Giant sublingual epidermoid cyst – An unusual case report

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Abstract Epidermoid cysts of the floor of the mouth represent <0.01% of all oral cysts. Only few cases have been reported so far. We report a case of a 24 years man with a large sublingual swelling for 17 years, unable to take solid meals. The swelling was approximately 13 × 13 cm, non-tender, non-transilluminant, with doughy consistency and with patent visible Wharton's duct openings on both the sides. Initial decompression of the lesion was done to facilitate nasotracheal intubation followed by partial excision of the cyst with marsupialization. Histopathological examination revealed a thick wall cyst lined with pseudostratified squamous epithelium without any evidence of dermal appendages suggestive of epidermoid cyst. Postoperative care included extraction of the loose infected dentures and physiotherapy of the ankylosed temporomandibular joints. Clinical progress was uneventful and cosmetic appearance was acceptable on 6 month and 2 years follow up with normal mastication function.

Keywords Epidermoid cyst · Floor of the mouth · Excision with marsupialization

Introduction

Although rare, a variety of congenital, inflammatory and neoplastic lesions occur in the region of the floor of the mouth. Rarer still is the epidermoid cysts occurring at the floor of the mouth. Epidermoid and dermoid cysts of the floor of the mouth account for <0.01% of all oral cavity lesions with dermoid twice as common to epidermoid [1]. We report an unusual case of a giant sublingual cyst.

Case report

A 24-year-old-gentleman presented with a swelling beneath the tongue for 17 years, unable to take solid meals and lived only on liquid diet since the age of 7 years.

Examination revealed a large spherical shaped approximately 13 × 13 cms, non-tender, non-transilluminant, doughy swelling at the sublingual region, with visible Wharton's duct openings on both the sides. The surface showed few ulcerations with loose infected dentures (Fig. 1). All investigations were within normal limits. However, X-ray mandible revealed a grossly enlarged mandible which grew along with the growth of his physique with malocclusion.

Both the Wharton's ducts were cannulated with malleable probes. Initial decompression of the cyst was done under local anesthesia, since nasotracheal intubations were not possible. Approximately 200 ml of dirty white odourless thick cheesy materials were evacuated. The Mallampati score before drainage was class 4 and after drainage was class 2. The modified Cormack and Lehane laryngoscopic view was grade II. Following which nasotracheal intubation was performed and the patient was subjected to general anesthesia.

Since the cyst wall was densely adherent to the ventral surface of the tongue, partial excision of the cyst with mar-

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Fig. 1 Preoperative photograph showing sublingual cyst occupying the whole of oral cavity

supialization was done preserving the lingual nerves and wharton's ducts. Operative findings included a cystic mass arising from the floor of the mouth. There were multiple caries and broken teeth and both the temporomandibular joints were found to be ankylosed. On the 2nd and 3rd postoperative days, extraction of the 7, 6, 5 and 4, 5, 6 lower teeth were done under local anesthesia. Patient recovered uneventfully but could not occlude the mouth fully (Fig. 2). He was discharged on the 4th postoperative day with advice to follow up for ankylosed temporomandibular joints reduction.

Histopathological examination revealed a thick wall cyst lined with a stratified squamous epithelium with acidophilic stratum corneum and basophilic dot like staining of stratum granulosum without any evidence of dermal appendages suggestive of epidermoid cyst.

Fig. 2 Postoperative day 1 showing dental malocclusion

On first follow up, the patient underwent reduction and physiotherapy of ankylosed temporomandibular joints. However, disparity in the size of the upper and lower jaw persisted with difficulty in mastication. The patient was then subjected to further physiotherapy. On 6 months follow up, the patient has been on regular solid food after 17 long years, with acceptable cosmetic appearance (Fig. 3). On 2 years follow up there is no evidence of any recurrence.

Fig. 3 On 6th month follow up - reasonable cosmetic appearance, no recurrence

Discussion

Sublingual epidermoid cysts are uncommon in the floor of the mouth. Majority of the few cases reported are histologically dermoid cysts. Epidermoid cysts are benign ectoderm lined inclusion cyst without any dermal appendages [2]. Clinically epidermoid cysts are painless, slow growing lesion with doughy consistency, often soft and well encapsulated without any associated lymphadenopathy. Large intraoral cysts usually present with dysphagia, dysphonia or dyspnoea. Contents of the cyst can be keratinous, caseous, sebaceous or purulent [3]. Only few cases have been reported and our case may be the largest ever reported so far.

Differential diagnosis of such longstanding large sublingual cysts include dermoid and simple ranula. Absolute distinction between a simple ranula, dermoid or epidermoid

cysts cannot be made by radioimaging studies [4]. Fine needle aspiration cytology (FNAC) helps in differentiating ranula from dermoid or epidermoids. However, imaging studies especially magnetic resonance imaging (MRI) provides essential information of the cyst location allowing optimal surgical approach. MRI was not done in our case as the swelling was clinically proven to be sublingual and the surgical approach would have been the same.

Surgical excision is often the treatment of choice [1]. Initial decompression of the cyst by aspiration has been reported if airway problem occurs [5]. In our case also initial decompression was done so as to facilitate intubation as it was not possible due to the large size of the cyst which occupied almost the whole of the oral cavity. Care has to be taken to delineate both the salivary gland ducts. To achieve this, lacrimal probes or gutta percha are being used to cannulate the ducts to avoid being injured during dissection [3].

Although rare, epidermoid cysts in sites other than the oral cavity harboring malignancy has been reported [6, 7]. However, no case of malignancy has been reported from the oral cavity epidermoids. Recurrence rate is low after excision. Various authors have reported different surgical approaches and techniques [8]. Our technique of initial decompression followed by partial excision and marsupialization is a simple, less invasive and effective procedure giving good cosmetic and functional results for such large sublingual cyst. In our view, the decisive factors for such an

approach are the large size of the cyst, relation to adjacent structures and the presence or absence of infection.

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