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Case Report

Life-threatening postoperative tongue oedema: a rare complication following lingual frenuloplasty

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ABSTRACT

Ankyloglossia, commonly known as tongue-tie, is relatively common in infants. The treatment of choice is frenotomy or frenuloplasty. Rare complications include prolonged bleeding and scarring of the connective tissue. We present a case of life-threatening tongue oedema following frenuloplasty. The aim is to describe an unusual postoperative complication of tongue oedema following lingual frenuloplasty. We present a case of life-threatening tongue oedema following frenuloplasty. A 4-year-old boy presented to the Otorhinolaryngology Outpatient Department (OPD) with the complaint of speech impairment. On examination, ankyloglossia was identified, and frenoplasty was performed under general anaesthesia. As the effect of the anaesthetic agents began to wear off, tongue oedema developed. The swelling was initially mild but gradually increased. The child was unable to maintain adequate oxygen saturation on room air and was therefore provided with supplemental oxygen. A hematoma was suspected, but immediate surgical wound exploration ruled out this possibility. After one hour of monitoring, the child maintained adequate oxygen saturation with supplemental oxygen, and the tongue oedema had stabilised. Laboratory investigations and chest radiography revealed no significant abnormalities. With continuous monitoring and treatment with antibiotics, a corticosteroid, an antifibrinolytic drug, and an antihistamine, the child's general condition gradually improved. Supplemental oxygen was no longer required, and the oedema gradually subsided. The child was discharged home with the instructions to continue antibiotic and antihistamine therapy for an additional five days. We recommended follow-up in the ENT Outpatient Clinic and further allergy evaluation. Frenuloplasty is generally considered a safe procedure; however, postoperative complications may occur. This case highlights the importance of careful surgical technique, early recognition of complications and appropriate management to ensure recovery and minimise morbidity

Keywords: Ankyloglossia, Tongue-tie, Tongue oedema, Lingual frenoplasty

INTRODUCTION

Ankyloglossia, commonly referred to as tongue-tie, is a developmental abnormality characterised by a short lingual frenulum that restricts tongue movement and may extend to the tip of the tongue.[1] Ankyloglossia is a relatively common finding in infants with reported prevalence ranging from 4.2% to 10.7%.[2] It may cause difficulties with sucking in infants and, in older patients, can lead to speech difficulties or malocclusion.[3-6] The treatment of choice is frenotomy, a minor surgical procedure generally considered safe.[7,8] Rare complications include significant bleeding and scarring of the connective tissue.[9,10] We present a case of life-threatening tongue oedema following frenoplasty in a child with ankyloglossia.

CASE REPORT

A 4-year-old boy presented to the otorhinolaryngology Outpatient Department (OPD) of a tertiary care centre with a complaint of speech impairment. On examination, ankyloglossia was diagnosed, and the child was scheduled for elective frenoplasty. The procedure was performed under general anesthesia using electrocautery and a scalpel. Lidocaine and adrenaline were administered for local anesthesia. Minor bleeding occurred during the procedure and was controlled by suturing the wound with non-absorbable synthetic sutures. As the effect of anesthetic agents began to wear off, tongue oedema developed.

The swelling was initially mild but gradually increased. A lingual hematoma was considered the most likely cause of the swelling. Immediate surgical wound exploration was performed; however, no hematoma was identified. Following the withdrawal of anesthesia, the child was unable to maintain adequate oxygen saturation on room air and was therefore provided with supplemental oxygen. After one hour of monitoring, the patient maintained adequate oxygen saturation with oxygen administered at a flow rate of 2L/min. The tongue oedema had stabilised but had not yet subsided.

The child was fed enterally, and medical management included an antibiotic (ceftriaxone), a corticosteroid (dexamethasone), an anti-hemorrhagic drug (tranexamic acid), and an antihistamine. Laboratory investigations and chest radiography revealed no significant abnormalities. On the following day, the child's general condition gradually improved. Supplemental oxygen was no longer required, and the tongue oedema began to subside. The swelling resolved completely by postoperative day 5. The child was discharged home with the instructions to continue antibiotic and antihistamine therapy for an additional five days. We recommended follow-up in the ENT Outpatient Clinic and further allergy evaluation. However, the subsequent allergy evaluation did not reveal any significant findings. There was also no history of previous allergic reactions.



Figure 1: Tongue oedema after removal of intubation tube.



Figure 2: Tongue oedema on postoperative day 2.



Figure 3: Tongue oedema subsides on postoperative day 5.

DISCUSSION

Frenotomy is a relatively simple procedure and most of the authors consider it safe.[8] Complications such as pain, bleeding, scarring and lingual nerve injury have been described in the literature.[1] Rare complications including airway obstruction, tongue oedema and hypovolemic shock have also been reported. However, descriptions of these complications are limited, suggesting that awareness of them within the medical community may be limited.[11,12] In the present case, the patient developed a complication following surgical treatment. An attempt was made to identify the possible cause of oedema. Hereditary angioedema (HAE), also known as Quincke's oedema, is a genetically determined disorder associated with dysfunction of the complement system, commonly resulting from a mutation in the gene encoding factor C1 esterase inhibitor (C1-INH). HAE was considered a possible cause of the oedema.[12] However, laboratory investigations did not support this diagnosis. Elevated immunoglobulin E (IgE) levels may indicate an allergic predisposition, despite negative results on food allergen panel testing and inhalant allergen testing.[13] Negative culture results and low inflammatory marker levels made bacterial infection a less likely cause of the lingual oedema. A sublingual hematoma was also excluded during surgical wound exploration. The oedema was suspected to be a reaction to lidocaine, which had been administered for local anesthesia during the procedure. However, subsequent provocation testing did not demonstrate hypersensitivity to lidocaine. Ultimately, the etiology of the lingual oedema remained undetermined. A

possible association between the suspected allergic episode, exposure to the anesthetic agent and the development of lingual oedema was considered; however, a causal relationship could not be established.

CONCLUSION

Frenoplasty is generally considered a safe procedure. However, postoperative complications may occur and, in rare cases, can lead to life-threatening conditions. This case highlights the importance of early recognition of postoperative tongue oedema, careful surgical technique, close postoperative monitoring and prompt appropriate management to facilitate recovery and minimise morbidity.

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