Sublingual epidermoid cyst in a neonate

Fadekemi Olufunmilayo Oginni, Taoreed Oladejo¹, Ramat Oyebunmi Braimah¹, Anthony Taiwo Adenekan²

Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Obafemi Awolowo University, ¹Department of Oral and Maxillofacial Surgery, Obafemi Awolowo University Teaching Hospital, ²Department of Anaesthesia, Faculty of Clinical Sciences, Obafemi Awolowo University, Ile-Ife, Osun State, Nigeria

Address for correspondence:
Dr. F. O. Oginni, Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, College of Health Sciences, Obafemi Awolowo University, Ile-Ife, Osun State, Nigeria.
E-mail: torera5265@yahoo.com

ABSTRACT

Epidermoid cysts (EC) in the head and neck region could be considered a rare condition representing only 6.9% of all ECs occurring in the body. They occur rarely in children and neonates. We present a case of sublingual EC in a Nigerian neonate.

Keywords: Cyst, epidermoid, neonate, sublingual

INTRODUCTION

In general, congenital cysts of the oral cavity are very rare.¹ This family of cysts comprises of ranula, lymphatic malformation, dermoid cyst, epidermoid cyst (EC), teratoma, heterotrophic gastrointestinal cyst and duplication foregut cyst.¹ They are differentiated histologically by the composition of their lining. ECs are benign cysts lined by epithelium only while dermoid cysts are epithelial lined cavities with variable types of skin appendages such as hair, hair follicle, sebaceous glands and sweat glands.²

Teratoid cysts are epithelial lined cavities with a capsule that contain mesodermal, ectodermal and endodermal derivatives. The presence of other tissues such as muscle, cartilage and bone is the hallmark of teratoid cyst.³

Most clinicians and researchers believe that ECs that appear in the midline floor of the mouth are a result of entrapped ectodermal tissue of the first and second branchial arches, which fuse during the 3rd and 4th weeks of intrauterine life.⁴ A second theory however suggests that midline ECs may be a variant of the thyroglossal duct cyst with ectodermal elements predominating.⁴

Anatomic classification divides the ECs of the floor of the mouth into three groups according to their relation to the muscles of the floor of the mouth: Sublingual cysts, located above the geniohyoid muscles; median geniohyoid cysts, located in the submental region between the geniohyoid and the mylohyoid muscles; and lateral cysts, located in the submandibular region.⁵⁶

The cystic mass can vary in size ranging from a few millimeters up to 10 cm in diameter. In general, the EC manifests only after attaining a considerable size, sometimes as late as second to third decade of life (its peak age incidence).⁷

Although uncommon in neonates, they may be absolutely significant as unrecognized or untreated EC in the floor of the mouth may impair feeding and respiration.

We present a case of sublingual EC in a neonate seen and managed at the Obafemi Awolowo University Teaching Hospital (OAUTH), Ile-Ife Nigeria.

CASE REPORT

A 26-day-old patient, product of a term pregnancy, who presented in our out-patient clinic with a sublingual swelling that was present from birth. Mother at the time was a 30-year-old hairdresser and father, a 32-year-old civil servant. Conception and delivery were uneventful. There was no difficulty with respiration or swallowing observed at birth but there was an initial difficulty with sucking. Parents were informed at the general hospital where he was delivered that swelling would recede spontaneously.
They however presented at a private hospital 72 h after his birth on account of gradual increase in size of the lesion. Needle aspiration at the hospital was followed by a gradual refill and this necessitated referral to our hospital. Examination showed a normal neonate weighing 4.16 kg with mouth kept open by a left sided oval mass measuring about 30 mm by 18 mm protruding from under the tongue and displacing the tongue to the right side and superiorly [Figures 1 and 2]. The overlying mucosa appeared clinically healthy and swelling was fluctuant to firm in consistency, non-reducible and non-pulsatile. Aspiration yielded thick yellowish brown watery fluid. Ultrasonography revealed a homogenous cystic well-defined submucosal oval mass in the floor of mouth mass measuring approximately 4.0 cm × 3.0 cm in diameter. We arrived at a provisional diagnosis of EC.

At age 8 months, there was no significant increase in size, child weighed 8.1 kg and was considered fit to undergo cyst enucleation under general anesthesia. Enucleation was carried out under general anesthesia after routine preoperative work-up and anesthetic appraisal.

Cautious inhalational induction was performed with 1-3% halothane in 100% oxygen after premedication with intravenous (IV) atropine 0.01 mg to dry the airway. Careful laryngoscopy and nasotracheal intubation with a size 3.5 mm plain portex® tube was achieved. Pharyngeal pack was inserted and anesthesia maintained with 1-1.4% isoflurane in oxygen/air mixture. Respiration was mechanically controlled and muscle relaxation was achieved with pancuronium bromide (0.8 mg IV). Intraoperative analgesia was provided and non-invasive monitoring of vital signs was performed.

Lesion was approached with a transverse sublingual incision [Figure 3a]. Blunt dissection was carried out to expose the entire cyst lining and facilitate its excision. Oral epithelium was then closed with absorbable sutures (3-0 Vicryl) [Figure 3b]. At the end of surgery, residual neuromuscular blockade was antagonized, pharyngeal pack was removed, airway sucked dry and the patient was extubated awake.

The postoperative period was uneventful. Postoperative drug regimen included IV cefuroxime 250 mg and metronidazole 150 mg 8 h for 3 days; IV drugamol 100 mg 8 h for 3 days and IV dexamethasone 1 mg 8 h for 2 days. Feeding was accomplished through the nasogastric tube for the first 3 days postoperatively; thereafter he commenced feeding per oral and oral toileting with warm saline mouth swabs. Patient was discharged home on the 4th postoperative day on oral antibiotics and oral hygiene instructions. He presented at the out-patient unit 1 week after discharge and 14 months postoperatively [Figure 4].

Histologic sections revealed a cystic cavity lined by parakeratinized, stratified squamous epithelium devoid of rete-ridges in most part. The cystic wall was composed of fibrovascular connective tissue with scanty infiltrate of chronic inflammatory cells. No dermal appendages were seen [Figures 5]. Findings were adjudged consistent with EC.
DISCUSSION

EC is extremely rare in neonates; not more than 30 cases of congenital cysts of the floor of the mouth have been reported.[7] This is the first case of intraoral EC in our unit.

Cases of EC and similar lesions have been diagnosed in the pre-natal period with sonography,[8] thus allowing for appropriate pre-natal counseling and preparation. While pre-natal diagnosis is most desirable, this was not achieved in our case. A pre-natal diagnosis in this case would have facilitated an appropriate referral and probably ensured that delivery took place in a tertiary facility. In addition, the immediate post-natal care delivery would have been more appropriate. It is important to point out the possibility of introducing infection into the lesion during aspiration and that this may have grave consequences.

Despite its increasing size, there was no active feeding or airway impairment observed in our patient. His adaptation to feeding with the initial gradual increase in size is substantiated by a satisfactory weight (8.1 kg) attained by 8th month of life.

While we waited for the patient to mature, source for the fund and present for surgery, a constant watch was kept and parents were requested to report any untoward change. Our wait no doubt, helped to improve the bulk of tissue to be handled and boost patient’s tolerance of the procedure.

Various imaging techniques have been applied in the investigation of EC. These include plain computed tomography (CT), contrast enhanced CT scan and magnetic resonance imaging (MRI). The contrast enhanced CT is a preferred method of imaging however MRI and plain CT are reported to allow more precise localization of the lesion and also enable the surgeon to choose the most appropriate approach.[9][10] Since our patient could not afford any of these, ultrasonography only was utilized.

Endotracheal intubation in cases like this is typically a challenge especially in the absence of fiber-optic intubation (a preferred method of intubation). The risk of possible rupture of the cyst especially during laryngoscopy was a reason to keep the cyst under close monitoring.

Surgical enucleation is the only effective treatment for intra oral EC. Several approaches are reported in the literature and these may be divided generally into intraoral and extraoral approaches. The choice of surgical access to EC depends on the location of the lesion in relation to the mylohyoid or geniohyoid muscles. If the cyst is located over the mylohyoid muscle, surgery is carried out through the oral cavity, whereas the extraoral incision is necessary only when the cyst is under the geniohyoid muscle or very large sublingual cysts.[8][10]

We found the intraoral approach appropriate in this case and that was employed for a total enucleation of the cyst lining.

The postoperative care of our patient included nasogastric tube feeding, close airway monitoring, prophylactic antibiotic regimen and analgesic. Oral toileting was accomplished with the use of warm saline in swabs applied carefully round the mouth.

In addition to careful dissection and tissue handling as precautionary measures against postoperative inflammatory edema, we utilized anti-inflammatory agent (dexamethasone).

Recurrence of EC is very rare. Our review shows no recurrence and normal tongue function.

CONCLUSION

EC of the oral cavity is rare in neonates. We present a case diagnosed in a neonate and successfully managed at the OAUTH.

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REFERENCES

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