

KERITINIZING DENTIGEROUS CYST : A RARE ENTITY

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Introduction

Dentigerous cyst is the most common of all odontogenic cysts and constitutes over 33% of all cases of jaw cysts. The mandibular third molar region is the most (77%) common site of dentigerous cyst [1]. However, keratinizing dentigerous cyst is very rare and to the best of our knowledge has been reported only thrice in literature [2,3,4]. We report the fourth case of keratinizing dentigerous cyst. The patient presented with a complaint of long standing severe trismus with 8 mm mouth opening due to masseter muscle spasm caused by recurrent infections of dentigerous cyst. The enucleation of cyst with release of masseter muscle was planned. Mouth opening showed significant improvement post operatively. The histopathological examination revealed an unusual variant of odontogenic cyst, keratinizing dentigerous cyst arising from mandibular third molar.

CASE REPORT

A 32 year old female patient reported to us with the chief complaint of inability to open mouth completely since 3 years. The patient had a history of recurrent swelling on right side of face which used to subside after taking antibiotics. On examination there was mild swelling on right side of face which was nontender, nonfluctuant and firm. She presented with 8mm interincisal opening of mouth (fig.1). On intraoral examination there were generalized calculus deposits and a small sinus opening with respect to apparently normal second molar, First molar was missing in fourth quadrant (fig.2). TMJ movements were felt on palpation. There was no history of any trauma. Patient was advised a CT-Scan of face to rule out any underlying tumor, mass or fibrous ankylosis of TMJ restricting the mandibular movements. The images revealed a huge cyst with impacted third molar extending from first molar region to ramus and involving the inferior border of mandible. There was also buccal and lingual cortical perforation (fig.3). The buccal perforation of



the ramus has led to the infection of masseter muscle and a small radiolucent area adjacent to cortex can be clearly seen suggesting a submassetric abscess. A mildly thickened masseter muscle is also evident in the same region (fig.4).

The trismus was suspected due to repeated infections of masseter muscle leading to fibrosis and spasm. Excision of cyst was planned along with the release of masseter muscle from ramus of mandible if required. Preanaesthetic checkup and medical consent was taken prior to surgery.

The procedure was carried under general anaesthesia with fibroptic nasal - endotracheal intubation. Extraoral approach was planned to prevent the intraoral contamination of the cystic cavity.

Extraoral submandibular incision was given two fingers below the mandible. The flap was raised while preserving the marginal mandibular nerve. Buccal cortical plate was removed to attain access to the dentigerous cyst. The cyst was removed in toto along with the removal of impacted third molar. The displaced Inferior alveolar nerve was preserved (fig. 5). The specimen was sent for the histopathological examination. The histopathological specimen showed a cystic lining along with the keratin in the lumen. The Lining was typical of the dentigerous cyst but keratin in the dentigerous cyst was not common. So, the diagnosis of Keratinizing Dentigerous cyst was given.(fig6,7,8)

The mouth opening was reassessed. There was no improvement in the mouth opening. The masseteric muscle was released from ramus of mandible. A small chronic abscess masseteric muscle was noticed in the area of buccal fenestrations. The abscess was thoroughly drained and curetted. A 40 mm mouth opening was achieved and closure was done in layers. Post operative physiotherapy was done. Healing bone can be seen in post operative x-ray taken two months after surgery (fig.9) Mouth opening of 35 mm was maintained at six months follow up checkup (fig. 10).

DISCUSSION

The keratinizing dentigerous cyst, to the best of our knowledge has been reported thrice previously in literature[2,3,4]. We present the fourth case of keratinizing dentigerous cyst. Characteristically, the epithelial lining of dentigerous cyst is not keratinized. Mostly odontogenic keratocyst which is now known as Keratocystic tumors is keratinized and such pathology shows basal palisading of epithelium, hyperchromatism of nuclei and not merely the presence of keratinization. The present case did not show any of these features. The histopathological section revealed focal areas of keratinizing cystic epithelium exhibiting a prominent granular layer. The cystic lumen is filled with keratinaceous material. The CT-scan shows the well defined radiolucency along with the impacted tooth. The significance of keratinization in dentigerous cyst is not yet clear. However, it is possible that dentigerous cyst might be arising from more primitive cells of the developing enamel organ apart from the reduced enamel epithelium.

When infected, the large dentigerous cyst can cause the perforation of cortical bone. In our case this perforation caused a submassetric abscess and subsequent fibrosis of the masseter muscle leading to the only presenting feature of persistent trismus for three years.

Trismus can occur due to diseases of TMJ and muscles of mastication. This may include internal derangement of TMJ, ankylosis, inflammation and fibrosis of masticatory muscle or masseteric space infection. The submassetric space (SMS) is located between the masseter muscle and the mandibular ramus.

Hoel et al have indicated that there is a zone of muscle insertion that is less dense and can accommodate abscess formation [5], while the anatomic study by Brarsky-zachary supported the presence of a true space [6]. He found a narrow bare area devoid of masseter muscle insertion extending from the anterior border of the mid portion of the ramus upwards and backwards to condylar neck region.

Louis Mondel reported two cases of submassetric abscess caused by a dentigerous cyst mimicking a parotitis [7]. Nishimena et al reported a case of chronic organized abscess of right masseteric muscle that developed following parotidectomy. Total eradication of abscess and myotomy of muscle resolved the problem completely [8].

Cooper suggested the division of muscle fibers as a method of relieving myofacial pain in the masseter and temporalis when non surgical measures have failed and pain is localized in these muscles [9]. The submassetric space (SMS) is located between the masseter muscle and the mandibular ramus.

Though the primary etiology in our patient was dentigerous cyst, the expansion and enlargement of the cyst has caused perforation of the buccal and lingual cortices of mandible leading to the submassetric space extension of the cyst. This has probably caused the pressure necrosis in the area leading to the submassetric abscess and repeated inflammations of the masseteric muscle. The resultant spasm and fibrosis have caused reduced trismus in the patient which was the only evident clinical sign apart from a small sinus opening intraorally. The purpose of this paper is to review another cause of trismus which might gets misdiagnosed as TMJ ankylosis , parotitis or might remain undiagnosed for a long time as in our case.

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Fig:1Pre Operative with reduced mouth opening



Fig:2 – intra oral view with sinus opening



Fig 3: Buccal and lingual cortical plate perforation

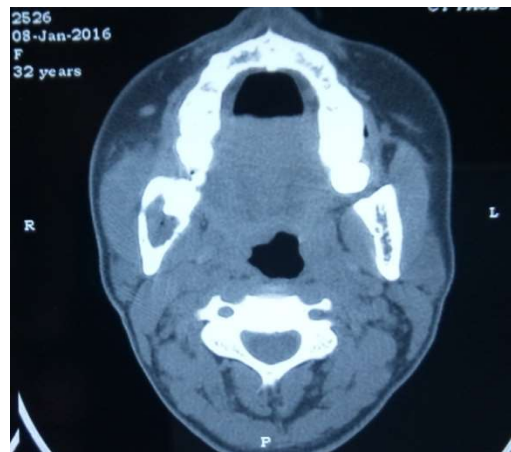


Fig:4 Right masseter muscle thickening



Fig:5- removal of the cyst along with the impacted tooth

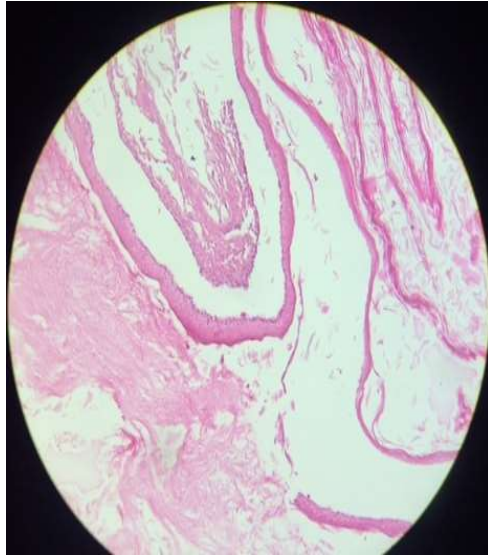


Fig:6– Dentigerous Cyst lining of varying thickness under 4X magnification

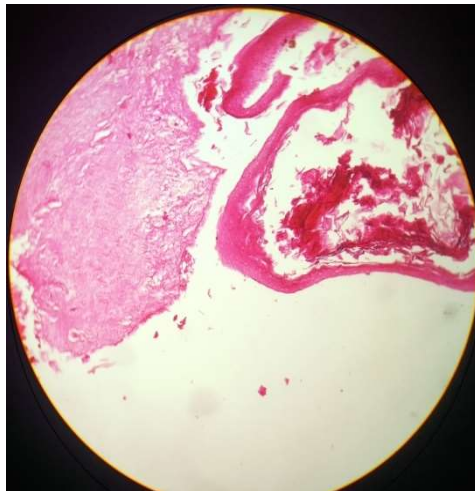


Fig: 7 Dentigerous cyst with Keratin in the lumen under 10x magnification



Fig:8 Keratinizing dentigerous cyst under 100X magnification



Fig:9 Two months Post operative xray showing bone formation



Fig: 10 Six month follow up with 35mm of m