Oral & Maxillofacial Surgery Keratocystic Odontogenic Tumour: An Incidental Finding

B. Gunachandra Rai¹, Ashwin K Harekal², Niti Sarawgi³

¹Professor & MDS in Department of Oral & Maxillofacial Surgery, Yenepoya Dental College, Mangalore, Karnataka, India
²MDS 3rd year Post Graduate student in the Department of Oral & Maxillofacial Surgery, Yenepoya Dental College, Mangalore, Karnataka, India
³MDS 3rd year Post Graduate student in the Department of Oral & Maxillofacial Surgery, Yenepoya Dental College, Mangalore, Karnataka, India

Running title: Keratocystic Odontogenic Tumour: An incidental finding

Abstract: Odontogenic Keratocyst is the second most common cyst after radicular cyst. In the year 2005, keratocysticodontogenic tumor was coined by WHO owing to its inclination more towards tumor than cyst. It was necessitated due to the aggressive behaviour of the lesion also associated with genetic and chromosomal variations consistent with progression towards neoplastic changes. In this case report, we present a young patient with KCOT involving a large section of mandible (extending from 33 to 46). Any aggressive approach would have ensured extensive morbidity. Hence, we went for a conservative approach of enucleation with peripheral ostectomy and apicectomy of involved teeth followed by placement of Carnoy’s solution. This ensured no remnants of satellite cells. The patient has been on regular follow-ups with normal healing and no signs of recurrence.

1. Introduction

Odontogenic keratocyst is the 2nd most common cyst of the oral cavity after radicular cyst. Philipsen described Odontogenic keratocyst in the year 1956. Its occurrence can be associated to cell rests of dental lamina. Being an aggressive cyst with a rapid growth rate it has a tendency to invade surrounding structures including the bone.

OKC has a high recurrence rate due to the presence of satellite or daughter cysts. Most common site of occurrence being posterior part of mandible. The clinical features generally donot co-relate with its radiographic presentation. The cyst can be associated with non-vital tooth that owes to its late diagnosis due to lack of clinical symptoms.

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KCOT clinically presents as a single lesion, asymptomatic, discovered during routine radiographic examination. If the lesion becomes gargantuan it may cause root resorption, poor positioning of teeth, cortical plate expansion causing swelling and intra-oral drainage of the cystic content.

Radiographically. It presents as a unilocular or multilocular cyst with well-defined scalloped margins.³

2. Case Report

A 30-year-old male presented to the Department of Oral and Maxillofacial Surgery, Yenepoya Dental College with the chief complaint of intermittent sharp shooting pain and pus discharge from the lower jaw since past 2 months.

On extra-oral examination there were no relevant findings. On intra oral examination a draining sinus opening was seen in the buccal aspect of 44. There were no associated caries or dental infections. There was no expansion of cortical plates. No associated intra-oral or extra-oral swelling was seen.

On intra-oral palpation, there was pain and discharge from the sinus.

Figure 1: Intra-oral Pre –operative photo of lingual side of mandible

Figure 2: Intra-oral pre operative photo of buccal side of mandible
An Ortho-pantomogram was advised, which revealed a well-defined radiolucency with scalloped margins of about (9cmX2.5cm) extending from the distal aspect of 33 to the distal aspect of 46.

**Figure 3: OPG showing radiolucency extending from distal of 33 to distal of 46**

**Pre-operative preparation**
A conservative treatment was planned. Root-canal treatment was done for 9 teeth from 33 to 46. In which, the final obturation was done one-day before the surgery.

**3. Intra-operative**

Patient was prepared under aseptic conditions with standard asepsis protocol. Nasal intubation was done for the patient. An intra-oral cervicircular incision was placed from 33 to 46. Two-vertical relieving incisions were placed on either end. Flap was reflected. A window was made to expose the cyst. Enucleation of the cyst was done followed by apicectomy of all 9 teeth and peripheral ostectomy of about 2 mm. A retrograde- filling of 44 was done with MTA – retrograde filler. Chemical cauterization with freshly prepared carnosy’s solution was done to attack any remaining daughter cysts. Time of contact was kept as 4 minutes to prevent any permanent damage to inferior alveolar nerve. It was further irrigated with betadine-saline. The cavity was checked for any remnants of the lining. The flap was placed back and sling-sutures were placed for closure using 3-0 vicryl.

**Figure 4: intra-operative post enucleation of cyst**

Immediate post-operative examination showed minimal parasthesia on the right side and no parasthesia on left side.

A Histopathological examination was undertaken on the enucleated cystic lining and was concluded to be Infected Keratocystic Odontogenic Tumour.

Histopathology Report:
Histopathological examination of H & E stained section showed cystic epithelial lining and soft tissue capsule. Epithelial lining was of stratified squamous parakeratinised type. Certain areas showing rete-peg formation, squamous metaplasia and keratin-pearl like structures.

Further, patient was advised soft-diet and monthly follow-up.

**4. Discussion**

Here, a case of incidental diagnosis of keratocysticodontogenic tumour of anterior mandible has been presented with minimal symptoms contrary to its actual radiographical presentation.

Keratocystic Odontogenic Tumor, which was known as odontogentickeratocyst initially, got its name so as to put emphasis on its neoplastic behavior. Inspite of being a benign developmental odontogenic tumor, it has many distinguishable clinical and histopathological features like local destruction and high recurrence rate. It can be associated with syndromes such as Nevoid Basal Cell carcinoma syndrome or Gorlin syndrome.

Keratocystic Odontogenic Tumor represents between 4-12% of Odontogenic cysts and is believed to originate from dental lamina. It has a male predilection and peaks around second and third decades. The mandible is more commonly involved than maxilla (65%- 83% of cases) with the most commonly seen area being angle of mandible. Even though, Keratocystic Odontogenic tumors are regarded as intraosseous lesions, peripheral involvement have also been reported like the gingiva or alveolar mucosa in the canine-premolar region.
Most of the time KCOTs are incidental findings during routine examinations. Rarely patient complains of swelling, pain, drainage or mobility of teeth except in case of secondary infection. Radiographic studies show it as unilocular or multilocular radioluencies with well-defined or scalloped margins. Sometimes it may mimic dentigerous cyst, if it is associated with impacted teeth.

Various treatment modalities have been considered for the treatment of KCOT such as decompression, marsupialization, enucleation with or without application of Carnoy’s solution and peripheral osteotomy. According to recent literature a much conservative approach that is enucleation with application of Carnoy’s with regular follow-ups can be advocated. This can specially be used in case of large lesions and patients’ age being less to go with aggressive modalities that can cause a disruption in the continuity of jaw.1

The treatment plan depends upon various factors such as patient age, size of lesion, location of lesion, soft tissue involvement, and previous treatment history and histological variant of the lesion. The main objective of the treatment modality is to have least morbidity and lowest risk of recurrence.3

The various modalities are as follows:

- **Marsupialization and Decompression**
  The main objective of this modality is to relieve pressure within the cyst and enable its size to decrease. Decompression can be done by making an opening in the cyst and keeping it patent with a drain. Marsupialization involves opening up the cyst to the oral cavity and suturing the cyst lining to the mucosa, creating a permanent opening of the cyst into the oral cavity. 
  
  Advantages: Allows decrease in size of cyst preserving vital structures like teeth or inferior alveolar nerve.

Disadvantages: As this technique does not remove the cystic lining completely, can lead to continued epithelial proliferation and increase recurrence rate.

- **Enucleation with peripheral ostectomy**
  Owing to its high recurrence rate, simple enucleation is not enough as it leaves behind daughter cysts, hence removal of surrounding 1–2 mm of bone beyond the visible margin is believed to be very effective in reducing the recurrence. Methylene blue or crystal violet stain can be used to mark the amount of bone to be removed.

  Advantages: reduced recurrence rate

  **Enucleation with chemical treatment with Carnoy’s solution**
  Average depth of bone penetration depends on duration of application (1.54 mm after 5 mins).5 Carnoy’s solution is a caustic tissue fixative. It is neurotoxic and chemically fixes the inferior alveolar nerve or lingual nerve if it comes in contact with them for more than 2 minutes.9 Most authors recommend a freshly prepared solution.

  Advantages: decreases recurrence

  Disadvantages: Neurotoxic - can cause permanent nerve damage if left in contact for more than 4 mins.

- **Physical treatment with cryotherapy using liquid nitrogen:**
  Cryotherapy with liquid nitrogen devitalizes organic tissue beyond the visible margin of the lesion, but leaves the inorganic bony framework intact. It devitalizes an area between 1 to 2 mm at a temperature of -20°C. Cryotherapy destroys cells by means of direct damage to the intracellular and extracellular surfaces because of formation of ice crystals that affect the osmotic and electrolytic balance.10

  **Advantages:** clean scaffold for bone formation, bone graft can be placed immediately, decreased intra-operative bleeding and post-operative swelling.

  **Disadvantages:** necrosis of healthy tissues.

- **Resection**
  It is an aggressive modality and results in considerable morbidity.

  **Disadvantages:** Morbidity

  Thus, there are no absolute guidelines for the treatment of Keratocysticodontogenic tumor as it is inclined more towards tumor than cyst. There are various factors upon which the treatment modality depends.

  In this case, since it was a young patient conservative approach was preferred. Enucleation and peripheral ostectomy with application of Carnoy’s solution was considered the most effective. The patient has been on regular follow up for 6 months now and shows normal healing and no signs of recurrence.

**References**

