Epidermoid Cyst of the Tongue Causing Airway Obstruction

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Abstract Epidermoid cyst occurring within the tongue is rare. The aim of this article is to discuss the management of an adult patient who presented with respiratory obstruction due to this congenital epidermoid cyst within the tongue. A 32-year-old male patient who was brought to the accident and emergency department of the University of Port Harcourt Teaching Hospital, Port-Harcourt, Rivers State, Nigeria was presented. He came with a history of difficulty in breathing as a result of a slow growing swelling of the tongue which started from birth. There was associated pain, difficulty in speech and mastication as the swelling increased in size. Detailed physical examination, sonographic imaging and histopathology were carried out. On examination, patient was acutely ill-looking, in severe respiratory distress, febrile, mildly pale and dehydrated but anicteric. Extra-oral examination revealed mildly protruded upper and lower dento-alveolar arch as well splaying and recession of the anterior and posterior teeth of the upper arch. Intra-oral examination revealed moderately tender, fluctuant and grossly enlarged tongue obstructing the oro- and nasopharynx. A diagnosis of dermoid cyst was made and the patient was booked for surgery after resuscitation. Excision of the cyst was done under general anaesthesia/

naso-tracheal intubation by an intra-oral approach. Ultrasound was able to give a thorough assessment of the cystic nature of the swelling while post-operative histopathology was used to confirm the specific type of teratoma. The relationship of the cyst to the intrinsic and extrinsic muscles of the tongue and mylohyoid muscle was relevant to the surgical approach to the lesion.

Keywords Epidermoid cyst · Intralingual · Airway obstruction

Introduction

Epidermoid cysts constitute about 15–35% of all teratomas which include epidermoid, dermoid and teratoid cysts [1]. These cysts are developmental or acquired (traumatic or iatrogenic) in origin [1]. Epidermoids or dermoids can occur as an embryologic disaster around the 3rd to 5th week of gestation resulting from failure of surface ectoderm to regress and the entrapment of the epithelium [2]. They may also result from implantation of sequestered ectodermal remnants at the union of first and second bronchial arches or from ectodermal differentiation of multipotential cells [2]. Epidermoid cysts are usually congenital lesions that begin to manifest shortly after birth while dermoid cysts manifest more between 1st and 3rd decade [3]. The epidermoid cystic cavity is usually lined with squamous epithelium without the skin appendages that is conspicuous in the wall of dermoid cysts. Generally, these cysts occur almost in every part of the body including the head and neck where they form about 1.6–6.9% and are mostly found in the midline sub-mental and sub-lingual regions [3]. The aim of this article is to report on the management of an adult patient who presented with

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respiratory obstruction due to this congenital and infected epidermoid cyst within the tongue.

Case Presentation

A 32-year-old man presented in the casualty department of the University of Port Harcourt Teaching Hospital, with a history of gradual respiratory distress due to a slow growing swelling of the tongue that started from birth. The distress became severe on the day of presentation and was then rushed to the hospital. There was associated difficulty in swallowing, speech and mastication. The patient had pain from the mass occasionally which subsides with the use of antibiotics and analgesics prescribed by general practitioners. There was no relevant previous medical and dental, surgical history. On examination, patient was acutely ill-looking, in severe respiratory distress, febrile, mildly pale, and dehydrated but anicteric. The sub-mandibular nodes on both sides were palpable, mildly tender and firm in consistency. Extra-oral examination revealed mildly protruded upper and lower dento-alveolar arch as well splaying and recession of the anterior and posterior teeth of the upper arch. Intra-oral examination revealed moderately tender, fluctuant, grossly enlarged tongue obstructing the oro-nasopharynx, and mildly obliterating the anterior and posterior regions of the lingual sulcus (Fig. 1). The mucosa over the tongue appeared mildly erythematous, thin and distended with indentations from cuspal bites. There was no weakness or loss of

Fig. 1 Large swelling obliterating the anterior and posterior regions of the lingual sulcus

sensations of the tongue musculature and mucosa respectively. Examination of the systems revealed no abnormality.

Resuscitation commenced immediately by placing patient in the lateral position with the neck extended and supported with pillow. Patient was given 1 atmosphere of 100% oxygen/air through a nasal catheter connected to the control knob of the oxygen gas cylinder under absolute pressure, intermittently for 2 h to avoid respiratory alkalosis. Intravenous fluids (5% dextrose saline 1 l 8 hrly), antibiotics (i.v. ceftriaxone 1 g daily and i.v. metronidazole 500 mg/100 ml 8 hrly for 5 days), anti-inflammatory (i.v. dexamethaxone 8 mg start, then 4 mg 8 hrly for 72 h) and analgesics (i.v. paracetamol 300 mg 12 hrly for 72 h) were also administered. Patient could not tolerate oropharyngeal tube and forward traction of the tongue was not done since he was conscious. Aspiration of the swelling was done to reduce the mass and this relieved the airway considerably. The aspirate was an odourless, dirty cream cheesy material which was sent for microbiology/culture sensitivity resulted in non-haemolytic streptococcal organisms. The above-mentioned empirical antibiotics was continued based on the good response obtained.

Pre-operative ultrasound (Fig. 2a, b) showed grossly enlarged tongue measuring 5.76 cm by 6.38 cm with a huge moderately demarcated cystic space of dimensions 5.76 cm by 6.3 cm. It contained sedimented, mixed echogenic debris. The normal fibrillar echopattern of the extrinsic muscles was intact while that for the intrinsic muscles was lost. A diagnosis of dermoid cyst was made. After the relief of the airway, the patient was booked for surgery four days later so as to allow control of the infection and stabilize the patient.

Excision was done under general anaesthesia/naso-tracheal intubation by an intra-oral approach with a 4 cm horizontal incision on the ventral surface of the tongue midway between the tip and base of the tongue. Blunt dissection was done to separate the submucosa and capsule of the cyst except at the attachment of the capsule to the genial tubercle on the mandible where sharp dissection was used. During surgery, the cyst was found to extend from the base (above the vestibule of the larynx and ary-epiglottic fold) of the tongue to the tip, between the intrinsic and extrinsic muscles. The whole capsule was removed. A drain was placed and mucosa was sutured with 2-0 silk suture. The drain was left in situ for 72 h. The wound healed without any complication.

The patient was discharged 1 week after admission. Figure 3 shows patient 2 weeks after discharge and post-operative ultrasound (Fig. 4a, b) taken 3 weeks later showed the size of the tongue had reduced to 2.1 cm by 2.24 cm. The intrinsic muscles showed better fibrillation though still echogenic, the cystic space had resolved and

Fig. 2 a, b Pre-operative ultrasound images showing grossly enlarged tongue measuring 5.76 cm by 6.38 cm with a huge moderately demarcated cystic space of dimensions 5.76 cm by 6.3 cm containing sedimented, mixed echogenic debris. The normal fibrillar echopattern of the extrinsic muscles was intact while that for the intrinsic muscles was lost

was now possible to distinguish between the anterior serous and distal mucous regions of the dorsum of the tongue. Histopathology showed areas of both stratified squamous and pseudostratified columnar epithelium in the cystic lining without skin appendages which is consistent with the diagnosis of epidermoid cyst (Fig. 5a, b).

Discussion

Anatomically, the sub-mental cysts occur between the mylohyoid muscle and mentalis above and deeper part of the investing layer of the deep cervical fascia. The sublingual cysts are located between genio-glossus above and mylohyoid and genio-hyoid muscles below. In this report, the intra-lingual cyst was found between the intrinsic muscles of the tongue above, hyoglossus laterally, and genio-glossus below [1–6]. The report highlights the airway obstruction caused by the epidermoid cyst that

Fig. 3 Three weeks post operative, tongue has remarkably reduced in size

occurred within the tongue, which is a rare site. Dermoid/epidermoid cysts commonly occur in isolation as in our case, but a rare case of co-existence with bronchogenic cyst has been reported by Obiechina et al. [4]. The cyst initially presented as a painless, slow-growing swelling which later affected feeding, swallowing, speech and breathing as reported in other studies [7–14]. De-ponte and his colleagues [10] reported a case of sublingual epidermoid cyst which caused difficulty in breathing. Other workers have also documented respiratory distress in a 6-year-old girl with intralingual dermoid cyst and they reported that the distress resolved after basic resuscitative measures and excision of the cyst [15]. Obstructive sleep apnea has also been reported to occur due to these cysts [13]. Secondary infection has been documented to cause tenderness of the swellings and complications accompany systemic spread [12].

We used ultrasound and histopathology to investigate the swelling because of the cystic nature with well defined capsule. Other workers have used magnetic resonance imaging in addition [14], they used sagittal MRI to evaluate these cysts and this was able to locate the level of obstruction with better clarity compared to ultrasound. The normal fibrillar pattern of the intrinsic muscles of the tongue of our patient, which was altered in the pre-operative sonographic image, due to longstanding pressure of the cyst was restored in the post-operative image.

Depending on the site, the cysts can be excised by either an intra-oral or extra-oral approach [15]. In this patient, we excised intra-orally with a horizontal incision and this

Fig. 4 **a, b** Post-operative ultrasound showed the size of the tongue had reduced to 2.1 cm by 2.24 cm. The intrinsic muscles showed better fibrillation though still echogenic, the cystic space had resolved and was now possible to distinguish between the anterior serous and distal mucous regions of the dorsum of the tongue

enabled direct and easy access to the cyst. In a study by Brusati and colleagues [15], midline sagittal glossotomy was used to excise the sublingual cyst. Although, a number of complications such as infections/sinus tracts and malignant transformation have been documented in the literature [16], there was pre-operative infection in our case but no post-operative complication.

In conclusion, this is a report of a rare case of epidermoid cyst of the tongue presenting as a slow growing swelling which caused respiratory symptoms as it enlarges. Ultrasound was able to give a thorough assessment of the cystic nature of the swelling while post-operative histopathology was used to confirm the specific type of teratoma. Airway was relieved mainly by decompression and positioning. The relationship of the cyst to the intrinsic and extrinsic muscles of the tongue and mylohyoid muscle was relevant to the surgical approach to the lesion.

Fig. 5 **a, b** Photomicrograph showing areas of pseudo-stratified columnar epithelium with Goblet's cells in the cystic lining. Magnification $\times 40$

References

1. Shaari CM, Ho BT, Shah K, Biller HF (1995) Lingual dermoid cyst. *Otolaryngol Head Neck Surg* 112(3):476–478
2. Pancholi A, Sameer R, Prakash AV, Vijay Vaidya (2006) Midline epidermoid cyst: a rare case. *Otorhinolaryngol* 4(2):1–2
3. Smirniotopoulos JG, Chiechi MV (1995) Teratomas, dermoids, and epidermoids of the head and neck. *Radiographics* 15(6):1437–1455
4. Obiechina AE, Arotiba JT, Ogunbiyi JO (1999) Coexisting congenital sublingual dermoid and bronchogenic cyst. *Br J Oral Maxillofac Surg* 37(1):58–60
5. Lohaus M, Hansmann J, Witzel A, Flectenmacher C, Mende U, Reisser C (1999) Uncommon sonographic findings of epidermoid cyst in the head and neck. *HNO* 47(8):737–740
6. Miles LP, Naidoo LC, Reddy J (1997) Congenital dermoid cyst of the tongue. *J Laryngol Otol* 111(12):1179–1182
7. Myssiorek D, Lee J, Wasserman P, Lustrin E (2000) Intralingual dermoid cyst: a report of 2 new cases. *Ear Nose Throat J* 79(5):380–383
8. Zeltser R, Milhem I, Azaz B, Hasson O (2000) Dermoid cysts of the floor of the mouth: a report of four cases. *Am J Otolaryngol* 21(1):55–60
9. Dimov Zh, Dimov K, Kr'stev N, Kr'stev D, Baeva N, Baeva M, Yar'mov N (2000) Dermoid, epidermoid and teratoid cyst of the tongue and oral cavity floor. *Khirurgiia (Sofia)* 56:30–32

10. De-ponte FS, Brunelli A, Marchetti E, Bottini DJ (2002) Sublingual epidermoid cyst. *J Craniofac Surg* 13(2):308–310
11. Matthews J, Lancaster J, O Sullivan G (2001) True lateral dermoid cyst of the floor of the mouth. *J Laryngol Otol* 115(4):333–335
12. Mrad DK, Zriq A, Mhiri-souri M, Arifa-Achour N, Khochtali H, Tlili-Graies K (2005) Intra-lingual dermoid cyst. *J Radiol* 86(5 Pt 1):502–505
13. Koeller KK, Alamo L, Adair CF, Smirniotopoulos JG (1999) Congenital cystic masses of the neck: radiologic and pathologic correlation. *Radiographics* 19(1):121–146
14. Vogl T, Steger W, Ihrier S, Ferrara P, Grevers G (1993) Cystic masses in the floor of the mouth. Value of MR imaging in planning surgery. *AJR* 161(1):183–186
15. Brusati R, Galiotos S, Tullio A, Moscato G (1991) The midline sagittal glossotomy for treatment of dermoid cysts of the mouth floor. *J Oral Maxillofac Surg* 49(8):875–878
16. Devine JC, Jones DC (2000) Carcinomatous transformation of sublingual dermoid cyst: a case report. *Int J Oral Maxillofac Surg* 29(2):126–127