
Congenital Sublingual Cyst: A Case Report

J. M. Simba¹, I. K. Marete², R.T. Kuremu³

¹Assistant Lecturer Child Health & Paediatrics Department,

²Lecturer Child Health & Paediatrics Department,

³Associate Professor Surgery & Anaesthesiology Dept,
Moi University, School of Medicine, Eldoret, Kenya.

Correspondence to: Dr. J. M. Simba, Email: maingij@gmail.com

Congenital sublingual cysts are usually rare. A neonate presented in our facility with feeding difficulties caused by a large sublingual cyst. Excision of the cyst was done and post operative period was uneventful. In this case we discuss the common presentation of such rare cyst and their management.

Introduction

Congenital sublingual cysts are generally rare. Over a period of 25 years only 83 cases of ranula were documented in Zimbabwe while Roscow et.al (2009) and Ellegard et.al (2010) noted less than 30 cases of cyst arising from duct atresia have been described in English literature. Cyst in the sublingual region could either be true cysts or pseudocyst; benign or malignant^{1,2,3,4,5,6}.

A ranula is a pseudocyst that is caused by the extravasation of mucus from the sublingual gland¹. It occurs specifically in the floor of the mouth associated with the sublingual or submandibular salivary glands as a fluctuant, unilateral, bluish soft tissue mass. They are cystic and are frequently blue owing to the Tyndall effect, whereby blue light is reflected more than red light at the interface of soft tissue and cyst. Ranula could occur following trauma or obstruction of the outlet of the glands^{1,7,8}. Similar cysts could arise from; an imperforate submandibular or sublingual duct orifice. This may result in a cystic swelling with a characteristic bluish, translucent appearance though it occurs rarely⁴. Unlike ranula cyst these have epithelial lining^{1,2,4,9}. Other differential diagnoses include dermoid cysts, lymphatic or vascular malformations, minor salivary gland tumors, and mucous retention cysts. Dermoid cyst usually occurs in the midline^{4,10}.

Patients usually present with non painful swelling noticed incidentally or by displacement of the tongue. It may also be suspected in a neonate with feeding difficulties. Interference with feeding may lead to failure to thrive if not addressed⁵. The methods, and the age of intervention are diverse in literature. Observation for 6 months has been suggested as is aspiration upto removal of ipsilateral sublingual gland^{4,7,8}. We present a case of a neonate with a congenital sublingual cyst who presented with feeding difficulties at birth.

Case Report

A 9 days old male neonate was referred to us from a district hospital. The neonate was born at term to a 21 year old primigravida through spontaneous vertex delivery assisted by a Traditional Birth Attendant at home, cried immediately after birth and was essentially normal at that point save for a swelling in the floor of the mouth that

interfered with breastfeeding of the baby. The swelling was noted on day one of life. The swelling progressively increased in size and by the time of being at our facility, the child was only able to feed by cup and spoon. No other reported problems, and the birth was uneventful. The mother had attended clinic from the 7th month of pregnancy with normal antenatal profile and nonreactive HIV screen. She did not smoke nor take alcohol during pregnancy. The pregnancy was uneventful with a normal labour of about 14hours. No history of prolonged rupture of membranes.

On arrival to our facility the neonate was in fair general condition, no pallor, cyanosis or edema, but had a tinge of jaundice. No features of dehydration noted. The vital signs were all within normal with pulse rate of 148, respiratory rate of 46, temperature of 37.1°C and had oxygen saturation of 92% in room air. Head examination was normal other than for the swelling in the mouth. Local examination revealed a cystic mass approximately 4cm in diameter on the floor of the mouth arising on the right side and pushing the tongue completely away to the left. The swelling had ulceration, approximately 1/2cm in diameter on the surface. It generally had no colour change compared to surrounding tissues. No sinus noted. The gum and lips were not involved. The submandibular and submental regions were normal. See [Figure 1](#). All other systems were normal.

The baby was reviewed and scheduled for theatre with a diagnosis of sublingual cyst. Preoperatively the complete blood count showed Haemoglobin of 15.6g/dl; White blood count of $9.7 \times 10^9/L$ (Granulocytes-26.9% & Lymphocytes-59.1%); Platelets of $383 \times 10^9/L$; Urea/electrolytes and creatinine were normal.

Pre-operative supportive treatment included antibiotics and intravenous fluids. Intraoperatively, the child was induced and intubated nasotracheally. Excision of the cyst was done, by dissecting it out, after packing the posterior oral cavity. Closure was done using absorbable suture. There was mucinous material in the cyst. See [Figure 2](#). Capsule was taken for histology. The neonate was reversed successfully and had restoration of normal anatomy in the oral cavity. See [Figure 3](#). The immediate post operative period was uneventful and the neonate was slowly reintroduced to feeds after 12hours.



Figure 1: Cyst covering most of oral cavity, tongue on extreme left

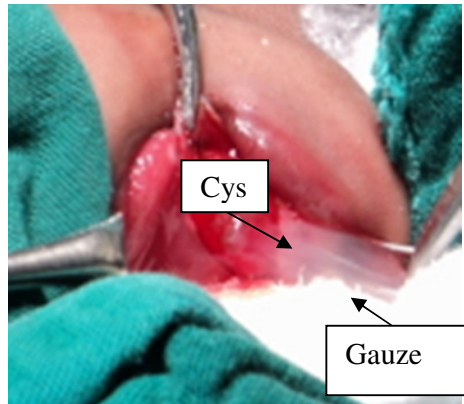


Figure 2: Mucinous substance from the cyst during the operation (Arrows)



Figure 3: Immediate postoperative period; tongue restored to its position

The neonate was discharged on the third day postoperatively able to breastfeed. The histology reported a fibrous cyst wall lined by stratified squamous epithelium. There were mucous glands seen in the wall, with a conclusion of a simple cyst.

Discussion

Cysts in the mouth are rare occurrences. The types of cysts in neonates include vascular malformations/lymphangioma, mucoceles of minor salivary glands, ranulas, mucous retention cyst or even due to imperforate salivary gland duct orifice^{1,2,4,5,11}. Sublingual cyst could be inflammatory, traumatic or neoplastic. Most oral lesions in infancy are benign¹¹. Our patient presented with a lesion at birth. This puts inflammatory/infective conditions low in our differentials. There was neither history of prolonged rupture of membranes, nor birth trauma. There was also no maternal illness. Ranulas incidence increase with HIV seropositivity but our neonate was not seroexposed^{6,12}.

Chidzonga⁶ documented only 83 cases of ranulas over a period of 25 years in Zimbabwe. So far only 29 cases of babies with sublingual/ submandibular duct atresia presenting with sublingual cyst have been described^{4,5}. Ranulas are slightly more common in females and mostly involve the left side while dermoid cysts are reported to be more common in males as are cysts arising from duct orifice atresia^{4,11,13}.

Ranula is formed following excretory duct rupture with extravasation and accumulation of saliva into the surrounding tissue. The accumulation of mucus in the surrounding connective tissue forms a pseudocyst that lacks an epithelial lining. Rarely though a ranula could occur without extravasation of mucous. The cause of ranula in neonates is however not known. In older children it is associated with trauma to the salivary duct. When the duct orifice is not patent this may end up with congenital sialocoele which is a true cyst with epithelial lining. This is thought to result from a congenital failure of canalization of the terminal end of the duct^{2,4,9}. Dermoid and epidermoid cysts 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 third and fourth weeks in utero¹⁰. The presenting complaint is usually a slow growing swelling noticed incidentally rarely causing any feeding difficulty¹⁰. Our patient presented with feeding

difficulties associated with the large size and displacement of the tongue. Any sublingual swelling large enough could present with feeding difficulties^{5,10,14}.

The clinical examination of the lesion showed a cystic lesion, whose differential diagnoses include ranula, but could also occur with cyst resulting from duct atresia and epidermoid cyst (10). Epidermoid cysts usually are in the midline while the others (ranula and duct atresia cyst) are usually lateral. The mucinous substance contained in the cyst points more towards salivary gland abnormality or related problems likely in this case ranula cyst or submandibular/sublingual duct atresia^{4,10,13}. The early presentation is in support of either a ranula cyst, sublingual duct atresia or epidermoid cyst. Most of cyst associated with duct atresia reported in literature presented at birth or as neonates^{4,5}.

Our investigations were limited on account of cost and availability. The clinical assessment satisfied the clinicians to go ahead with the operation. However, imaging is not always necessary in preoperative diagnosis and surgical planning, neither is fine needle aspiration. Investigations such as ultrasound, CT scan and MRI have been done preoperatively in other studies¹⁰. Histology done showed a fibrous cyst wall lined by stratified squamous epithelium. There were mucous glands seen in the wall. The conclusion was that of a simple cyst. Strictly speaking, ranulas have no epithelial lining though some literature classifies them into two with simple cystic lesions (true cyst) and the extravasation type which is a pseudocyst. Considering this histology report then salivary duct atresia is very likely, though very rare. Considering the stratified squamous epithelium, epidermoid cyst is a possibility but since no skin appendages were seen it becomes unlikely^{1,2,4,7,9,15}.

Treatment of sublingual cyst in neonates includes observation for spontaneous resolution in 6months^{2,16}. This in our case was not feasible since it was already interfering with feeding of the neonate. The excision of the cyst done is among several options for treatment of such lesions. Other methods which can be used include marsupialization, resection of ipsilateral sublingual gland for recurrent ranulas, needle aspiration, cryotherapy or sclerotherapy^{2,4,7,8,11,16}. Simple surgical resection is usually successful in treating most benign lesion in the mouth, including epidermal cyst and ranulas, with occasional re-excision being necessary in lymphangiomas^{4,5,11}.

Acknowledgement

We wish to acknowledge the Director, The Moi Teaching and Referral Hospital for giving us permission to publish this case.

References

1. Ryan R., German P. Management of the pediatric ranula Operative Techniques in Otolaryngology-Head and Neck Surgery Volume 20, Issue 4, December 2009, Pp 260-262 Surgical Techniques in Pediatric Otolaryngology II
2. Keqian Z., Yumin W., Wenhao R., et.al Management of infant ranula International Journal of Pediatric Otorhinolaryngology Volume 72, Issue 6, June 2008, Pages 823-826

3. Tolga K., Murat K., Enver V., et.al Sublingual epidermoid cyst: a case report Journal of Medical Case Reports 2007, 1:87
4. Rosow D.E., Ward R.F., April M.M. Sialodochostomy as treatment for imperforate submandibular duct: a systematic literature review and report of two cases. Int J Pediatr Otorhinolaryngol. 2009 Dec;73(12):1613-5.
5. Ellegård E., Kjellmer I. Atresia of the submandibular duct orifices: an unusual cause of feeding problems and failure to thrive in an infant. Acta Paediatr. 2010 Jul;99(7):1100-1.
6. Chidzonga M.M., Rusakaniko S. Ranula: another HIV/AIDS associated oral lesion in Zimbabwe? Oral Dis. 2004 Jul;10(4):229-32.
7. Chidzonga M.M., Mahomva L. Ranula: experience with 83 cases in Zimbabwe. J Oral Maxillofac Surg. 2007 Jan;65(1):79-82.
8. Mark M., Josiah E., Bethan T. et.al Conservative Treatment of Oral Ranula by Excision With Minimal Excision of the Sublingual Gland: Histological Support for a Traumatic Etiology Journal of Oral and Maxillofacial Surgery Volume 66, Issue 10, October 2008, Pages 2050-2057
9. Amin M.A., Bailey B.M. Congenital atresia of the orifice of the submandibular duct: a report of 2 cases and review. Br J Oral Maxillofac Surg. 2001 Dec;39(6):480-2.
10. Tsirevelou P., Papamantinos M., Chlousidris P., et.al Epidermoid cyst of the floor of the mouth: two case reports. Cases J. 2009 Dec 20;2:9360.
11. Ralf-Bodo T., Elinor M., Thomas F., et.al Oral tumors and tumor-like lesions in infants and children Pediatric Surgery International Volume 19, Numbers 9-10, 639-645.
12. Baurmash H.D. Ranula and HIV: an explanation of their relationship. J Oral Maxillofac Surg. 2007 Aug;65(8):1670-1;
13. Longo F., Maremonti P., Mangone G.M., et.al. Midline (dermoid) cysts of the floor of the mouth: report of 16 cases and review of surgical techniques. Plast Reconstr Surg. 2003 Nov;112(6):1560-5.
14. Saheeb B.D. Recurrent congenital bilateral ranula: a case report. SADJ. 2001 Aug;56(8):366-8.
15. García C.J., Flores P.A., Arce J.D., et.al. Ultrasonography in the study of salivary gland lesions in children. Pediatr Radiol. 1998 Jun;28(6):418-25.
16. Bernhard M.K., Hüchel D., Hamala D. Congenital ranula in a newborn [Article in German] HNO. 2007 May ;55(5):388-91.