

Ankyloglossia: Analysis of 56 Cases Seen in Port Harcourt, Rivers State, Nigeria

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Abstract

Introduction: Ankyloglossia is a Greek word meaning “agkilos” –curved, and “glossa” –tongue, also known as tongue-tie. It refers to the presence of an unusually short lingual frenulum which restricts the tongue in achieving its full range of mobility. It causes poor co-ordination of swallowing, with its attendant feeding problems, and subsequently may lead to failure to thrive in some infants. Kotlow in 1999, stated a classification of Ankyloglossia, based on the distance between the insertion of the lingual frenulum and the tip of the tongue. Some patients have persistent associated problems, which only improve with surgical treatment of the tongue-tie. Surgical treatment of ankyloglossia can either be frenulotomy, frenulectomy or frenuloplasty, depending on the degree of the tongue-tie. This aim of this study is to chronicle and analyse the 56 cases of Ankyloglossia seen in Port Harcourt, Rivers state, Nigeria. To highlight peculiar characteristics or patterns identified in these cases.

Patients and method: This study is a descriptive analysis of 56 cases of Ankyloglossia seen in Port Harcourt, Rivers state, Nigeria from July 2014 to June 2019. These cases were pooled together from medical records of the Ear, Nose and Throat theatre, Hospital Main theatre, and ENT clinic of the University of port Harcourt Teaching Hospital, Rivers state, Nigeria. The degree of tongue-tie was categorized using Kotlow’s classification of Ankyloglossia. Results are represented in simple tables and figures.

Results and conclusion: The study is a retrospective study of 56 cases of Ankyloglossia seen in the University of Port Harcourt Teaching Hospital (UPTH) from July 2014 to June 2019. There were 38 males and 18 females and an age range of 1 month to 10 years. Associated problems include poor cry and poor speech.

Keywords: Ankyloglossia; Tongue-tie; Frenulum; Frenulotomy; Port harcourt

Introduction

Ankyloglossia is a Greek word meaning “agkilos” –curved, and “glossa” –tongue, it is also known as tongue tie [1]. It refers to the presence of a short lingual frenulum which restricts the tongue in achieving its full range of mobility [2]. Lingual frenulum is a fibro-mucosal fold that connects the ventral surface of the tongue to mucosa of floor of the mouth [1]. During embryology of the primitive foregut, there is an apoptotic process that occurs in the frenulum, with the aim to separate the embryologic structures that form the tongue and the floor of the mouth [2]. However, a failure in this planned cell death can result in an unusually short frenulum. The extent of the failure of this programmed cell death, also determines the severity of the ankyloglossia [1]. The first use of the term “ankyloglossia” was in the 1960s, when Wallace defined tongue-tie as “A condition in which the tip of the tongue cannot be protruded beyond the lower incisor teeth because of a short lingual frenulum” [3]. It varies in degree, from mild ankyloglossia where the lingual frenulum is composed of mucous membrane bands alone, to the complete tongue-tie or ankyloglossia where tongue is tethered to floor of mouth.

This restriction of tongue movement in ankyloglossia interferes with several functions, such as sucking, crying, swallowing, and speech [2]. This results in poor cry, poor breastfeeding of affected babies from inability to latch on to the breast, and maternal nipple pain during breastfeeding. It causes poor co-ordination of swallowing, with its attendant feeding problems, and subsequently may lead to failure to thrive in some infants [2]. In post lingual children, it can also cause poor articulation of speech, especially with the articulation of certain phonemes that involve lifting or protrusion of the tip of the tongue [4]. Ankyloglossia can be asymptomatic, may resolve spontaneously during growth, and some individuals may compensate adequately for their

decreased tongue mobility [1]. However, some patients have persistent associated problems, which only improve with surgical treatment of the tongue tie [4].

Kotlow in 1999, stated a classification of ankyloglossia that has four types based on the distance between the insertion of lingual frenulum and tip of the tongue [5]. He noted that the closer the insertion of the lingual frenulum to the tip of the tongue, the more the restriction of tongue mobility. Surgical treatment of ankyloglossia can either be frenulotomy, frenulectomy or frenuloplasty, depending on the degree of the tongue-tie. Frenulotomy is done in mild tongue-tie where the short frenulum is composed of only mucous membrane and is only incised to releases the tongue [6]. Frenulectomy is done in moderate to severe tongue-tie where the frenulum is thick, and part or the entire band has to be removed to release the tongue [6]. Frenuloplasty is the surgical repair carried out in cases of complete ankyloglossia where the tongue is plastered to the floor of the mouth, and there is need to refashion the under-surface of the tongue and the floor of the mouth following tongue detachment [6].

This aim of this study is to analyse the 56 cases of ankyloglossia seen in Port Harcourt, Rivers state, Nigeria. To highlight peculiar characteristics or patterns identified in these cases.

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Patients and Methods

This study is a descriptive analytical survey of 56 cases of ankyloglossia seen in Port Harcourt, Rivers state, Nigeria. These cases were pooled together from medical records of ear, nose and throat theatre, Main theatre, and ENT clinic of the University of Port Harcourt Teaching Hospital, Nigeria. The University of Port Harcourt Teaching hospital is a tertiary hospital located in the city of Port Harcourt, which is the capital of Rivers state. It receives patients referred from the primary and secondary health care facilities and private clinics within Rivers state. Data collected were age, sex, associated problems, degree of tongue tie, and type of surgical procedure done. Due approval was gotten from the hospital medical records department before medical records collection, and confidentiality was applied. The degree of tongue tie as reported on surgical operation notes tie was categorized using Kotlow’s classification of ankyloglossia, and all cases were done under general anaesthesia, The procedure of tongue tie release done, depended on the degree of ankyloglossia. Frenulotomy was done for mild cases of ankyloglossia with thin mucous membrane bands, while frenulectomy was done for the release of thick frenulum bands. Results are represented in simple tables and figures.

Results

This is a retrospective study of 56 cases of ankyloglossia seen in the University of Port Harcourt Teaching Hospital (UPTH) from July 2004 to June 2019. The gender distribution was 38 males and 18 females and an age range of 1 month to 10 years. The commonest associated problem among the pre-lingual subjects was poor cry and breastfeeding problems, while the commonest in the post-lingual population was poor speech. Three classes of ankyloglossia were seen in this study, types F1, F2 and F3. Only frenulotomy and frenulectomy was done in this study. Details of the results are given in the tables and figures below.

Figure 1 shows the gender distribution of the study subjects. There were 38 (68%) males and 18 (32%) females in the study population, with a male:female ratio of 2.2:1.

Table 1 and Table 2 show the age distribution of the patients with ankyloglossia. The highest presentation was seen in children below 1 year of age with 39 patients, among which 27(62.9%) were less than 6 months old. There were 4 (7.1%) patients who were older than 5 years, with the oldest being 10 years old.

Signs and symptoms of ankyloglossia in this study include poor cry, feeding problems like inability to latch on the breast, poor feeding due to inability to sustain sucking, failure to thrive, poor speech. Breastfeeding problems in the mother include painful nipple during breastfeeding.

Figure 2 shows the type of lingual frenulum insertion seen in the subjects. The commonest type of insertion was the F2 insertion, which was in 30 patients. The F1 insertion was seen in 24 patients while F3 insertion was seen in 2 patients.

Age range	Number of subjects	Percentage
<1 year	39	69.70%
1-5 years	13	23.20%
6-10 years	4	7.10%
Total	56	100%

Table 1: Age of subjects with ankyloglossia.

Age of children less than 1 year	Number of subjects	Percentage
0-5 months	27	69.20%
6-11 months	12	30.80%
Total	39	100%

Table 2: Age of children with ankyloglossia (less than 1 year).

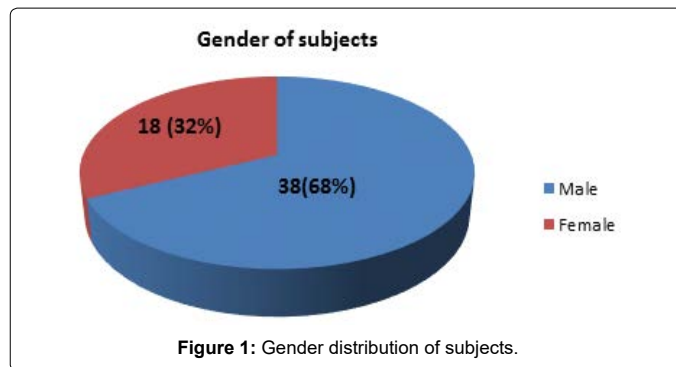


Figure 1: Gender distribution of subjects.

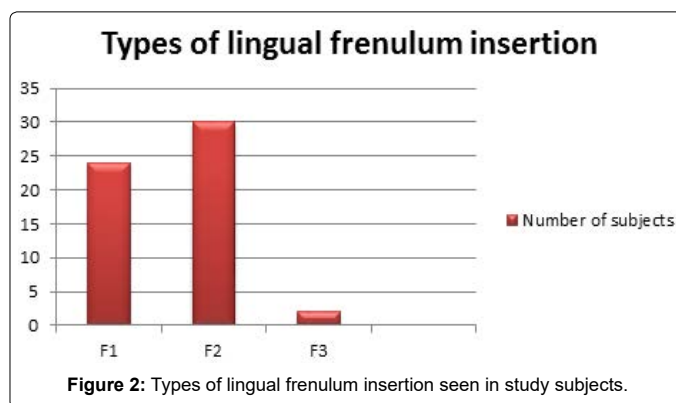


Figure 2: Types of lingual frenulum insertion seen in study subjects.

Discussion

In this study, it was noted that ankyloglossia is commoner in males than females in the ratio of 2.2:1. As much as there may not be conclusive explanations for this, it may be attributed to its X-linked genetic characteristic [7], which makes it more expressible in males. Ankyloglossia has been found to be a relatively common pathology, which has been widely reported [1,8]. As much as its cause is said to be unknown, it has been reported by Ashtankar et al. to be due to a sporadic mutation in the transcription factor TBX22, which may lead to an X-linked ankyloglossia, with or without associated cleft lip, cleft palate or hypodontia [7]. This is buttressed by the case report by Morowati et al. in 2010 of familial ankyloglossia in five individuals from five consecutive generations of a family lineage [9].

However, this needs to be validated in genetic based studies in patients with ankyloglossia. This male predominance was similarly noted in the study done by Osifo et al. in Benin, Nigeria where the male to female ratio was 1.7:1 [10] and Osama in Jordan, who found a male female ratio of 2.7:1 [11].

Problems associated with ankyloglossia in this study include poor cry, feeding problems like inability to latch on the breast, poor feeding due to inability to sustain sucking, failure to thrive and fever. Breastfeeding problems in the mother include painful nipple during breastfeeding, and poor speech was seen in children that were post lingual. Our results show that majority of the cases presented at age

less than one year and this is in keeping with the study by Osifo et al., with a mean age of 5 months of age [10]. This may be due to the early manifestation of poor cry, breastfeeding difficulty, with associated difficulty in latching on to the breast, failure of prolonged sucking, with maternal nipple pain and mastitis which commonly characterizes ankyloglossia in this neonatal and infantile age group [2]. This factor most likely prompts the mothers of these babies to present to hospital for treatment of this pathology. However, contrary to this, the Canadian paediatric society in their position paper [12] states that symptomatic tongue tie is not common and must be of very severe degree before it can cause breastfeeding difficulties [12].

This study noted that the point of insertion did not correlate with the thickness of the short frenulum, in the sense that thick bands were seen for both F1 and F2 insertions. The lingual frenulum usually composes of only mucous membrane, but occasionally, some fibers of the genioglossus muscle migrates into the frenulum during embryogenesis, making the band thicker than usual [7]. However, in all cases seen in patients aged greater than 5 years, the frenulum were thicker than just mucosal folds. Vivek et al. [13], reported a case of ankyloglossia in an 11 year old male and Antony et al. [6], reported two cases in males aged 16 and 21 years old, both studies noted similar findings of thick frenulum, and did frenulectomy in all cases [6,13]. Thus the spontaneous resolution of ankyloglossia with growth of the child can be said to be more unlikely in cases of thick band frenulum. However, the claims of spontaneous resolution of tongue-tie in neonates in the course of their growth can be due to thin frenulum and can only be substantiated by longitudinal studies with serial assessment of the frenulum.

Frenulotomy and frenulectomy was done in this study, and it was noted that the type of surgery done, was not necessarily dependent on the type of insertion, but rather on the thickness of the frenulum. In this study, excision of the frenulum with simple closure of defect was done as Frenulectomy in cases with thick band and this was seen in all cases older than 5 years. This thick frenulum seems to be more associated with speech defect [14], as seen in our study. This is also similar to the findings by Anand et al. [13] and Antony et al. [6]. We noted good outcome with all cases, no record of any complication in our patients, and there was no mortality. Following frenulectomy, the older patients were referred for speech therapy.

Conclusion

The study found that ankyloglossia is commoner in males and the point of frenulum insertion does not correlate to thickness of the band. Furthermore, the thick band frenulum is commoner in older children and more likely associated with speech defect. The type of surgery done depends on the thickness of the band and not necessarily on the frenulum insertion.

Conflict of Interest

There is no conflict of interest.

References

1. Dezio M, Piras A, Gallottini L, Denotti G (2015) Tongue-tie, from embryology to treatment: a literature review. *J Pediatr Neonat Individual Med* 4: e040101.
2. Kupietzky A, Botzer E (2005) Ankyloglossia in infants and young children: clinical suggestions for diagnosis and management. *Paediatr Dent* 27: 40-46.
3. Wallace AF (1963) Tongue tie. *Lancet* 2: 377-378.
4. Baker AR, Carr MM (2015) Surgical treatment of ankyloglossia. *Oper Tech Otolaryngol Head Neck Surg* 26: 28-32.
5. Kotlow LA (1999) Ankyloglossia (tongue-tie): A diagnostic and treatment quandary. *Quintessence Int* 30: 259-262.
6. Antony VV, Khan R (2013) Management of ankyloglossia – Case report. *IOSR J Dent Med Sci* 6: 31-33.
7. Ashtankar M, Baburay MD, Tambe L, Kode S (2018) Ankyloglossia-A review and Case report. *European J Pharmaceut Med Res* 5: 426-430.
8. Opara PI, Eke GK (2013) Tongue-tie in Children; Mothers perception, attitude, and practice in Port Harcourt, Nigeria. *Global J Med Res* 13: 19-23.
9. Morowati S, Yasini M, Ranjbar R, Peivandi AA, Ghadami M (2010) Familial ankyloglossia (tongue-tie): a case report. *Acta Med Iran* 48: 123-124.
10. Osifo OD, Osaigbovo EO (2008) Ankyloglossia in Benin City. *Port Harcourt Med J* 3: 49-54.
11. Osama MA (2016) Tongue tie (Ankyloglossia) *Global J Otolaryngol* 1: 1-4.
12. Canadian paediatric society Position paper (CP 2002-02) (2002) Ankyloglossia and breastfeeding. *Paediatr Child Health* 7: 269-270.
13. Anand K, Vivek S, Asish M (2014) Ankyloglossia or tongue tie –A case report. *IOSR J Dent Med Sci* 13: 52-54.
14. Chaubal TV, Dixit MB (2011) Ankyloglossia and its management. *J Indian Soc Periodontol* 15: 270-272.