CASE REPORT

Sublingual dermoid causing stertor in an infant

P Naina,¹ Kamran Asif Syed,² Lisa Koshy,¹ Suma Susan Mathews³¹Department of ENT, Christian Medical College and Hospital Vellore, Vellore, Tamil Nadu, India²Department of Otorhinolaryngology, Head & Neck Surgery, Christian Medical College and Hospital Vellore, Vellore, Tamil Nadu, India³Department of ENT, Christian Medical College, Vellore, Tamil Nadu, India**Correspondence to**
Dr P Naina, drp.naina@hotmail.com

Accepted 1 September 2016

SUMMARY

Sublingual dermoids are uncommon lesions in the floor of mouth. The most common age of presentation is early adulthood, but presentation in infancy has also been reported. This report highlights the clinical presentation and treatment challenges in infants. An 8-month-old infant presented with inability to close mouth, intermittent stertor and snoring. MRI suggested a sublingual dermoid which was confirmed on histopathology of the surgically enucleated specimen. Sublingual dermoids are uncommon lesions of the floor of mouth. The age of presentation of a sublingual dermoid depends on the initial size of the lesion. Large sublingual dermoids in neonates can present with respiratory embarrassment and need early surgical intervention. Surgical enucleation via an intraoral approach is the treatment of choice with external approach reserved for very large cysts below the mylohyoid. Large sublingual cysts can present with anaesthetic challenges and may need fiberoptic intubation.

BACKGROUND

Sublingual dermoid cysts are uncommon cysts in the floor of the mouth which can present across different age groups. A large sublingual dermoid causing stertor in an infant is a rare presentation. This report highlights the clinical presentation and treatment challenges of a large sublingual dermoid in an infant.

CASE PRESENTATION

An 8-month-old infant was brought by his parents with symptoms that the child always had an open mouth posture with snoring and stertor but no apnoeic episodes. His mother also noticed that the baby was unable to suckle properly during breast feeding and had to take frequent breaks. He was born of non-consanguineous marriage and there were no significant antenatal or postnatal complications. Examination of the oral cavity showed a large, cystic swelling in the floor of mouth displacing the tongue posteriorly and superiorly. The rest of the head and neck and general examinations were normal.

INVESTIGATIONS

Ultrasound screening showed a cystic lesion with round mobile echogenic foci within while MRI showed a heterogeneous cystic well-defined lobulated lesion measuring 3.5×3.7×4.3 cm. The lesion was hyperintense on T2-weighted image and hypointense to isointense on T1-weighted images (figure 1).

DIFFERENTIAL DIAGNOSIS

The differential diagnosis for midline cystic swelling of floor of mouth includes ranula, lipoma, vascular malformations such as haemangioma, lymphangioma, salivary mucocoele, duplication foregut cyst, etc. Ranula usually has a bluish grey hue and may be bimanually palpable if it is a plunging ranula. Lipoma in this region would be yellowish and soft in consistency while lymphangioma and haemangioma do not tend to be well defined. On ultrasound scanning the epidermoid cysts were seen as well-defined cysts with multiple well-defined dependent echogenic nodules within the cyst. CT scan shows a unilocular fluid-filled cyst with homogeneous hypoattenuating fluid material with multiple hypoattenuating fat density nodules within. This is typically described as 'sack of marbles' appearance and is pathognomonic of a dermoid cyst. MRI helps in knowing the exact location and extent of the lesion and the relationship to the surrounding muscles. The lesion appeared hyperintense in T2-weighted images and hypointense in T1-weighted images.

TREATMENT

Surgical enucleation via an intraoral approach was performed under general anaesthesia with fiberoptic nasotracheal intubation (figure 2). A large serous fluid-filled cyst was seen above the mylohyoid and below the tongue musculature pushing the tongue posteriorly. The tongue muscles were noted to be thinned out. The wound was closed by primary suturing. Histopathological examination showed the cyst wall was lined by stratified squamous epithelium containing flakes of keratin in the lumen suggestive of a dermoid cyst (figure 3). Adnexal structures such as hair follicles were seen with mild-to-moderate subepithelial infiltrates composed of lymphocytes and occasional plasma cells.

Figure 1 T2-weighted MRI coronal image of the infant showing hyperintense cystic lesion involving the sublingual space.

CrossMark

To cite: Naina P, Syed KA, Koshy L, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2016-217135

Figure 2 Intraoperative view of the sublingual dermoid.

OUTCOME AND FOLLOW-UP

The postoperative period was uneventful and the tongue went back to its normal position. The child was able to close his mouth, there was no stertor and the child was able to suckle well within 1 week. The child continues to be symptom free after 3 years follow-up.

DISCUSSION

Dermoid cyst of the floor of the mouth is a term loosely used to encompass a group of histologically related cysts—dermoid, epidermoid and teratoid cysts. In the largest series so far Mayo clinic in their review of 1495 dermoids in 1937, found only 6.9% occurred in the head and neck and 1.6% in the floor of the mouth. More than 250 cases have been reported in the literature so far.¹

There are two common theories regarding its origin, one suggesting that they arise from entrapment of ectoderm elements during midline fusion of first and second branchial arches while the other suggesting that it arises from tuberculum impar of His. Traumatic and thyroglossal anomaly theory has also been suggested.² Meyer *et al*³ were the first to classify these cysts based on the type of germinative layers in the cyst wall into three types. An epidermoid cyst is lined with squamous epithelium without dermal appendages while a dermoid cyst has dermal appendages in addition. Teratoid cysts on the other hand have tissues from all germ layers within the cyst. Dermoid cyst is a term loosely used to describe all types of these cysts. Cysts of the head and neck are most commonly epidermoid followed by dermoid and teratoid cyst.⁴

Colp *et al* were the first to describe an anatomical classification to the cysts of the floor of the mouth based on muscle relationships. This was further modified by Teszler *et al*⁵ based on relationship to mylohyoid and whether medial or lateral in position. They believe this anatomicosurgical classification can guide in determining the appropriate surgical approach although others do not consider this relation to be an important decisive factor.⁶

Sublingual dermoid cysts are rare in children and usually present in the second or third decade of life. Gradual growth or a sudden increase in size due to infection is the common reason for seeking medical intervention. Rapid expansion and

Figure 3 Photomicrograph showing the cyst wall lined with stratified squamous epithelium with keratin flakes (thick arrow) and hair follicle (thin arrow) within.

extension into the neck as early as in a 3-month neonate has been reported.⁷ Sudden blockage of the glands within the cyst, increased secretion of sebum during adolescence, trauma and seeding by oral flora are the commonly reported causes of infection.^{4 8} When cysts involve the submental region they can present as neck swelling or a double chin.

The differentials of a swelling floor of mouth include ranula, other embryological anomalies such as lymphangioma, thyroglossal cyst, branchial cleft cyst, infectious causes and benign and malignant tumours of the salivary glands.^{5 7 9–11} Ultrasound scanning is an easy and inexpensive way to diagnose dermoid cysts and typically shows a midline well-circumscribed unilocular mass with cystic or heterogeneous contents. CT scan delineates the relationship of the cyst to the muscle planes while MRI can confirm the diagnosis as well rule out other lesions such as lymphangioma. Dermoid cysts are isointense to hypointense to muscle on T1-weighted images and hyperintense or heterogeneous on T2-weighted depending on the internal content.¹²

Complete surgical excision is the treatment of choice and is associated with low recurrence. Risk of infection and fistula and the potential for sudden rapid increase in the swelling causing airway and swallowing problems are indications for early surgery. The ideal time for surgery would be whenever the patient seeks attention. Larger cysts can pose considerable challenge to the anaesthetist and the surgeon. The surgical approaches include both the intraoral and the extraoral. Intraoral is used mainly for sublingual lesions while extraoral for large submental and submandibular lesions. Teszler *et al*⁵ have used the anatomicosurgical classification to decide on the surgical approach. For large cysts with airway compromise options described include fibreoptic nasotracheal intubation, decompression of the cyst by aspiration of cyst and even tracheostomy.^{1 13 14} Fine-needle aspiration cytology as a diagnostic and therapeutic modality has been described; however, its role in the era of imaging is debatable.¹⁵

CONCLUSION

Sublingual dermoids are uncommon lesions in the floor of mouth. The commonly reported age of presentation is early adulthood although presentations in infants have also been reported. Presentation in the early infancy can present anaesthetic challenges. Fibreoptic nasotracheal intubation would be

the ideal option in these situations. The age of presentation of a sublingual dermoid depends very much on the initial size of the lesion and the compartment involved. It is essential to know the extent of the lesion and the compartments involved to plan appropriate exposure. Intraoral approach is generally suitable for most lesions except large cysts below the mylohyoid.

Learning points

- ▶ Sublingual dermoids are uncommon lesions of the floor of mouth.
- ▶ Large sublingual dermoids in neonates can present with respiratory embarrassment and stertor.
- ▶ They need early surgical intervention. Complete surgical enucleation is the treatment of choice.
- ▶ Very large sublingual dermoids can present with anaesthetic challenges and may need fibreoptic intubation.

Contributors PN contributed to acquisition of data, drafting of manuscript, critical revision of manuscript for important intellectual content. KAS contributed to the acquisition of data, critical revision of manuscript for important intellectual content. LK was involved in acquisition of data, drafting of manuscript. SSM was involved in the acquisition of data, critical revision of manuscript for important intellectual content and supervision.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Boko E, Amaglo K, Kpemi E. A bulky dermoid cyst of the floor of the mouth. *Eur Ann Otorhinolaryngol Head Neck Dis* 2014;131:131–4.
- 2 Longo F, Maremonti P, Mangone GM, *et al.* Midline (dermoid) cysts of the floor of the mouth: report of 16 cases and review of surgical techniques. *Plast Reconstr Surg* 2003;112:1560–5.
- 3 Meyer I. Dermoid cysts (dermoids) of the floor of the mouth. *Oral Surg Oral Med Oral Pathol* 1955;8:1149–64.
- 4 Milam M, Hill SA, Manaligod JM. Lingual dermoid cysts. *Otolaryngol Head Neck Surg* 2003;128:428–9.
- 5 Teszler CB, El-Naaj IA, Emodi O, *et al.* Dermoid cysts of the lateral floor of the mouth: a comprehensive anatomico-surgical classification of cysts of the oral floor. *J Oral Maxillofac Surg* 2007;65:327–32.
- 6 King RC, Smith BR, Burk JL. Dermoid cyst in the floor of the mouth. Review of the literature and case reports. *Oral Surg Oral Med Oral Pathol* 1994;78:567–76.
- 7 Bloom D, Carvalho D, Edmonds J, *et al.* Neonatal dermoid cyst of the floor of the mouth extending to the midline neck. *Arch Otolaryngol Head Neck Surg* 2002;128:68–70.
- 8 Black EE, Leathers RD, Youngblood D. Dermoid cyst of the floor of the mouth. *Oral Surg Oral Med Oral Pathol* 1993;75:556–8.
- 9 Ohta N, Watanabe T, Ito T, *et al.* A case of sublingual dermoid cyst: extending the limits of the oral approach. *Case Rep Otolaryngol* 2012;2012:634949.
- 10 Modolo F, de Melo Alves SJr, de Almeida Milani B, *et al.* Congenital teratoid cyst of the floor of the mouth. *Otolaryngol Head Neck Surg* 2007;136:134–6.
- 11 Kyriakidou E, Howe T, Veale B, *et al.* Sublingual dermoid cysts: case report and review of the literature. *J Laryngol Otol* 2015;129:1036–9.
- 12 Göl IH, Kiyici H, Yildirim E, *et al.* Congenital sublingual teratoid cyst: a case report and literature review. *J Pediatr Surg* 2005;40:e9–e12.
- 13 Raveenthiran V, Sam CJ, Srinivasan SK. A simple approach to airway management for a giant sublingual dermoid cyst. *Can J Anaesth* 2006;53:1265–6.
- 14 Dutta M, Saha J, Biswas G, *et al.* Epidermoid cysts in head and neck: our experiences, with review of literature. *Indian J Otolaryngol Head Neck Surg* 2013;65 (Suppl 1):14–21.
- 15 Babucco O, İşıkşacan Ozen O, Hoşnutter M, *et al.* The place of fine-needle aspiration in the preoperative diagnosis of the congenital sublingual teratoid cyst. *Diagn Cytopathol* 2003;29:33–7.

Copyright 2016 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow