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Management of Non-Syndromic Parakeratinized Odontogenic Keratocyst with Adjunctive Chemical Cauterization and Peripheral Odentectomy– A Case Report

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INTRODUCTION

Since the middle of the 20th century, the odontogenic keratocyst (OKC) has been one of the maxillofacial region's most contentious pathological phenomena. [1] Since its conception, the OKC has been renowned for its vagueness. It was initially misdiagnosed as a primordial cyst. Later, due to its aggressive clinical behavior and propensity for recurrence, cysts were no longer considered to be a part of its classification and it was given the name intraosseous benign neoplasm, keratocystic odontogenic tumor (KCOT). [2] Ironically, there have been accounts of OKC responding to marsupialization, and as a result, the benign intraosseous neoplasm, KCOT, was reclassified as a cystic lesion in the WHO's revised classification, which was released in 2017[3]. We still do not agree on the behavior of this mysterious entity despite our comprehensive research. The mandibular angle is where around half of all keratocysts are found. They can expand in different directions into the ascending ramus and onward into the body. Until the cysts grew to a great size and included the maxillary sinus and the entire ascending ramus, including the condylar and coronoid processes, patients are frequently amazingly symptom-free. [4] Its unique palisaded pattern and uniform nuclei are visible in the basal cell layer.

Its typical histological highlights include a thin parakeratinized stratified squamous epithelium, around 5 to 8 cells thick, secured by a thin ridged layer of parakeratin. [6] The daughter cysts that are created when the basal layer buddings into the surrounding connective tissue is a significant characteristic of OKCs. [7] The fibrous cyst wall is typically not infiltrated with inflammatory cells and is rather thin. [6] Recurrence rates ranging from 0% to 100% have been documented. [8] It is thought that these dramatic disparities are related to the length of postoperative follow-up intervals, the surgical techniques used, or the inclusion of cases with nevoid basal cell carcinoma disease. [9] Treatment plans have been developed based on molecular research and thorough literature reviews that propose the modalities with the lowest risk of recurrence. In their comprehensive evaluation of 14 studies, Blanas et al. [10] discovered a recurrence rate of roughly 17% to 56% when treated by simple enucleation. After enucleation, they've also recommended adding Carnoy's solution for three minutes. Because of this, the recurrence was decreased to 1.6%, which is comparable to resection but without the associated morbidity. On the basis of OKC's behavioural pattern, Stoelinga[11] has also suggested a therapeutic plan.

In locations where the cyst was linked to soft tissues, he advised applying electro coagulation or Carnoy's solution, as well as carefully enucleating the cyst and removing any covering mucosa from such areas. With an average of 2.9 years of follow-up, Pogrel[12] recommended marsupialization alone as the only therapeutic option for OKC. Even teeth eruption and uprighting were discovered in the cyst. Immunohistochemistry data of increased interleukin-1 alpha levels in OKC that drastically decreased following marsupialization served as his justification for this. [13] Histological samples taken following marsupialization in all of his instances revealed normal epithelium without daughter cysts, remnants, or basal epithelial layer budding. He discovered that preoperative bcl-2 protein expression was strictly

restricted to the basal layer and that postoperative normal oral mucosa specimens were bcl-2 negative. [12] The classic work of Pogrel [12], who had shown that marsupialization might be thought of as a final therapy technique for OKCs, had an impact on the path of management in our case.

CASE REPORT

A 25-year-old male visited the Department of Dentistry with the complaint of pain on chewing and swelling on the right side of the face. Medical history was not contributory. On intraoral examination, the only positive finding was a grossly carious, tender, right mandibular first molar. Radiographic examination revealed an extensive, multilocular radiolucency bounded by a radiopaque (sclerotic) margin all around. Figure 1 shows radiolucency extended across the whole angle of mandible on right side with complete resorption of lower border. After thorough examination of the case the patient was diagnosed as having an infection involving the right buccal and submandibular space, secondary to a carious right mandibular first molar, along with a panmandibular cystic lesion, most probably an OKC. A provisional diagnosis was made as OKC due to presence of keratin on aspiration.

The submandibular and buccal space infections were treated surgically and the offending tooth was extracted. Incisional biopsy was taken from the extraction wound and the diagnosis was confirmed as nonsyndromic parakeratinized OKC. Marsupialization was considered as an appropriate treatment plan, as the lesion was quite extensive. So, bone windows were made by excising mucoperiosteum along with the bone in the left mandibular buccal vestibule and anterior mandibular labial vestibule. Since the extraction socket of mandibular right first molar provided a potential means of irrigation of the cystic cavity, it was enlarged by removal of interradicular bone by means of bone rongeur and rotary instruments with copious saline irrigation. The cystic contents were evacuated, and the cystic cavity was packed with tape gauze soaked (and squeezed) in 2% povidone-iodine for 3 days. This was followed by periodic irrigation and suction of the cystic cavity with 2% povidone-iodine and normal saline (1:1 proportion). The irrigation of the cystic cavity was initially done every alternate day for 15 days, then twice weekly for about 4 months followed by weekly irrigation. Orthopantomograms were taken at regular intervals to monitor the progress. Histopathological examination was carried out from the base of the resolved lesion in the anterior mandibular area.

The pretreatment biopsy specimen and the lesional tissue from the base of the resolved lesions were subjected to immunohistochemical analysis to assess the expression of Ki-67, a proliferative marker, and bcl-2, an antiapoptotic marker (BioGenex reagent and Super Sensitive polymer horseradish peroxidase kit Sigma Aldrich, Germany). The preoperative radiograph revealed that the cystic lesion involved whole of the angle region of mandible on right side. Progressive reduction in the radiolucency was noted in successive radiographs taken. The radiographs taken at an interval of 4 months after marsupialization depicted gradual resolution of the lesion (as evidenced by a radiographic reduction in the radiolucency accompanied simultaneously by replacement with radiopaque bone), except for a small area in the mandibular right quadrant (4th quadrant). Clinically the bone windows have largely filled up from within, leaving a 1.5cm depth. The immune-histochemical report of the pretreatment biopsy specimen showed the characteristic parakeratinized epithelium of OKC with an abundance of Ki-67 and bcl-2 in the basal and suprabasal layers. So the area was curetted out and chemical cauterization was done with Carnoy's solution under local anesthesia. Regular follow-up revealed total uneventful bone healing with no evidence of recurrence or new lesion.

DISCUSSION

Based on extensive research, recommended treatment modalities for OKC that are known to reduce/prevent recurrence include enucleation, excision of overlying mucosa followed by application of Carnoy's solution,[15] marsupialization [16]/decompression followed by cystectomy, [17] and mandibular resection. [18] However, in our case enucleation had the possibility of pathologic jaw fracture, whereas cystectomy and resection had their own share of morbidity including functional, psychologic, cosmetic, and financial implications. Marsupialization as a potential treatment modality for parakeratinized OKC has received a great deal of attention after the research work of Pogrel [12]; however, the findings of this case suggests otherwise. The reduced expression of Ki-67 after marsupialization indicate the reduced proliferative activity and potential for recurrence and that of bcl-2 indicates the conversion of the classic OKC epithelium to the stratified squamous epithelium. The bcl-2 gene, located at chromosome 18q21, is characterized by its ability to stop apoptosis (programmed cell death) without promoting cell proliferation. [19] Bcl-2 inhibits apoptosis to facilitate cellular proliferation in the basal and suprabasal layers, whereas apoptosis maintains the homeostasis of the thickness of the lining epithelium and allows the synthesis of large amounts of keratin in the surface layer of OKCs. There is a regulated balance between cell proliferation, cell differentiation, and cell death in this type of lesion, this may explain why OKCs do not transform into tumor masses, instead of having neoplastic behavior with an increased potential to proliferate. [19] Ki-67 antigen is more specific marker of proliferating cells, maximally expressed during S-

phase. The levels of Ki-67 in the epithelial linings were found to be double in syndrome-related OKCs as compared to the sporadic cysts, indicating a greater level of proliferative activity in the former. This was reflected in the multiplicity of cysts and the increased numbers of satellite cysts and epithelial islands in the OKCs of NBCCS patients. This suggested that it was a genetic factor, possibly related to defective tumor suppression functions, that was reflected by the higher proliferative activity in their epithelial linings.[20] It was also observed that Ki-67 expression was found to be more common. Postoperative radiograph at a 10-year follow-up showing complete resolution of the lesion as evidenced by replacement of radiolucency by radiopaque bone.

CONCLUSION

The authors claim that marsupialization must be viewed as the first line of management, particularly for an OKC of such a large size, but it cannot be viewed as a final form of treatment for OKC. When the lesions are not responding, however, more invasive surgical procedures including removing the cortical plate and the mucoperiosteum that lies on top of it should be taken into consideration. Our example furthers the paradox that, despite OKC being classed as cystic tumors by WHO[3] in 2017, these tumors respond to straightforward procedures like marsupialization and enucleation with a variety of adjuvant techniques such peripheral ostectomy, chemical cauterization, and cryosurgery.

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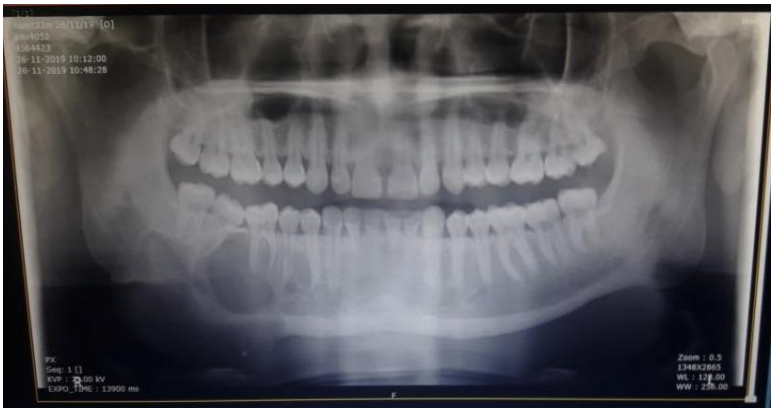


Figure 1:

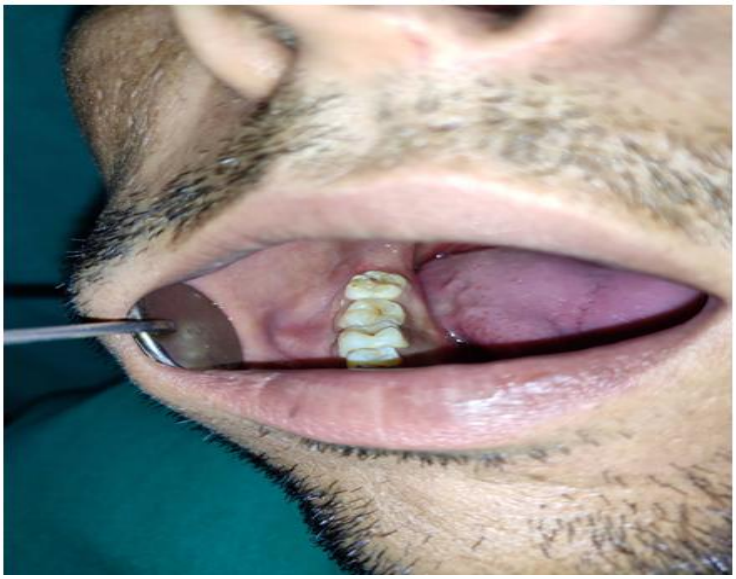
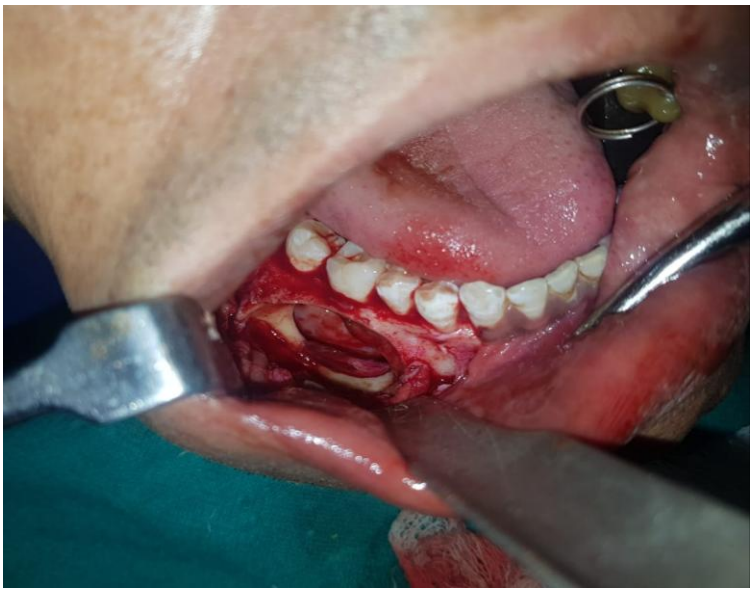


Figure 2



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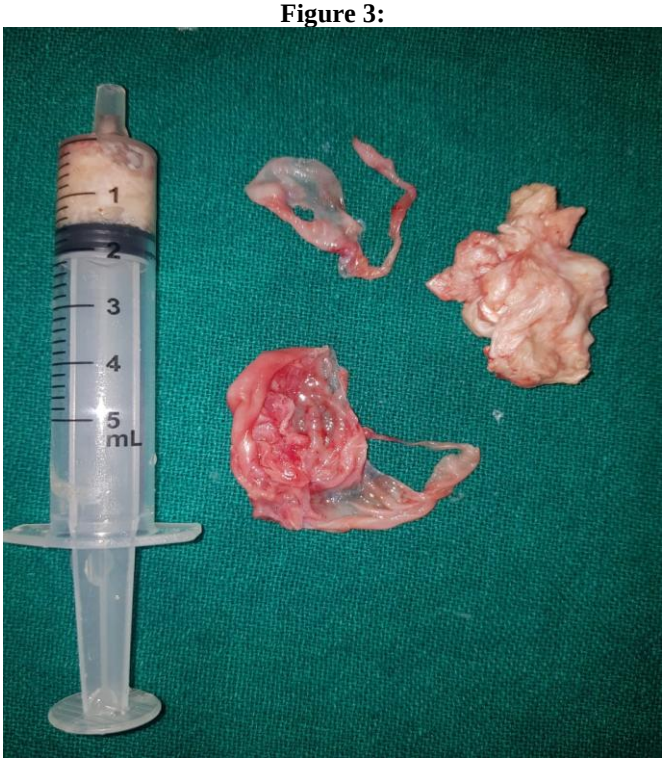

Figure 4: