Aetiology, Diagnosis and Treatment of Ankyloglossia

SUMMARY

This review paper occupies with the frequency, etiology, diagnosis, treatment and the possible complications of Ankyloglossia (AG). AG is a congenital anomaly and its range varies from 0,1% to 4,8%. There are several methods for the diagnosis of AG. The most popular method is the “Hazelbaker”, which assesses seven different tongue movements and five appearance characteristics. As far as the management of AG is concerned, there are two options, the “wait-and-see” and the invasive procedure. The operator can choose between the frenotomy and the frenectomy. The difference is that in frenectomy the clinician removes the whole frenulum. Few complications have been mentioned, such as ulcers, pain, bleeding and noticeable scar, which were brought on to a second operation.

Key words: Ankyloglossia, Breast Feeding, Frenectomy

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REVIEW PAPER (RP)

Introduction

Ankyloglossia (AG), also known as tongue-tie, is a congenital oral disorder that may decrease mobility of the tongue tip. The cause of AG is an unusually short, thick lingual frenulum, which is the membrane connecting the underside of the tongue to the floor of the mouth. AG is a controversial issue that concerns many researchers. Some of them declare that it is rarely symptomatic, whereas others observe a variety of complications caused by it. It has been found that AG affects breastfeeding, oral hygiene, speech, as well as the development of swallowing and occlusion. The above indicate the severity of tongue-tie, affecting both families and dentists. Treatment of AG encompasses a surgical approach, including frenotomy and frenectomy and a conservative one, such as “wait-and-see” method. The present literature review aims to cite the etiology of AG, introduce the way it can be diagnosed and discuss the possible outcomes as well as the management of this condition.

Aetiology

AG is a congenital anomaly and its range varies from 0,1% to 4,8%. Both genetic and environmental factors are involved in its etiology1. Molecular analyses show that point mutations on the TBX22 gene can cause cleft palate together with AG1,2,3. This gene is located on Xq21 and generally eight point mutations have been identified as the cause of CPX3. A hereditary condition which is reported to provoke AG is epidermolysis bullosa (EB) and specifically the recessive dystrophic subtype. EB is characterized by soft tissue blistering, which results in tissue separation and scarring. As an outcome, adhesions are developed which result in reduced tongue mobility (tongue tie)4. Correlated AG appears as an isolated anomaly, but it is also linked to several craniofacial abnormalities such as the X-linked cleft palate that is mentioned above. Other syndromes are: Opitz, orofacialdigital, Backwith-Wiedemann, Simpson-Golabi-Behmel, Van der Woude and Pierre-Robin1,2,5.
Diagnosis

AG is a developmental abnormality, which is characterized by a short, thick lingual frenum either attached distally to the floor of the mouth or onto (or close to) the alveolar ridge or it extends from the tip of the tongue. It is a quite common condition in neonates and it may even be asymptomatic or resolve spontaneously. Other infants and toddlers face difficulties in breastfeeding, swallowing and speech, while some of them manage to compensate for their condition. During the clinical assessment the tongue’s functional and appearance characteristics should be evaluated. A normal range of tongue motor function is shown in Table 1. The patient presented with AG, because of the restricting lingual frenulum, is not usually able to protrude the tip of the tongue beyond the lower incisors (or the lower lip), moves with strain the tongue from side to side and also faces difficulty in lifting the tip to the upper alveolar ridge. In addition, the tongue appears heart-shaped or notched upon protrusion because of the force applied by the sublingual membrane. It is important to note that a short sublingual frenum is not necessarily fibrotic or inelastic. In that case, especially if the mouth floor maintains its elasticity, it may not affect the tongue’s normal mobility.

In order to clinically assess and treat AG, several investigators have developed a few criteria. Hazelbaker assessed seven different tongue movements and five appearance characteristics and according to the results he suggested the proper treatment (Table 2). Kotlow measured the “free tongue” length as the distance between the point the frenum is attached to the tongue and the tip of the tongue. According to the length measured, he classified AG in four groups: mild, moderate, severe and complete and suggested proper treatment for each of them (Table 3). Fletcher and Meldrum determined the relative “free tongue” length which is correlated to speech impairment. Williams and Waldron introduced a different method which emphasizes in assessing the tongue functions. According to this method, the patient is asked to place the tip of the tongue on the maxillary alveolar ridge and while maintaining the contact, opens the mouth as wide as possible. At this position, the distance between upper and lower incisors is measured and the tongue function is evaluated. In addition, the patient is asked to articulate specific sounds in order to evaluate possible speech impairment. Messner and Lalakea evaluate the tongue function in correlation with speech disorders. They rely on the Williams and Waldron technique and also measure the tongue length from the tip of the tongue to the lower incisors when it is protruded forward.

<table>
<thead>
<tr>
<th>Table 1. Normal range of tongue motion</th>
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<tbody>
<tr>
<td>Tip of the tongue should protrude outside the mouth without clefting</td>
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<td>Tip of the tongue should be able to sweep the lips easily – without straining</td>
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<td>When the tongue is retracted, it should not blanch the tissue lingual to the anterior teeth</td>
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<td>The lingual frenum should not create a diastema between mandibular central incisors</td>
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<td>The lingual frenum should not prevent the infant from attaching to the maternal nipple while nursing</td>
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<td>Children should not exhibit speech difficulties</td>
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<tr>
<th>Table 2. Hazelbaker’s criteria regarding appearance and function of the tongue in cases with AG</th>
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<tr>
<td>Appearance</td>
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<tr>
<td>When Lifted: round/square, slight cleft in tip apparent, heart/V-shaped</td>
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<tr>
<td>Elasticity of frenulum (lift the tongue and palpate the frenum)</td>
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<tr>
<td>Length of lingual frenulum when tongue lifted</td>
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<tr>
<td>Attachment of lingual frenum to the tongue</td>
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<tr>
<td>Attachment of lingual frenum to inferior alveolar ridge</td>
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<tr>
<td>Function</td>
</tr>
<tr>
<td>Lateralization</td>
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<tr>
<td>Lift Of Tongue</td>
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<tr>
<td>Extension of tongue</td>
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<tr>
<td>Spread of anterior tongue</td>
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<tr>
<td>Cupping</td>
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<td>Peristalsis</td>
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<td>Snap Back</td>
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<th>Table 3. Kotlow’s classification of AG according to the range of tongue movement</th>
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<td>Kotlow’s classification</td>
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<tr>
<td>Clinically accepted</td>
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<tr>
<td>Class I mild AG</td>
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<td>Class II moderate AG</td>
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<td>Class III severe AG</td>
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<td>Class IV complete AG</td>
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Complications

A lot of studies have been researching the correlation of AG with breastfeeding and ingestion. Many investigators declare that AG in infants is the cause of breastfeeding problems. This condition applies to both the mother and the neonate. On the one hand, the infant fails to nurse effectively and on the other hand, the mother suffers from nipple trauma. The tongue’s limited mobility hinders the infant from obtaining an adequate seal when nursing and usually, they use their gum pads to keep the nipple in the mouth. As a result, the milk transfer is inadequate, the baby gets frustrated and dissatisfied,
it may chew the breast, the feedings are frequent, prolonged and the weight gain is below average while in some cases the neonate fails to thrive. As far as the mother is concerned, common findings in researches are traumatized nipples, severe prolonged breast pain while feeding, incomplete drainage of the milk and consequently breast infections and clogged ducts\cite{1,2,6,8,12,13}. Early weaning and switching to bottle feeding have also been noticed\cite{11}. On the contrary, a lot of researches declare that the effect of AG in breastfeeding is not significant. A study conducted by Messner et al.\cite{11} notices that for at least two months postpartum, breastfeeding in infants with AG was not affected in a statistically important way. There is also discordance in how frenotomy affects the quality of breastfeeding. In several studies, it has been observed a remarkable enhancement in breastfeeding effectiveness after the surgical procedure\cite{10,12-14}. Research showed that infants who had undergone frenotomy, improved their weight gain, maternal nipple pain was reduced and the breastfeeding was prolonged\cite{10,12-15}.

There is a big controversy whether AG can affect the child's speech ability. Although not correlated with speech delay, it may interfere with articulation and intelligibility. Pronunciation of certain sounds such as "t, d, l, th, s" may be impaired due to restriction of the tongue tip, which is unable to reach the palate or the palatal gums of the upper incisors. In case a patient articulates all those sounds properly, the surgical approach is not recommended\cite{2,10,16,17}.

Among children with AG, maintaining infantile swallowing pattern is common. It is known that the swallowing mechanism in adults differs from infants. During infancy, deglutition takes place with the jaws open and the tongue thrusting between them. This pattern changes at the age of 2.5 years with the completion of primary teeth eruption. To be specific, the mouth is closed during swallowing and the tongue elevates on the palatal surface of upper incisors. However, in children with AG, tongue fails to elevate in order to create a seal and thus continues to thrust forward\cite{18}. The ability to elevate the tongue is crucial for the proper development of the adult swallowing mechanism.

Other problems mentioned in the bibliography are orthodontic anomalies, inadequate retention of orthodontic mobile devices, mechanical issues related to oral hygiene and social difficulties\cite{1,2,16,17}.

Cases are reported where children with AG develop malocclusion and specifically open bite. In a study conducted by Vaz and Bai\cite{19}, it became perceptible that children with AG may present maxillary protrusion and open bite. Normally while deglutition, the tongue elevates on the palate to create a seal, thus supporting it in order to obtain its normal shape. In children with AG though, the short frenum restricts the upward movement of the tongue, which thrusts forward during swallowing, pressing the upper anterior up and out\cite{8,10,19}. What is more, in cases of severe AG, the tongue is positioned lower in the mouth than normal, which may result in the disproportionate growth of the two jaws (growth of the maxilla is not stimulated as there is no or little contact with the tongue) and cause a Class III malocclusion\cite{17}. It is important to note that as opposed to what one may think, greater teeth irregularities were observed in milder cases of AG. The reason is that frenulums with better mobility appear in grades I or II of AG and in this way the tongue is able to apply greater force on the incisors, contrary to types III or IV of AG where the tongue function is severely limited\cite{19}.

Limited tongue movement impedes the self-cleaning of anterior teeth and lips, resulting in poor oral hygiene, as it is difficult to sweep food debris from tooth surfaces. The presence of a short frenum may lead in gingival recession, caused by the force applied on the gums and the degradation of oral health. Receded gums have been located especially in lower incisors. It has also been observed that frenotomy caused immediate improvement\cite{1}.

As far as the social outcomes are concerned, children with AG may be mocked by their peers because they experience difficulties in licking ice-cream or their lips, drooling, playing a wind musical instrument or find trouble in kissing. These may affect their self-esteem and be the cause of psychological issue\cite{2,11,16}. In addition, Chinnadurai et al. focused on the positive outcomes of treating AG, other than enhancing feeding impairment. The above include speech improvement and social issues as the gain of self-esteem\cite{16}.

**Management and treatment of AG**

There are two aspects for the management of AG in all existing studies. One option is the method of "wait-and-see"\cite{11}, in this case the clinician watches the patient in regular periods. If there are difficulties with breastfeeding, counseling and education could help the mother. However, Marmet et al. found out that it didn’t improve the breastfeeding after 12 weeks in 10 of 13 infants\cite{6}. Speech problems can be handled with the help of a speech therapist with beneficial results. Patients with AG have difficulty in articulation of ‘t’, ‘d’, ‘th’, ‘l’, ‘s’. Moreover, dentists should give special instructions for oral hygiene\cite{4}, in order to avoid minor injuries of the frenum and the plaque formation.

On the other side, there are several studies which recommend the invasive procedure. We cannot compare the effects of surgical and no-surgical treatment on breastfeeding due to the fact there are not adequate surveys for non-invasive management. However, it is mentioned that frenotomy improves breastfeeding the same day or within five days\cite{20}. Not only has surgical division of the tongue positive results on breastfeeding,
articular and hygiene, but also on infant's weight due
to the sufficient attachment to the nipple and the adequate
supply of milk.

There are two kinds of surgical approaches, frenotomy and frenectomy. The first procedure releases
the frenulum, while the frenectomy removes the frenum
entirely. As far as the anesthesia is concerned, some
recommend general and others local. It has dominated
that local anaesthesia is preferable for adults and older
children and general for young children. There are
several studies which support the absence of general
or local anaesthesia during the surgical procedure on
neonates. The anaesthetic which is preferred is articaine
4% with epinephrine 1:100000. The clinician needs to
have an assistant to hold the tongue upward, usually with
a sterile gauze. The incision is horizontal- rhomboidal and
it’s located next to the tongue, in distance of the
floor of the mouth, so as to avoid the incision of any
vascular tissue. Argiris et al. mentioned 24 patients with
excessive blood loss.

Nowadays, laser is used a lot for this procedure, because of the safe and easy use, the avoidance of
any bleeding and sutures. There are different types of
laser for this operation, CO₂ laser, diode laser, erbium:
yttrium-aluminium-garnet laser (Er:YAG laser) and
erbium, chromium:yttrium-scandium-gallium-garnet
laser (Er,Cr:YSGG laser). If diode laser is chosen for this
approach, firms recommend 1.5-3 W, CV (Continuous
Wave), 810/940/980 nm with initiated tip, always in
contact with the tissue.

Sutures are usually not necessary as well as antibiotics. Lampa et al. presented a case report, using
Er,Cr:YSGG laser (wavelength 2780 nm, short pulse “H”
mode with 600 µm sapphire tip at 1.5 W power, 13% air
and 9% water in contact mode). The advantage of using
Er laser is the sterilization of the wound. Nicoloso et al.
cite one more benefit of diode laser on frenotomy and this
is the coagulation that is succeeded.

Apart from conventional operation and laser, there
is the option of electrosurgery although it is more painful
than the previous techniques. When the operator decides
to suture the wound, usually chooses 4/0 polyglycolic
acid. Patients can handle with pain, receiving analgesic
drugs, like paracetamol (acetaminophen). When the
frenulum is thick and vascular, frenectomy (z-plasty)
is preferred. The reason is to avoid bleeding and
reattachment due to the scar. In this case, the operator
has to suture the wound. Steehler et al. mention 8 patients
who underwent additional procedure due to the scar
or incomplete clipping. Argiris et al. mentioned 3 of 46
patients who required repetitive surgery and 24 of 46
patients with noticeable blood loss.

Some surveys detail other complications, like ulcers
over the injury or pain. Ferrés- Amat et al. used a
specific protocol of orofacial rehabilitation. The patient is
asked to lift the tongue towards the palate with the mouth
open. This exercise begins 1 week before the surgery and
it continues 45 days postoperative, concluding that 96 %
of patients had valuable correction. The basic inquiry is
which the appropriate age to interfere is. Lalakea et al. support the procedure after
birth in order to avoid impediment. Ketty et al. and
Paradise recommend we do not anything till the age of
4 years old, in order to assess the complications of AG.
The survey of Glynn et al. in 48 children demonstrated
that the median age (months) for frenotomy is 3.5 in
case of breastfeeding difficulties and 38 in difficulty of
articulation.

Conclusions

1. AG has a low prevalence and both genetic and
environmental factors are implicated in its appearance.
2. The diagnosis of tongue-tie is based on the medical
record of the patient and the clinical examination.
The presence of AG may be either asymptomatic or
leading in several complications.
3. The treatment of AG can be either symptomatic or
surgical, with the last one including frenectomy and
frenotomy.

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