

Osifo OD
Akpoduado DD

Management challenges and outcome of congenital giant intra-lingual fibroma in a 2-year old boy.

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Osifo OD (✉)
Paediatric Surgery Unit, Department of
Surgery,

Akpoduado DD
Department of Anaesthesia,
University of Benin Teaching Hospital,
Benin City, Nigeria.
Email: Davidosifo4@Gmail.Com,
Leadekso@Yahoo.Com

Abstract: Oral fibromas are often acquired painless benign slowly growing tumour usually seen on the buccal mucosa along the plane of occlusion of the maxillary and mandibular teeth. Congenital intra-lingual fibroma is rare and not much has been reported on it in literature. A giant intra-lingual fibroma which was present at birth that completely filled and protrudes out of the mouth thereby resulting in feeding diffi-

culty was diagnosed in a two-year-old boy. The case is reported to draw attention to this rarely congenital tumour found in an unusual location and highlights the challenges of surgical management in a financially constrained African setting.

Key words: Intra-lingual, Fibroma, Management challenges, Outcome.

Introduction

Intra-oral tumours are very common pathology in children and the majority are congenital benign lesions which can be located in any region of the mouth. The floor of the mouth is the commonest site for the frequently diagnosed paediatric oral tumours such as ranula, haemangioma, lymphangioma, dermoid cyst, mucous retention cyst and calculi.¹⁻³ A purely intra-lingual tumour is very rare in children and there are very few documentation on tumour occurrence in this location.⁴ Similarly, although fibroma can be seen in any part of the body, including oral cavity, an intra-lingual location is rare.^{4,5} They are painless benign slowly growing tumour that hardly produce life threatening symptoms.⁴ A giant intra-lingual fibroma present from birth that grew and could not be contained within oral cavity but completely filled and protrudes out of the mouth thereby resulting in feeding and anaesthetic difficulties was diagnosed in a two-year-old boy.

The case is reported to draw attention to this rarely congenital tumour found in an unusual location and highlight the challenges of surgical management in a financially constrained African setting.

Case

Master M.W, a two-year-old boy, is the last of three children in a monogamous family. He was brought to the hospital on account of swelling of the tongue which was noticed as a small intra-lingual lump at birth. The swelling increased gradually and at the age of one year had filled and protruded out of the month as the oral cavity could no longer contain the massive tongue (fig. 1). There was difficulties closing the mouth, talking and eating which was associated with drooling of saliva.

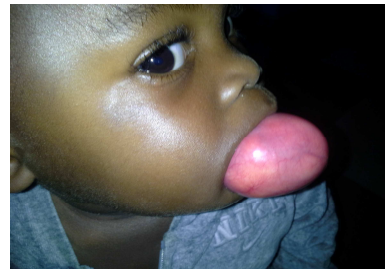


Fig 1: A two-yr-old boy with huge intra-lingual mass noticed as a small lump at birth.

Ingestion of liquid feeds was accomplished by the parents insinuating a straw between the cheek and the massive tongue. Financial constraints and ignorance resulted in the parents seeking help from quacks and traditional health care providers without improvement.

However, apart from the lingual mass the child was clinically stable, looking well fed and active on arrival. The tongue was grossly enlarged with a firm non tender and non mobile smooth mass within its substance. Haematocrit was 11.5mg%, other blood parameters and chemistry were within normal ranges and lung fields were clear on plain chest radiograph. He was booked for excision biopsy under general anaesthesia but the anaesthetist assessment showed that there was no space to insinuate laryngoscope for endotracheal intubation to secure the airway during surgery. The parents were counselled for tracheostomy but declined consent due to fear of its outcome and inability to pay the additional bills.

Consequently, the mass was excised with anaesthesia provided using intravenous ketamine 2mg/kg, midazolam 1mg/kg, atropine 1mg/kg to dry up secretion and simultaneous local infiltration of the tongue with 1% xylocaine in adrenaline 1:1000 6mg/kg. He was placed on a left lateral position with continuous suctioning of

the operating field during the operation which lasted 25 minutes. A 6 x 8 cm intra-lingual mass weighing 250g, which was histopathologically confirmed as a lingual fibroma, was well circumscribed and easily enucleated (figs. 2). Post operative course was uneventful following thrice daily oral warm saline lavage, prophylactic metronidazole and paracetamol which provided adequate analgesia. Oral feed was commenced and tolerated 4 hours after operation and he was discharged to out patient follow up on the evening of surgery. Redundant lingual tissues resolved within two weeks of follow up and he has been doing well.



Fig 2: The resected congenital intra-lingual mass which weighed 250g and was histopathologically confirmed as fibroma.

Discussion

Fibromas are said to represent reactive focal fibrous hyperplasia due to trauma or local irritation which cannot account for the occurrence of this lesion in this case. It was reported⁵⁻⁷ to be seen most often on the buccal mucosa along the plane of occlusion of the maxillary and mandibular teeth unlike in this patient where it was found within and grossly enlarged the tongue. Also in the index case, the fibroma was present at birth and diagnosed in a two years child which supports a congenital rather than acquire origin. Oral fibromas which are either sessile or pedunculated are often submucosal in location. They are painless and are usually seen in older patients following repeated abrasions from chewing and chemical irritation of oral mucosa⁶⁻⁸. Earlier authors⁹ recorded excellent results in paediatric

operations performed with parenteral ketamine and simultaneous local infiltration of operation site with xylocaine in adrenaline. These operations were performed in children older than a month with lesions located outside the mouth, and the duration of each operation lasted less than one hour. In neonates and older children with intra-oral lesions, however, securing the airway to prevent aspiration during operation is of paramount importance. Endotracheal intubation, nasotracheal intubation, laryngeal mass, or creation of tracheostomy where these are not possible, are advised in such cases^{9,10}. The huge intra-lingual mass which filled the mouth could not allow space for endotracheal intubation or placement of laryngeal mass in this child. Coupled with financial constraints and ignorance which resulted in parents refusal to consent to tracheostomy posed major challenges in surgical management of index case.

As a last resort, anaesthesia was provided using intravenous ketamine, midazolam, atropine which reduced oral secretion, and simultaneous local infiltration of the tongue with xylocaine in adrenaline⁹. Aspiration during the operation was further prevented by placing the child on a left lateral position with continuous suctioning of the operating field throughout the operation. The successful outcome recorded in excising this huge intra-lingual mass with this method may have been possible because the mass was well circumscribed and easily enucleated within 25 minutes. A less well defined mass with poorly developed plane for dissection that can result in longer duration of surgery may not have given comparably good outcome as recorded in this case⁹.

Congenital huge intra-lingual fibroma which completely filled and protrudes out of the mouth thereby resulting in feeding and endotracheal intubation difficulties was successfully excised with excellent outcome in a two-year-old boy. The case is reported to draw attention to this rarely congenital tumour found in an unusual location and highlighted the challenges of surgical management in a financially constrained African setting.

References

- Liang Y, Yang YS, Zhang Y. Retrospective analysis of ten cases of congenital sublingual teratoid cyst. *Eur J Paediatr Dent* 2012; 13: 333-336.
- Schwanke TW, Oomen KP, April MM, Ward RF, Modi VK. Floor of mouth masses in children: proposal of a new algorithm. *Int J Pediatr Otorhinolaryngol* 2013; 77:1489-1494.
- Alves TP, Pinheiro RS, Braga DN, Monteiro LP, Castro GF. Successful management of oral ranula: a rare case in an infant. *Gen Dent* 2013; 61:26-28.
- Alaeddini M, Barghamadi R, Eshghyar N, Etemad-Moghadam S. An analysis of biopsy-proven tongue lesions among 8,105 dental outpatients. *J Contemp Dent Pract* 2014; 15: 1-7.
- Kale L, Khambete N, Sodhi S, Sonawane S. Peripheral ossifying fibroma: Series of five cases. *J Indian Soc Periodontol* 2014;18: 527-530.
- Ojo MA, Omoregie OF, Altini M, Coleman H. A clinico-pathologic review of 56 cases of ossifying fibroma of the jaws with emphasis on the histomorphologic variations. *Niger J Clin Pract* 2014; 17: 619-623.
- Parwani S, Parwani RN. Diagnosis and management of focal reactive overgrowths of gingiva-a case series. *J Mich Dent Assoc* 2014; 96: 36-47.
- Durmaz EO. Denture-induced fibroma. *Clin Exp Dermatol* 2014; 39: 763-765.
- Osifo OD, Emeangui KN, Aghahowa SE. Safety profile of parenteral ketamine and lignocaine infiltration in paediatric operations. *Saudi Med J* 2008; 29: 60-64.
- Tsukamoto M, Hirokawa J, Sako S, Fujiwara S, Yokoyama T. Anesthetic management of a patient with osteogenesis imperfecta combined with mandibular defect. *Masui* 2014; 63: 679-681.