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Large Sublingual Dermoid Cyst with airway compromise - An uncommon presentation

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Abstract : Sublingual dermoid cysts are very rare entities with only few cases reported in literature. They are most likely caused by the retention of germinal epithelium during the growth of mandible and hyoid arches. Here we report an unusual case of a sublingual dermoid cyst in an infant presenting with inability to close mouth and stertor since birth. The baby also had difficulty in breast feeding since birth.

Keyword : Dermoid cyst, Congenital lesions, Floor of mouth

INTRODUCTION

Epidermoid cysts and dermoid cysts are benign lesions that are encountered throughout the body. About 7% of these occur in the head and neck region and out of these 1.6% is reported to occur in the oral cavity (1). The cysts are defined as epidermoid when the lining of the lining of the cyst is only epithelium, dermoid when skin adnexa are also found and teratoid cysts when tissue such as muscle, cartilage and bone are found within the cyst. They represent less than 0.01% of all oral cavity cysts (2). They are usually found in the midline, ie submental and sublingual regions. Epidermoid cysts are congenital lesions that usually manifest shortly after birth while dermoid cysts mostly manifest between 1stand 3rd decade (3). Here we report an unusual case of sublingual dermoid cyst and review the relevant literature.

CASE REPORT

A one year old male infant was brought by the parents with history of difficulty in closing the mouth since birth. The parents also complained that the child had stertor at rest and during sleep, however there were no apnoeic episodes. The child slept well through the night. 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 consanguinous marriage and there were no significant antenatal or postnatal complications. The baby had undergone tongue tie release at 3 months of age for the similar complaint but there was no improvement with surgery. On examination, the child was active and playful. His weight and length was appropriate for age. The child had an open mouth posture with mild stertor but was not in distress. Oral cavity examination showed a large globular swelling of about 3x3 cm over the ventral aspect of the tongue obliterating the sublingual sulcus. The swelling was non tender and cystic in nature. The mucosa over the swelling was normal. There were no engorged veins over the swelling. The mass displaced the tongue posteriorly and superiorly. There was no other swelling in the neck or the rest of the body. Ultrasound screening of the oral cavity showed a cystic lesion with round mobile echogenic foci within (Figure 1). An MRI scan of the face and neck was done which showed a large heterogenous cystic well defined lobulated lesion involving the inferior and anterior aspect of the tongue. The lesion measured approximately 3.5 x 3.7 x 4.3 cm. The lesion was predominantly hyperintense on T2- weighted image and hypo to isointense on T1- weighted images. There were lobulated regions seen within the lesion (Figure 2). A possibility of a dermoid cyst was considered and it was planned for surgical excision of the lesion under general anaesthesia.

Fig.1 Ultrasound scan showing cystic lesion with echogenic foci within.

Fig.2 T2 weighted MRI coronal cuts showing hyperintense cystic lesion involving the tongue.

As the lesion was large and occupying the whole of the oral cavity, oral intubation was not considered for this child. A flexible bronchoscopy guided nasotracheal intubation was done with a 4.5mm size cuffed endotracheal tube. An oral approach was used and an incision was made over the mucosa overlying the swelling and the cyst was delineated and dissected intoto. A large cyst was seen above the mylohyoid and below the tongue musculature, pushing the tongue laterally and posteriorly. The tongue muscles were noted to be thinned out. The wound was closed by primary suturing. 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 in a week's time. On gross appearance , the cyst was filled with greyish white pultaceous material and the cyst wall measured about 0.1 cm. Histopathological examination showed cyst wall lined by stratified squamous epithelium containing flakes of keratin in the lumen. Adnexal structures like hair follicles were seen with mild to moderate subepithelial infiltrates composed of lymphocytes and occasional plasma cells. These features were consistent with dermoid cyst.

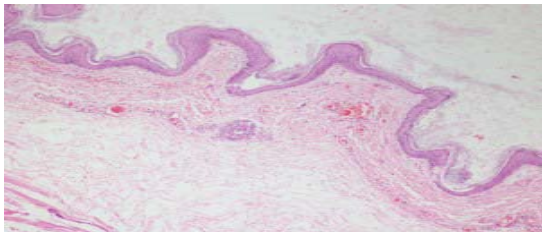

Fig.3 Photomicrograph showing the cyst wall lined with stratified squamous epithelium with keratin flakes and hair follicle within .

DISCUSSION

Dermoid cysts of the mouth are more frequently located in the midline of floor of mouth. The most popular theory for their appearance in the midline is the retention of germinal epithelium during the growth of mandibular and hyoid arches which normally fuse during third and fourth weeks of embryonic life. They might also result from prior surgical or accidental traumatic implantation of epithelial cells into deeper tissue. Clinically, they present as slow growing painless masses at the sublingual, submetal or submandibular regions. In the oral cavity they can be either sublingual or submental (4). The floor of mouth is the second most common site for dermoid cyst in the head and neck region, first being lateral eyebrow. Anatomically three different types of dermoid cysts are described in floor of mouth. They are median genio-glossal or sublingual, median geniohyoid or submental and lateral according to the anatomical relationship of the cyst with muscles of the floor of mouth. Meyer in 1955 described the histological differentiation of the cysts of floor of mouth into epidermoid, dermoid and teratoid cysts (5). The teratoid type is the only variety that can have malignant potential. The differential diagnosis for midline cystic swelling of floor of mouth includes ranula, lipoma, vascular malformations like haemangioma, lymphangioma, salivary mucocoele, duplication foregut cyst. Ranula usually have a bluish grey hue and may be bimanually palpable if it is a plunging ranula. Lipoma in this region would be yellowish and nodular. Hence imaging in the form of ultrasound or CT Scan or MRI should be performed which gives a clue for preoperative diagnosis (6). Ultrasound is the initial diagnostic modality.

Epidermoid cysts are seen as well defined cysts with multiple well defined dependent echogenic nodules within the cyst. CT scan shows a unilocular fluid filled cyst with homogenous hypoattenuating fluid material with multiple hypo attenuating fat density nodules within. This is typically described as “sack of marbles” appearance and is pathognomonic of dermoid cyst. MR imaging helps to know the exact location and extent of the lesion and the relationship to the surrounding muscles. The lesion appears hyperintense in T2 weighted images and hypointense in T1 weighted images. The mylohyoid muscle in the floor of mouth is the key structure which decides the type of surgical approach to be planned. If the cyst is above the muscle, an intraoral approach is used and cervical approach if the location of the cyst is below the muscle (7). Surgical excision is the treatment of choice and intra oral or cervical or combined approach is used depending on the location and size of the lesion. In our case, the cyst was above the mylohyoid muscle and the entire cyst was removed intraorally. Recurrence is rare if the cyst wall is removed completely.

CONCLUSION

To conclude, epidermoid cysts and dermoid cysts are relatively less common lesions in the head and neck region. To our knowledge this is the largest sublingual dermoid cyst noted in an infant. Surgical excision is the treatment of choice. Prognosis is good with low incidence of recurrence if cyst wall is completely excised. When large they can present with airway compromise in which case a flexible bronchoscopy guided intubation must be considered. A 5% rate of malignant transformation has been reported in cysts of teratoid type in literature (8). Hence it is important that these lesions are identified and treated accordingly.

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