Plunging Sublingual Dermoid: A Review of Three Different Cases and Management Protocol

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Abstract

Epidermoid and dermoid cysts are rare in oral cavity. We are presenting here three such cases of sublingual dermoid cyst with submental extension (plunging dermoid) who presented in outpatient department with complaints of a slow growing painless swelling in the oral cavity and difficulty in chewing and swallowing solid foods. On clinical examination cystic swelling was seen in floor of mouth displacing the tongue superiorly and posteriorly. Ultrasonography followed by contrast CT scan of neck revealed well defined cystic swelling involving floor of mouth with submental extension. Surgical excision is treatment of choice in these cases. Although the cysts were large and extending through the mylohyoid muscle, intraoral approach was adopted for excision of cyst in all three cases avoiding extraoral neck incision for cosmetic reasons.

INTRODUCTION

Epidermoid cysts are benign swellings that are predominantly seen in areas of embryonic fusion [1]. Most of the cases have been reported in the ovaries and the testicles (80%), 7% in head and neck with most common location of external third of eyebrow [1,2]. These cysts are uncommon in the mouth and constitute less than 0.01% of all the oral cysts [2,3]. These cysts can be categorized as epidermoid when the lining presents only epithelium, dermoid cysts when skin adnexa are found and teratoid cysts with other tissue such as muscle, cartilage, and bone [4]. Dermoid cysts are congenital but they usually present in the second or third decade of life as painless slowly growing swelling. Such a swelling in the floor of the mouth can occasionally cause dysphagia, dysphonia and difficulty in breathing [5,6]. Complete surgical excision is treatment of choice for dermoid cysts which can be done either by intraoral, extraoral or combined approach, depending on the size and extension of cyst. On the basis of site of location, cysts are classified into three types (1) sublingual, (2) submental, and (3) submandibular cysts. Oral approach is suitable only for small sublingual cysts while large sublingual cysts with submental extension are preferably excised by extraoral neck incision.

CASE PRESENTATION

We are presenting here three cases of sublingual dermoid cyst with submental extension (plunging dermoid) who presented in outpatient department with complaints of a slow growing painless swelling in the oral cavity and difficulty in chewing and swallowing solid foods. There was no complaint of difficulty in breathing in any of the cases. Among three cases, 2 were females of 10 years and 13 years age respectively while 1 patient was 18 years old adult male. On clinical examination cystic swelling was seen in floor of mouth displacing the tongue superiorly and posteriorly in all three cases and largest cyst measuring 6.5 × 5 cm in one of the patient (Figure 1). Cysts were nontender, fluctuant...
and soft with normal overlying mucosa and none of the patient had associated cervical lymphadenopathy. Ultrasonography followed by contrast CT scan of neck (to see the deep extension and relations with other structures) was done in all three of them as a part of management protocol which displayed well defined cystic swelling in floor of mouth with well encapsulated margins and submental extension suggestive of plunging variety of cyst (Figure 2). Two patients underwent cyst excision under general anesthesia with nasotracheal intubation while in adult male excision (cyst size 3.5× 4.5cm) was done under local anesthesia and sedation with co-operation of the patient. The patient was explained about the chances of need of extraoral approach if required and consent was taken for the same but with meticulous dissection and traction, cyst was excised successfully via intraoral approach taking all the safety precautions to prevent aspiration.

An intraoral midline horizontal incision was given through the mucosa overlying the swelling and the cyst was separated from the surrounding tissues with proper traction and countertraction. The cyst was successfully removed avoiding extraoral neck incision in all three patients. The incision was closed in layers and post operative period was uneventful (Figure 3). Specimen on histopathological examination with hematoxylin-eosin staining revealed a cystic lesion with a stratified squamous epithelium lining and a fibrovascular connective tissue capsule, consistent with dermoid cyst.

**DISCUSSION**

Although epidermoid cysts may be categorized as congenital or acquired based on their origin, there is no difference found between the two either clinically or histologically in the literature [2,6,7]. According to proposed theories, congenital variety of cysts usually form due to entrapment of epithelial remnants during midline closure of the bilateral first and second branchial arches in the third and fourth embryonic weeks while traumatic or iatrogenic inclusion of epithelial cells or the blockage of a sebaceous gland duct have been proposed as the causative factors behind the formation of acquired cyst [1,6,7]. Roser was the one who termed dermoid cysts in the floor of the mouth as epidermoid tumours [8]. These cysts are mainly seen between 15-35 years of age with no gender predilection. Symptoms of dysphagia, dyspnoea and dysphonia may occur due to upward displacement of tongue by these sublingual swellings while more growth in a inferior direction may give rise to appearance of characteristic “double chin” [1,6,9]. On CT scans, the dermoids appear as well encapsulated cystic masses filled with a homogeneous, hypoattenuating fluid with numerous hypoattenuating fat nodules giving the pathognomonic “sack-of-marbles” appearance. On MR imaging dermoid cysts show variable signal intensity on T1-weighted images and are usually hyperintense on T2-weighted images [3]. The various differential diagnoses kept in mind for these sublingual swelling were ranula, infections of sublingual salivary glands, lipoma, thyroglossal cyst, cystic hygroma and salivary gland tumor (Figure 4). Radiological investigations allow more precise localization of the lesion in relationship to the geniohyoid and mylohyoid muscles, and they also enable the surgeon to choose the most appropriate surgical approach, especially in the case of very large lesions. Treatment is excision by via an intraoral or extraoral approach. An intraoral approach is recommended by most authors for sublingual cysts of small or moderate dimensions (less than 6 cm) above the mylohyoid muscle, whereas an extraoral approach is preferred for larger sublingual cysts (more than 6 cm). In our cases, the cysts were large (largest being 6.5 cm) and extending through mylohyoid muscle and an intraoral approach was adopted for surgical enucleation of the cyst for cosmetic purpose [10]. Recurrence is very rare with complete excision of the cyst, but a 5% rate of malignant transformation of oral dermoid cysts into the teratoid type has been reported in the literature [6,8].

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**Figure 2** Contrast CT scan of neck showing cystic swelling in floor of mouth with well encapsulated margins and submental extension.

**Figure 3** (a) Showing intraoperative dissection of cyst & (b) in immediate post operative after wound closure.

**Figure 4** (a) Showing gross appearance of excised specimen.
CONCLUSION

Complete surgical excision is treatment of choice for dermoid cysts which can be done either by intraoral or extraoral approach, depending on the size and extension of cyst. Intraoral approach is suitable only for small sublingual cysts while large sublingual cysts with submental extension are preferably excised by extraoral neck incision. With traction and counter traction most sublingual dermoids can be excised intraorally as done in our patients. Although diagnosis in such cases is mainly clinical but radiological investigations like Ultrasonography, CT scan or MRI are advocated to assess the extent and to know the relations of cyst with underlying structures that will help in surgical planning and thus achieving good surgical outcomes.

REFERENCES