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The Oji Technique: A New Method of Tongue Reduction in a Rare Combination of Multiple Orofacial Anomalies

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Abstract

We present the case of a newborn with a rare combination of trifid tongue, macroglossia, ankyloglossia, median cleft of the lower lip, bony Tumour on the mandible and cleft of the palate. Our surgical procedure consisted in the release and repair of the trifid tongue; removal of the Tumour on the mandible and repair of the cleft of the lower lip. Finally, we carried out a novel technique to reduce the enlarged tongue. This new technique of achieving uniform tongue reduction has its essential character in a rocket-like incision in the tongue, which is different in construction and quality from the over plus of techniques described so far. Among its variety of skills is the reduction of the square-like form and preclusion of the midline trough of the keyhole technique. It gives a functionally satisfactory and aesthetically pleasing result. The surgeons who apply this surgical procedure call it the Oji technique.

Keywords: Macroglossia; Novel reduction technique; Rare orofacial anomalies

Key Message

We wish to present this very rare and interesting case and the innovative surgery that we used to reduce the macroglossia to our colleagues locally and worldwide.

Introduction

Structures derived from the upper half of the first branchial arch have developmental anomalies, which are common and which result to deformities such as cleft lip or cleft palate. In contrast, abnormal or incomplete development of structures derived from the lower half of the first branchial arch is rare [1,2]. When it occurs, it may present as a complete or incomplete cleft of the lower lip, mandible, and tongue [1,3,4]. The case we present is a rare combination of Tessier 30 facial cleft (midline cleft of the lower lip; mandible and bifid tongue with ankylossia), and Pierre Robin sequence (cleft palate, micrognathia, glossoptosis). The difference, however, is that our case differs from the classical Tessier 30 because it has a trifid tongue and a mandibular midline Tumour, which have never been reported as part of Tessier 30. Again, this case does not have a glossoptosis but a macroglossia and an additional uvula cleft, thereby deviating from the classical Robin sequence. The Tessier 30 cleft, which is not commonly seen5 and the Robin sequence in our case are therefore not classical.

Macroglossia may occur as a congenital or acquired condition and the enlarged tongue may cause significant symptoms such as sleep apnea, respiratory distress, drooling, difficulty in swallowing, and dysarthria6. The reduction of the whole tongue and the preservation of its neurosensory function should be the aim in any case of macroglossia surgery. The plethora of techniques described to achieve this aim shows that there is no widely-accepted technique. Consequently, we describe a new technique that we believe will overcome most of the problems of the existing techniques. We hence considered this novel technique and this rare combination of symptoms unique to merit reporting.

Case Report

We admitted the patient in the maxillofacial unit of a tertiary hospital in eastern Nigeria when she was five days old. Her weight on admission was 3.5 kg. Examination at birth revealed a newborn with the following anomalies: macroglossia; trifid tongue; ankyloglossia; incomplete cleft of the lower lip; bony tumour in the middle of the mandible; micrognatia; cleft of the palate and uvula; nodular soft tumour on the left side of the palate (Figure 1). She had transient cessation of respiration, could hardly suck her mother's breast and drooled.

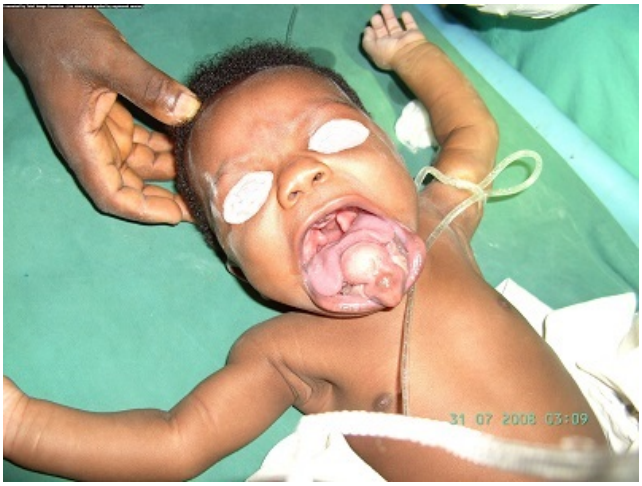

Figure 1: Newborn with multiple anomalies of the oral region.

The remainder of the physical examination was unremarkable. We designed the surgical approach individually to treat this constellation of conditions. We carried out the surgery under general anesthesia and it consisted in a repair of the ankyloglossia and the trifid tongue, removal of the tumour in the middle of the mandible and repair of the lower lip cleft, and finally the reduction of the over-large tongue. We released the ankyloglossia and repaired the trifid cleft of the tongue by initially undercutting the base of the tongue. Thereafter, we made incisions along the margins of the cleft tongue and carried the approximation in layers. We sutured using chromic catgut sutures. In the next surgery, we excised the large, firm mass in the middle of the mandible, whose pathological report was a benign fibrous tissue. In the same session, we repaired the incomplete cleft of the lower lip by a V-plasty procedure (Figure 2). In the final surgery, we carried out the tongue resection along the lines shown in Figure 2. The posterior triangular resection extended to the anterior of the circumvallate papillae for adequate reduction. The limbs of the incision diverged laterally one-third of the anterior to meet the lateral margin. Together with the posterior equilateral incision, it gives the appearance of a rocket (Figure 2).

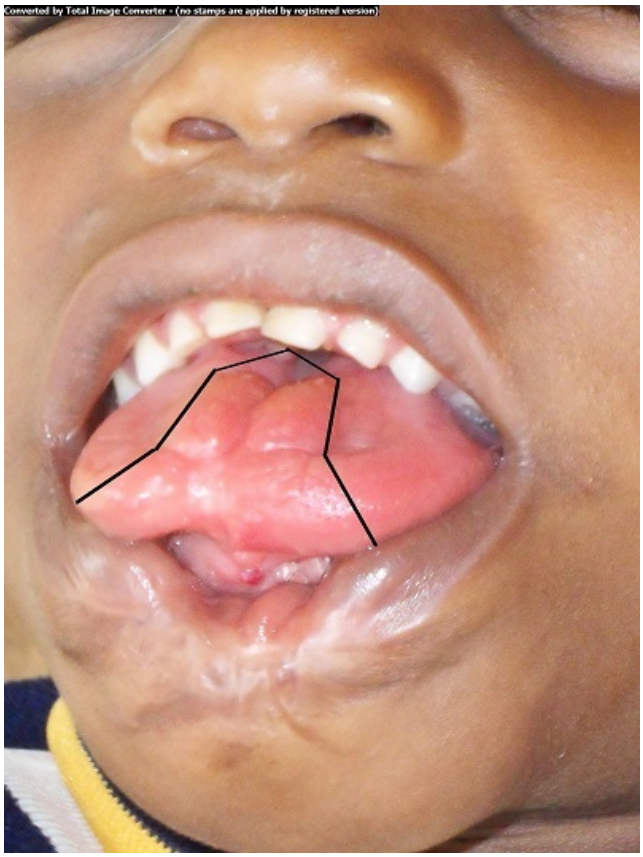

Figure 2: Markings for the V-plasty and the rocket-like incision for partial glossectomy.

We used cutting diathermy to raise lateral muco-muscular flaps, to enable excision of two horizontal triangular wedges from the central muscular part of the tongue to reduce the thickness and at the same time preserve the lingual and hypoglossal nerves. We closed the resected area in three layers with chromic suture into a straight line or an "I" shape, which reduced the length as well as the width of the tongue (Figures 3 and 4).

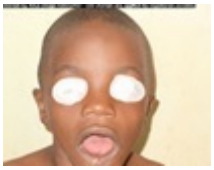

Figure 3: Patient showing a uniformly reduced tongue with good lip mobility and a satisfactory contour of the lip five years after surgery.

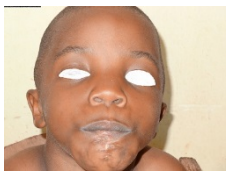

Figure 4: satisfactory mouth closures.

The tongue healed without any problems. After three years the tongue mobility and the feeding were good. She was able to identify and differentiate sweet, sour, and salty. Her speech was not satisfactory because of the cleft palate (Figure 5).

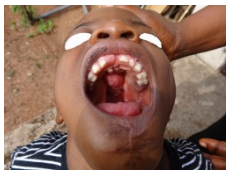

Figure 5: Condition before palatoplasty.

At the age of six, we successfully performed the palatoplasty (Figure 6).

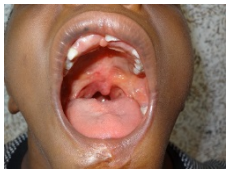

Figure 6: Condition after palatoplasty.

The delay was because the parents could not afford the cost of the CT scan and the hospital charges. The CT scan was necessary in this peculiar case because we wanted to ensure that the protrusion from the roof of the mouth through the cleft was not an extension of cranial neural tissue. Her speech has improved considerably.

Discussion

A combination of trifid tongue, macroglossia with ankyloglossia, incomplete median cleft of the lower lip, bony tumour on the center of the mandible, micrognatia, and cleft of the palate and uvula are rare [1,2,5,8]. Because this and resembling cases are not widely known, and because they vary in severity, there is lack of consensus on the mode of management and timing of surgical procedures [3,7,8] The strong parental desire for a "normal" looking child and the expected speech difficulties motivated us to perform early repairs of these anomalies. With the exception of the cleft palate, we treated all these anomalies within a space of three years. We could not perform the cleft palate surgery because the parents of the child had no money at the time to pay for further surgical intervention. However, we carried out this surgery successfully at a later date (Figures 5 and 6). Surgeons have advanced many different surgical procedures since Harris originally described the surgical treatment of macroglossia in 1835 [9]. Dingman and Grabb [10], Gupta [11] described the peripheral excision, which reduces the bulk of the tongue at the periphery while leaving the center and base bulky. Edgerton [12] performed a central elliptical excision with a view to sparing the nerves, arteries and papillae. This procedure lengthened the tongue and reduced the breadth. Kole13 described a triangular wedge excision of the tongue that was wider at the tip. This method does not address the base of the tongue and leaves it thick and wide. The novel surgical technique described in this report (Figure 2) provides a wide variety of skills. It allows increased tongue reduction with simple modifications. Although this technique is new, it may be seen as a modification of the keyhole subtotal glossectomy that was originally described by Morgan et al. [14] In contrast to the keyhole technique; this technique has the benefit of precluding the classic midline trough scar that often appears with the keyhole technique. It allows for more uniform reduction of the tongue and reduces the "square-like" form of the classic keyhole reduction technique. The square-like form may alter speech as the child grows. Expansion of the anterior wedge provides greater reduction in width and expansion of the posterior triangular incision provides increased reduction in tongue bulk and length. The versatility of this resection allows its use in almost all cases of macroglossia. In this case, we achieved uniform reduction and an improvement in cosmetics and function. In conclusion, we wish to share this innovative technique with our colleagues worldwide.

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