A Huge Epidermoid Cyst Endangering Life

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Abstract Epidermoid cyst of the neck or auricular area are relatively more common than that of the oral cavity. In most cases about 80% they remain asymptomatic. But in about 20% cases it becomes painful because of secondary infection seeking treatment. Here we present a case report of biopsy proved Epidermoid cyst with life threatening infection.

Keywords Epidermoid cyst · Oral cavity · Life threatening infection

Introduction

Epidermoid cysts seldom occur in the floor of the mouth though they can occur anywhere in the body. The reported incidence of dermoid cysts of the head and neck region range from 1.6 to 6.9% and those located in the mouth make up less than 0.01% of all oral cysts [1–3].

Epidermoid cysts develop from the entrapment of ectodermal elements at the fusion sites of the first and

second branchial arches during the third and fourth embryonic week [4, 5].

It commences shortly after birth, grows slowly and painlessly. That is why it draws little attention until it causes discomfort [2, 6, 7]. These cysts present either as solid or cystic masses in the midline of the neck between suprasternal and submental region.

They can occur lateral to submandibular gland and or rarely in the floor of the mouth.

Here we present a case which attained a huge size causing obstructive signs like dysphagia, dyspnoea, and dysarthria with a life threatening situation after being secondarily infected.

Case Report

A 61 year old female patient reported to the maxillofacial clinic of Dr. R. Ahmed Dental College & Hospital, Kolkata, with a huge, soft and fluctuant swelling involving right side of the neck crossing the midline [Fig. 1]. The patient could not tell the onset and duration of the swelling precisely but complained of small swelling in submental region since her childhood. Since last 2 months there was remarkable increase in the rate of growth of the swelling leading to progressive difficulty in speech, eating and later on respiratory problems. Intraorally it occupied almost whole of the floor of the mouth pushing the tongue against the oropharynx [Fig. 2]. Patient was facing respiratory difficulty in supine position due to further fall back of the tongue against the oropharynx because of weight of the swelling on the tongue. So she preferred to seat upright with head bent forward to keep the airway patent. She was almost unable to speak or eat anything including the fluid which lead to urgent hospitalization.

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Fig. 1 Showing submandibular swelling extending to the neck and crossing the midline

Fig. 2 Showing intraoral view of the swelling occupying almost entire oral cavity

On clinical examination, the swelling was found to be soft and fluctuant. The overlying skin in the neck was glossy and erythematous. With these clinical findings, a differential diagnosis of epidermoid cyst, dermoid cyst, and plunging ranula was made.

An USG of the affected area was performed which revealed cystic space occupying lesion (SOL) measuring 51×39 mm with homogenous low level internal echoes seen just below the (Rt) mandible with well defined margin. CT scan of the affected area revealed a well defined cystic SOL 48×68 mm (approx.) seen at the floor of the mouth extending from the under surface of the tongue inferiorly towards submental space to the right side. Due to pressure effect, the genioglossus muscle was pushed to the right side and the lesion was extending to the right side submental region below the mylohyoid muscle. Posteriorly the lesion was seen extending up to oropharynx obliterating the right space. No bony erosion was seen. Surrounding soft tissue showed inflammatory changes. Multiple enlarged lymph nodes were seen at internal jugular group.

Fig. 3 CT scan of the lesion in axial section showing the extent of the lesion

Rest of the visualised part of oropharynx was normal. Right pyriform fossa was normal. Neck vessels were normal. Parotids and left submandibular gland were normal, right submandibular gland was not seen separately. The CT scan did not show the presence of any specific structures [Figs. 3, 4].

Aspiration of the cystic lesion intraorally through the floor of the mouth revealed thick yellowish-white, granular fluid. This led us to focus our clinical diagnosis more towards epidermoid cyst. Based upon CT scan report, an operative plan of excisional biopsy was formulated. Other reports of relevant laboratory investigations were within normal limit. But suddenly she developed acute respiratory distress probably as a result of development of secondary infection and was immediately relieved by incision and drainage through dependent submandibular region. At least 150 ml of yellowish granular fluid was drained. Parenteral administration of cefotaxime sodium and metronidazole in suitable dosage were proved useful. Four days after the admission to the hospital, the patient was operated under endotracheal intubation. Enucleation of the cystic lesion was done mainly by intraoral approach, partly supplemented by extraoral procedure. Incision was given at the lingual mucogingival line and blunt dissection was performed to expose the whole mass which was then dissected out and sent for histopathological examination.

The histopathological report revealed keratinized squamous cystic epithelium of variable thickness. The surrounding fibrous capsule revealed moderate inflammation with chronic inflammatory cells, increased vascularity and extravasated RBCs [Fig. 5]. No odontogenic tissue was

Fig. 4 CT scan of the lesion in coronal section showing the extent of the lesion

Fig. 5 Photomicrograph of the cystic lesion showing keratinised squamous epithelium of variable thickness and fibrous capsule with moderate infiltration of chronic inflammatory cells, increased vascularity and extravasated RBCs

found to be associated with the lesion. The overall histopathological features were suggestive of epidermoid cyst. The post-operative healing period was smooth and uneventful. The patient was discharged after suture removal on seventh post-operative day [Figs. 6, 7].

Fig. 6 Extraoral view after suture removal on seventh post-operative day

Discussion

Various theories have been proposed regarding the origin of the dermoid cyst. As mentioned earlier, they may develop from the entrapment of epithelial remnants at the fusion site of first and second branchial arches during third to fourth fetal week. Some authors consider it as a variant of the thyroglossal duct cyst. Finally, previous surgical or accidental events may cause traumatic implantation of

Fig. 7 Intraoral view of the patient on seventh post-operative day

epithelial cells into deeper tissues which may give rise to an implantation dermoid [4, 8].

Histologically the dermoid/epidermoid cysts are lined by the stratified squamous epithelium. In our case the cystic lumen contained only keratin, no skin appendages in the cystic wall or any special structures like bone, muscle or odontogenic tissues were associated with it. Therefore it was diagnosed as epidermoid cyst [9, 10, 11].

The differential diagnosis of swelling of the floor of the mouth includes—I: infection of the floor of the mouth involving sublingual or submandibular spaces, II: ranula (plunging type), III: blockage of Wharton's duct, IV: lipoma, V: thyroglossal duct cyst, VI: cystic hygroma, VII: branchial cleft cyst, VIII: benign and malignant tumour of the floor of the mouth and adjacent salivary gland [9].

Dermoid/epidermoid cyst often remain asymptomatic for many years and grows painlessly. Usually attention is drawn when complication like masticatory or breathing difficulty arises as a result of huge growth like this case. Clinician must be cautious before needle aspiration or performing biopsy on the floor of the mouth because of its high risk of developing secondary infection and intensive inflammatory response due to high vascularity of the area. To perform this procedure of aspiration or biopsy in hospital set up are always better and preferred option [2]. Endotracheal intubation in this kind of patients is also a challenging job. There is every chance of rupture of the cystic lesion at the time of laryngoscopic examination, which may lead to aspiration of the cystic fluid. The

surgeon should keep the tracheostomy set ready to manage the airway in case of emergency to avoid catastrophe. Though fiberoptic intubation is a preferred procedure in this case, the challenge can be taken care of with caution, if this facility is not available.

Treatment of the dermoid cysts is complete surgical excision. Recurrence is rare. Many surgical accesses have been described depending on the location and size of the lesion. Surgical approaches, such as transcutaneous, extended median glossotomy, median glossotomy and mid line incision, may be performed [8, 12]. In our case surgical procedure involved both extra and intra oral approach because of the massive size of the lesion to enucleate it and one year follow up period showed no recurrence.

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