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Case Report

SUBLINGUAL EPIDERMAL INCLUSION CYST: A CASE REPORT

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ABSTRACT – Presenting a case of 13 years old female with a swelling in the sublingual space with submental extension since 2 years with difficulty in speech due to impaired movements of the tongue. On oral cavity examination, a solitary mass measuring about 4x3 cm was seen in the midline in the sublingual region. On palpation, the swelling was soft to cystic in consistency with normal overlying mucosa. Extra orally, there was diffuse fullness in the submental region. FNAC suggested epidermoid or dermoid cyst. USG showed a well-defined lesion measuring 3.3x2.9x3.6cm (18cc) in midline in sublingual region, no evidence of any vascularity or calcification seen within the lesion. Surgical enucleation was done intraorally to avoid cosmetic disfiguration and swelling was removed in toto. The histopathological report was suggestive of epidermal inclusion cyst of sublingual region. The patient did well postoperatively and there was no recurrence in 1 year follow up period.

Keywords: Sublingual, Epidermal inclusion cyst, Horizontal incision.

Introduction

Keratinous cysts have been known for many years as sebaceous cysts, a misnomer born of a mistaken gross interpretation of the cyst content and perpetuated by uncritical repetition. The more common (90%), known as the epidermal or epidermoid type is lined by cornified epithelium, has distinct granular layer, and contains lamellated keratin without calcification^[1]

Epidermoid and dermoid cysts are benign lesions encountered throughout the body, with 7% occurring in the head and neck area and 1.6% within the oral cavity.^[2]

this represent less than 0.01% of all oral cavity cysts.^[3] Epidermoid cysts are the ones lined only by epithelium, whereas dermoid cysts contain skin adnexa, and teratoid cysts contain other tissue such as muscle, cartilage, or bone.^[4] Histologically, Meyer divided the cysts of the floor of the mouth into three groups: epidermoid, dermoid, and teratoid.^[5] Surgical excision remains the treatment of choice for dermoid cysts. Floor of mouth cysts can be removed by an intraoral or extra-oral route according to the localization and size of the mass.^[6] They may reach a large size, involving more than one anatomical area.^[7]

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Such swelling in the floor of the mouth can occasionally cause serious problems for swallowing and speaking.^[8,9]

Case Report

A 13 years old girl presented in our department with the chief complaint of a swelling below the tongue since 2 years. It was initially pea sized and markedly increased to the present size in last 4 months. The patient complained of difficulty in speech due to impaired movements of the tongue. There were no complaints of pain, difficulty in chewing food and difficulty in breathing.

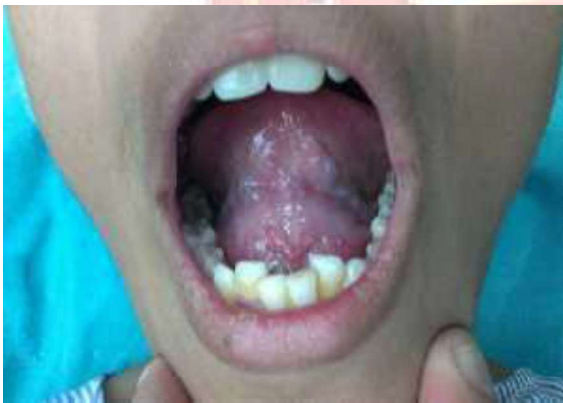


Figure1: Preoperative intraoral view of the patient showing diffuse swelling of about 4x3cm

On oral cavity examination, a solitary mass measuring about 4x3 cm was seen in the midline in the sublingual region. On palpation, the swelling was soft to cystic in consistency, non-tender, fluctuant, immobile with normal overlying mucosa. Extra-orally, there was diffuse fullness in the submental region. Transmission of movements between intraoral and submental swelling were felt.

There was no regional lymphadenopathy associated with the swelling.

A provisional diagnosis of sublingual swelling was dermoid cyst or epidermoid / epidermal inclusion cyst or plunging ranula.

Routine hematological investigations were within normal limits.

USG showed a well-defined lesion measuring 3.3x2.9x3.6cm (18cc) in midline in sublingual region, no evidence of any vascularity or calcification was seen within the lesion.

FNAC showed many squamous cells and debris (both nucleated and a nucleated squamous cells), features suggestive of epidermoid, dermoid cyst.

CT Scan showed a well-defined homogenous lesion of cystic attenuation in midline in sublingual space. No evidence of any internal calcification or heterogeneity. It is also extending into the submental space.

Excision of swelling was done under general anesthesia with nasotracheal intubation. A horizontal incision was placed intra orally on the floor of mouth yielding a smooth surfaced oval mass of tissue measuring approximately 4x3x3 cm. It was excised in toto. It was soft in consistency and cystic in nature. Post – operative period was uneventful. She was discharged on the 8th postoperative day. She was seen in the outpatient department after 2 weeks and then every 3 months for 1 year and no recurrence was noted.



Figure2: Per-operative photo showing intraoral horizontal incision

The histopathological investigation showed cystic cavity filled with cheesy material and wall thickness 0.2cm on gross examination. Microscopic sections showed features of epidermal cyst. Hence, it was conclusively diagnosed as epidermal inclusion cyst, sublingual region.

Discussion

Epidermoid and dermoid cysts of the oral cavity represent less than 0.01% of all oral cavity cysts. Histologically, this distinction of the cysts in the floor of the mouth was presented by Meyer in 1955^[11]. The cyst is described as epidermoid when the lining presents only epithelium, dermoid when skin adnexa are found, and teratoid when other tissues such as muscle, cartilage, or bone are present within the cyst^[12]. Dermoid cysts of the floor of the mouth are dysembryogenetic lesions derived from the entrapment and Subsequent growth of epithelial cells during

midline fusion between the first and second branchial arches in the third and fourth embryonic weeks^[13]. Acquired forms are derived from either iatrogenic or traumatic inclusion of epithelium and skin appendages.



Figure 3: A per-operative view of the cyst Longo et al prepared a report of 16 cases of midline (dermoid cyst of floor of mouth) with review of surgical techniques. They observed that male patients were more frequently affected with the male to female ratio of 3:1 (12:4 cases). Patients ranged in the age from 5 to 51 years¹¹. The case presented here was a 13 year old girl.

Longo et al also used ultrasonography in all cases but one, computed tomography in 8 cases and magnetic resonance imaging in 3 cases^[11].

Treatment is by enucleation 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) and above the mylohyoid muscle, whereas an extraoral approach is preferred for larger sublingual cysts (more than 6 cm) or for very large dermoid cysts affecting the submandibular and submental spaces and in cases of infection that could compromise the patient's

airway. El- Hakim and Aryaman used an intraoral approach for large, deep-seated uninfected lesions, obtaining good aesthetics and function [12-13]. Recurrence is very rare with complete excision of the lesion, but a 5% rate of malignant transformation of oral dermoid cysts into the teratoid type has been reported in literature^[14].



Figure 4: Specimen following intraoral removal

Conclusion

Epidermal inclusion cyst in the oral cavity is an uncommon entity. This is a slow growing painless mass of sublingual region which may extend into submental region through mylohyoid muscle. The cyst was completely removed through intraoral approach without any complications and no evidence of recurrence in 12 months follow up period. It should be emphasized that it is not possible to determine the specific histologic subtype through fine-needle aspiration, USG or CT scans. Thus, microscopic examination will always be required following excision of the lesion.



Figure 5: Post-operative day 8th photo

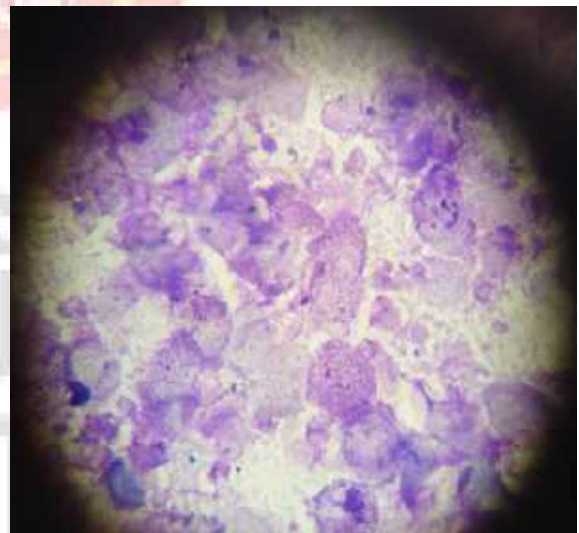


Figure 6: Fine needle aspiration cytology slide showing squamous cells and debris.



Figure 7: Histopathology slide showing a section of cyst wall lined by stratified squamous epithelium

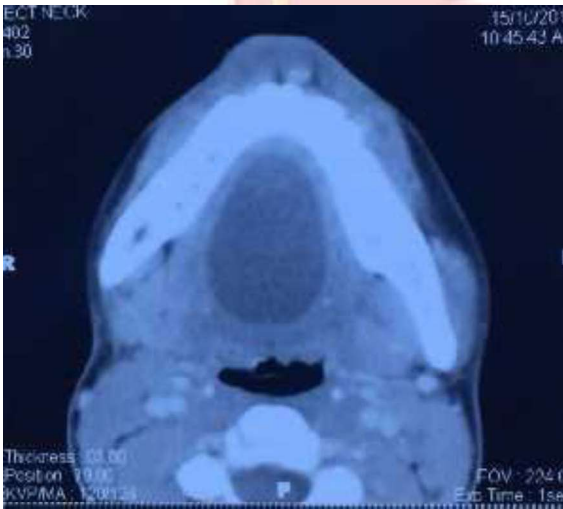


Figure 8: CT scan showing well defined homogenous lesion of cystic attenuation in midline in the sublingual space.

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