with multiple symptoms in this study, and this is further elaborated in table 2.

Table 2: Clinical features

Diagnosis	Number	Percentage (%)
Delayed developmental	22	23.4
milestones	18	19.2
Articulation disorder	11	11.7
Hearing impairment	43	45.7
Ankyloglossia		
Degree of ankyloglossia		
Mild (class 1)	26	60.5
Moderate (class 2)	10	23.3
Severe (class 3)	6	14.0
Complete (class 4)	1	2.2

Note: A patient presented with multiple symptoms in this study

No prior intervention was instituted in 8.5% of the patients. Family physicians have treated (by using sterilised scissors and gloved fingers to dissect the frenulum) majority (43.6%) of the patients prior to presentation in our facility. Traditional treatment by the use of herbs was offered to 20 (21.3%) of the patients. This is shown in figure 2. Normal tongue was recorded in 51 (54.3%) patients that presented with complaints of tongue-tie.

Of the 51 patients presenting with complaints of tongue-tie who were not diagnosed with ankyloglossia, 22 (23.4%) were diagnosed of delayed developmental milestones, 18 (19.1%) had articulation disorders and

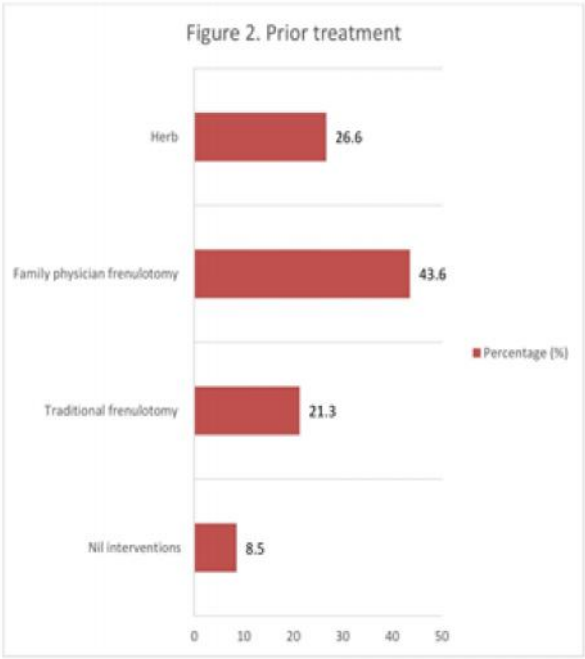
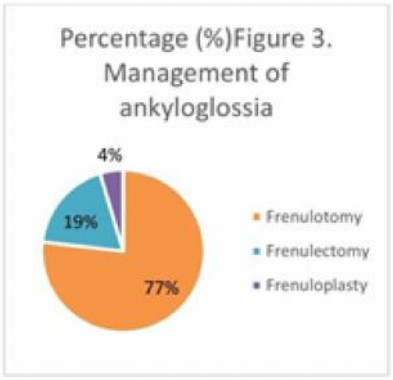


Table 3: Diagnoses and degree of ankyloglossia

Diagnosis	Number	Percentage (%)
Delayed developmental milestones	22	23.4
Articulation disorder	18	19.2
Hearing impairment	11	11.7
Ankyloglossia	43	45.7
Degree of ankyloglossia		
Mild (class 1)	26	60.5
Moderate (class 2)	10	23.3
Severe (class 3)	6	14.0
Complete (class 4)	1	2.2

11 (11.7%) were diagnosed with hearing impairment as shown in (table 3). The commonest degree of ankyloglossia was mild (class 1) ankyloglossia in 26 (27.7%)patients. Ten patients (10.6%) had moderate (class 2) ankyloglossia and severe (class 3) ankyloglossia was seen in 6 (6.4%) patients as shown in table 3.



All the patients diagnosed with ankyloglossia had surgery done. The commonest surgery was frenulotomy in 33 (35.1%) cases followed by frenulectomy in 8 (8.5%) cases and frenuloplasty in 2 patients. For further details see figure 3. Patients with delayed developmental milestones and poor articulation were treated by speech pathologist and therapist.

DISCUSSION

In this study, it was noted that ankyloglossia was an uncommon presentation in otorhinolaryngological practice in Nigeria. Although the prevalence of ankyloglossia among the studied children and adolescents in this study was low, higher prevalence was recorded in a neonatal study.[19] Detailed history and examination are required to establish a diagnosis, as parents often attribute other conditions leading to poor speech as being related to tongue-tie. An accurate diagnosis is essential before education of the parents and treatment of ankyloglossia is offered. Low prevalence in this study may be due to the inclusion of older children and adolescents. From this study, all the studied age groups were affected with peak prevalence during the preschool age group. This finding was also recorded in a previous study.[20] Furthermore, there is a higher preponderance in males than females as reported in other research work, which concurred with another study.[20,21] Urban dwellers were more common than rural dweller patients as this may be due to their proximity to the center. There was no family history of ankyloglossia in majority of the patients in this study while contrary findings were recorded in a previous study.[22]

The majority of the patients with suspected ankyloglossia were brought to the hospital based on independent observation by their parents, this is in keeping findings from another study that studied the persons identifying the problems associated with tongue-tie.[23] Some of the patients were directed by their family or referred by their family physicians for further experts review and management.

In this study, majority of the parents and families were concerned with the inability of their children to speak or speak properly. Some families associated this speech disability with tongue immobility and wanted the tongue to be freed by experts as in another study.[24] The commonest presenting features were problems of verbal communication by patients or poor articulation. In addition to this, parents also related tongue immobility to poor breast milk sucking and poor licking. Also associated with ankyloglossia in this study were poor crying patterns, poor swallowing and breast nipple pain with cracking among the mothers.

Based on the challenges faced by patients with their children, different interventions were sought, which included home remedies, traditional treatment, hospital-based treatment and finally specialist care as was obtained from a previous study.[25] Only a few parents had no prior interventions before specialists care. In this study, after a detailed history was taken and thorough clinical examinations performed, patients with normal tongue mobility were identified among these patients presenting with suspected ankyloglossia

in this study and concurred with other studies.[26-28] However ankyloglossia of various degrees was noted in other patients. The commonest degree of ankyloglossia was mild (class 1) ankyloglossia in this study and the same was recorded in another paper.[29] The less common ankyloglossia were moderate (class 2), severe (class 3) and complete (class 4).

Hearing impairment in severe cases leads to nonverbal speech development and has a serious effect on speech development.[30] All patients were managed based on their clinical diagnosis with remarkable parental satisfaction. Patients diagnosed with ankyloglossia had surgery done based on the degree of ankyloglossia. The surgeries included frenulotomy, frenulectomy and frenuloplasty with good parental satisfaction.

Strengths and limitations

This study involved more than 18,000 patients, and it was carried out over a ten-year period, thus providing a large study population for the evaluation of this condition. The study has opened more insights into ankyloglossia. However, it is a cross-sectional study in which causal relationships could not be determined. Experimental studies could be conducted to determine causal relationships between ankyloglossia and articulation problems.

CONCLUSION

Ankyloglossia is a congenital anomaly in children and adolescents. Many patients with speech problems can be wrongly attributed to having ankyloglossia. Assessments by otorhinolaryngologists, including hearing assessments are advised for children with suspected ankyloglossia, especially when they also have difficulties with verbal communication and development delays because of possible articulation problems. Treatment of ankyloglossia is determined by the severity of ankyloglossia. In this study, genetic study was not carried out. Genetic study is advised in future studies on ankyloglossia.

Conflict of Interest

There is no conflict of interest.

Authors' contributions

Conception and Design, WAA, IAA, OF, MB, OWJ, MO, AKA,OS; Acquisition of Data, WAA, IAA, OF, MB, OWJ, MO, AKA,OS ; Analysis and Interpretation of data, WAA, IAA, OF, MB, OWJ, MO, AKA, OS; Drafting the article, WAA, IAA, OF, MB, OWJ, MO, AKA, OS; Revision, WAA, IAA, OF, MB, OWJ, MO, AKA,OS; Final approval, WAA, IAA, OF, MB, OWJ, MO, AKA,OS . All the authors agreed to be accountable for all aspects of the work.

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