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MANAGEMENT OF ODONTOGENIC KERATOCYST (OKC) WITH SEGMENTAL RESECTION: A CASE REPORT

Maxillofacial Surgery

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ABSTRACT

An odontogenic keratocyst (OKC) is renowned for its distinctive behaviour, diverse origins, contentious development, unique tendency to recur, and debated treatment approaches. Consequently, it has been the focus of extensive research over the past four decades. Previously labelled as odontogenic keratocyst (OKC), it was renamed keratocystic odontogenic tumor (KCOT) by the WHO in 2005, only to be reclassified as OKC again in 2017. Here, we present a case involving a 37-year-old male patient who experienced delayed developmental milestones and was initially diagnosed with an extensive OKC featuring lingual perforation. The patient underwent treatment with segmental resection of the mandible followed by reconstruction using a reconstruction plate. Histological examination of the specimen ultimately confirmed the diagnosis of OKC with ameloblastomatous changes. The clinical, radiological, and histological characteristics of this tumor, as well as its surgical management, are thoroughly discussed.

KEYWORDS

ODONTOGENIC KERATOCYST (OKC), MANDIBLE SEGMENTAL RESECTION, AMELOBLASTOMOUS TRANSFORMATION

INTRODUCTION:

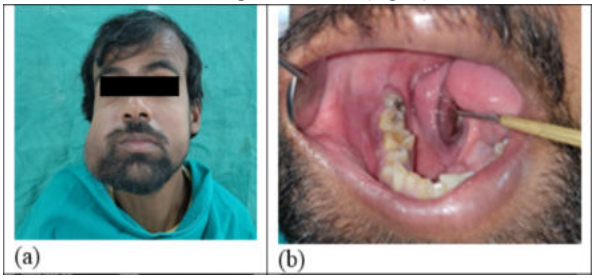
The entity previously known as Keratocystic Odontogenic Tumor (KOT) has beenrebranded as Odontogenic Keratocyst (OKC) in 2017 and stands out as a contentious subject in the realm of maxillofacial pathology. [1] OKC is distinguished by a characteristic structure featuring a thin, regular lining of keratinized stratified squamous epithelium adorned with palisading hyperchromatic basal cells. Unique to OKCs is their propensity for local aggressiveness, coupled with a notably high rate of recurrence and a tendency for multiplicity.[2] Recurrence often ensues due to inadequate treatment approaches, incomplete excision of the cyst or lingering epithelial islands post-enucleation, a heightened mitotic index in epithelial cells, considerable cyst size, and the challenge posed by lesion locations associated with complex surgical access.

CASE REPORT:

A 37 years old male presented swelling on lower right side of the face since past 7 years. The swelling has increased gradually to attain its present dimension along with intra oral pus discharge since past 2 week from that site. Fine Needle Aspiration cytology of the lesion has revealed features suggestive of Acute Necrotizing Inflammation.

Though there is absence of any accompanying comorbidities, the patient exhibits delayed developmental milestones.

On extra oral examination a diffuse, firm, non-tender, non-fluctuant swelling approximately 4 cm in diameter was present on the right cheek region extending antero posteriorly from the angle of the mouth up to the right ramus of mandible. Supero inferiorly extending from 3 cm below the right orbit up to 0.5 cm above the right lower border of mandible. The overlying skin appeared stretched with no changes in colour of overlying skin. There was no rise in temperature noted. (Fig 1 a)



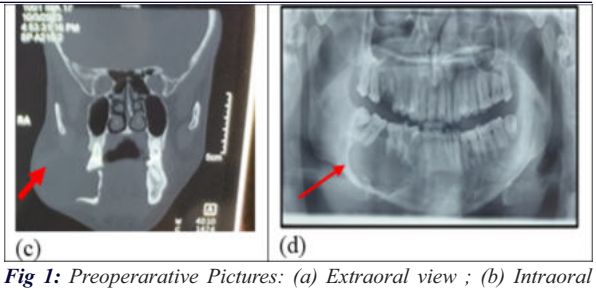


Fig 1: *Preoperarative Pictures: (a) Extraoral view ; (b) Intraoral view; (c) CECT scan (d) OPG*

Intra orally a diffuse firm, non-tender swelling of size approx. 3 cm x 3 cm X 1cm was present on the right lower buccal vestibule with vestibular obliteration from 43 to 48. On palpation, it was revealed to have a bucco lingual expansion bleeding and pus discharge was elicited.(Fig 1b)

On aspiration, a straw colored fluid mixed with blood was obtained, which was sent for protein estimation and report revealed 0.5 gm % protein present in the cystic fluid.

Orthopantogram revealed a well-defined well corticated Unilocular radiolucency, measuring approx. 5cm x 3 cm is size extending A-P from mesial side of 44 to distal root of 48 alongwith root displacement but no root resorption of teeth was observed. (Fig 1c)

CECT face revealed a large expansile space occupying lesion arising from right side body of mandible alongwith lingual perforation. (Fig 1d)

Histopathological report from incisional biopsy gave an impression of features suggestive of Orthokeratinized cyst.

Treatment decided involved surgical resection in form of segmental Resectioning of mandible followed by reconstruction with Reconstruction Plate

After routine pre anesthetic examinations, Department of Anesthesia provided the clearance for Surgical procedure under general anesthesia. The patient was intubated with naso endotracheal

intubation and was prepped and draped as per standard surgical protocol. Anatomic landmarks were marked on skin including the ramus condylar unit alongwith the extraoral extent of the lesion. (Fig 2a)

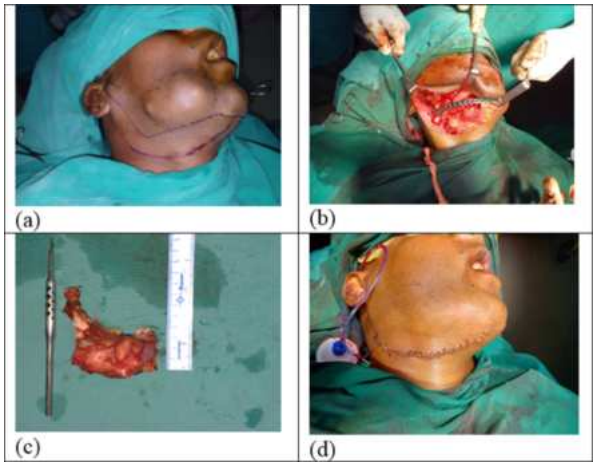


Fig 2:*Intraoperative Procedure: (a) Marking of anatomic landmark; (b) Mandibular segmental resection with reconstruction ; (c) Excised specimen ;(d) Closure*

An extraoral submandibular incision was placed and blunt dissection was carried through skin, subcutaneous tissues, superficial cervical fascia and platysma muscle. Sub platysmal flap was elevated. Blunt dissection was carried out in superior direction while preserving the facial artery and submandibular gland. Tissue dissection was done to separate the lesion from overlying skin. Segmental resection of mandible from distal to 41 to ramus along with coronoidectomy of mandible was performed. The lesion was excised along with the mandible and associated teeth and overlying mucosa. Reconstruction plate was placed to strengthen the remaining bone tissue and to reduce the risk of fracture. Mandibular movement was checked (Fig2b)

A drain was secured and extraoral closure was done in layers by using 3 – 0 vicryl for subcutaneous sutures and followed by skin sutures with 4 – 0 ethylon.(Fig 2d). Intraoral sutures were placed with 3 -0 vicryl. Specimen was sent for Histo pathological examination (Fig 2c)

Post operative healing was uneventful and there was adequate mouth opening.

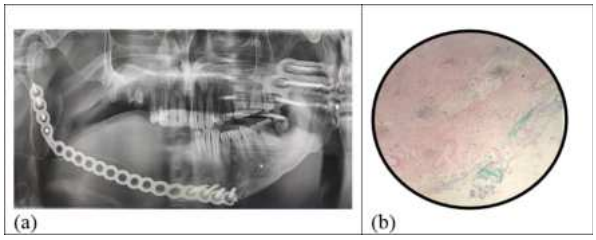


Fig 3: *Postoperative :(a) 7 day post operative OPG (b) HP slide of final specimen revealing features of parakeratinized OKC with ameloblastic changes*

Final Histopathology report revealed it to be an inflammed parakeratinized OKC alongwith some features suggesting of ameloblastoma.(Fig 3b)

No complication was observed during the postoperative follow-up. There was no problem in the functional jaw movements and sensory functions of the patient.

DISCUSSION

The treatment approach for OKCs remains a subject of debate. As far as we know, existing literature does not provide conclusive evidence to aid surgeons in choosing the most effective treatment options. Clinicians typically rely on their own experience to determine the most suitable course of action. Surgical methods range from conservative to more aggressive approaches.[3]

Enucleation is the most frequently employed method, despite its high recurrence rate.[4] Decompression and marsupialization are utilized as conservative treatments, though some practitioners are hesitant due to the potential for residual cystic tissues to contribute to recurrence. Opting for a more aggressive approach may mitigate this risk.

Recommendations suggest reserving aggressive resection for OKCs that have recurred multiple times or have undergone malignant transformation. Worrall advocates for radical excision in cases where OKCs have cortically perforated[5], while Gosau et al. suggest that enucleation coupled with curettage using carnoy's solution yields a recurrence rate comparable to resection excision.[6] Jackson et al. stress the importance of complete excision for OKCs with any indication of soft tissue involvement.[7] Consequently, decision-making regarding OKC resection varies widely. While radical resection boasts the highest cure rate, it also carries significant morbidity, such as jaw continuity loss or facial disfigurement. Thus, assessing patients' post-aggressive treatment is imperative.

The decision to pursue radical resection was primarily influenced by several factors, including multiple recurrent lesions, primary OKCs exhibiting aggressive clinical characteristics such as perforation of the jaw's cortical plates or infiltration into neighbouring tissues, extensive lesions, and localization in the mandibular molar-ramus region.

The occurrence of ameloblastomatous transformation originating from an OKC is uncommon, leading to these lesions being termed combined or hybrid odontogenic lesions. A case report documented histopathological findings indicating an ameloblastoma-like lining epithelium within certain cystic structures, with the treatment approach involving en bloc resection.[8]

The occurrence of an ameloblastoma or its progression into malignancy originating from the epithelial cells of odontogenic keratocysts (OKCs) is exceptionally uncommon. Ameloblastomatous transformation arising from the epithelium of odontogenic keratocysts may be linked to Nevoid Basal Cell Carcinoma Syndrome (NBCCS), suggesting a higher prevalence of ameloblastoma in patients with NBCCS. Nonetheless, instances of non-syndromic cases have also been documented.[9]

CONCLUSION:

The current case illustrates a male patient who experienced delayed developmental milestones and was subsequently diagnosed with an OKC featuring lingual perforation. Given the patient's mental condition and the extent of the lesion, resection alongwith coronoidectomy followed by placement of reconstruction plate was selected as the treatment modality. The final histopathological report, which indicated ameloblastic changes in the lining of the OKC, aligns with our treatment protocol as treatment is dictated by more aggressive lesion. The presence of ameloblastoma as one of the lesions shows the importance of proper histopathological assessment after surgery for further management of the patient as there is a higher chance of recurrence and need of extensive surgery like resection in cases of ameloblastoma.

Declaration Of Patient Consent:

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict Of Interests:

The authors declare that there is no conflict of interests regarding the publication of this paper.

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