Novel treatment (new drug/intervention; established drug/procedure in new situation)

Use of decompression tubes in the management of excessively large odontogenic keratocyst

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Summary

Odontogenic keratocyst (OKC) is a unique cyst because of its locally aggressive behaviour, high recurrence rate and characteristic histological appearance. A radical surgical approach is commonly advocated with morbid and disfiguring results. This approach also presents several reconstructive obstacles especially in anterior mandible. In this case report the authors present a 25-year-old male patient with a large OKC and treatment with decompression followed by enucleation, and chemical cauterisation. This approach though demanding prolonged treatment time and postoperative follow-up decreases the morbidity to a great extent in a young individual. At the same time, it allows an opportunity for the maxillofacial surgeon to preserve the natural dentition, maintain function and safeguard cosmesis. In our case, the authors effectively achieved all the aforementioned objectives. At the same time a radical option of treatment is still left in the armoury if required at a later date.

BACKGROUND

The age old controversy regarding effective management of odontogenic keratocyst (OKC) still exists in spite of current advances in surgery. The inherent property of an OKC for aggressive behaviour and high recurrence has always posed a problem for the maxillofacial surgeon treating it. Several treatment modalities have been proposed but none of them have proven to be foolproof. A radical approach proposed by few involving wide local excision followed by microvascular reconstruction is associated with several comorbidities. Overtly conservative approach on the other hand may heighten the chances of recurrence. We hence came up with a combination of both approaches in an attempt to reduce the morbidity associated with the radical approach as well as the recurrence rate after the conservative one. This technique may prove invaluable in the management of patients on either extremes of age.

CASE PRESENTATION

A 25-year-old male reported to our outpatient department with a chief complaint of swelling in the lower jaw since 1 year. The patient had already undergone incisional biopsy at another maxillofacial centre and on the diagnosis of OKC had been advised mandibular resection and reconstruction using microvascular fibula graft. The biopsy was repeated and the diagnosis was confirmed. The radiograph (orthopantomogram (OPG)) showed a multilocular cystic lesion extending from angle of the mandible on one side to the angle on the other side (figure 1). Associated with the lesion was an impacted canine displaced to the inferior border of the mandible in the left parasymphseal region. The lesion appeared to have involved the inferior border of the mandible, making it impossible to save the same at this stage.

INVESTIGATIONS

Serial OPG demonstrating the presence of a large OKC and subsequent decrease in size.

TREATMENT

Considering the patient's age and the morbidity of mandibular resection and reconstruction into account, it was decided to adopt a novel approach that combined an age-old technique with one of the latest technical advances in dentistry.

At the site of the repeat biopsy that is, left premolar region, a bony window (approximately 5 mm in diameter) was left patent at the depth of the buccal sulcus after extraction of the impacted canine. The retained deciduous canine in the arch was also extracted. An indigenously fabricated decompression tube fashioned out of a needle

Figure 1 Orthopantomogram showing large multilocular radiolucent lesion with extent from angle of mandible on one side to angle of mandible on the other side with impacted canine.

cap was inserted through this and secured with sutures. Another tube was placed through a bony window in the right premolar region (figure 2). The patient was instructed in self irrigation of the cavity and oral hygiene maintenance (figures 3–5).

The patient was followed up monthly with sequential OPG investigation. The lesion showed radiographic evidence of decompression followed by decrease in size. Surprisingly, over the following 8 months there was also an evidence of restoration of normal anatomical structures like inferior alveolar canal and inferior border of the mandible.

Along with this treatment, the patient underwent endodontic therapy of all teeth except third molars in the mandibular arch (figures 6–10).

At the end of 8 months, the cyst was enucleated through an intraoral approach. Following enucleation, the residual bone was immediately cauterised with Carnoy's solution (figure 11). Apical sealing of all endodontically treated teeth was achieved. Primary closure was done.

Postoperative recovery was uneventful (figure 12).

Figure 2 Decompression tubes fashioned out of needle covers inserted and fixed on either side.

Figure 3 Surgical stent fabricated to cover the decompression tubes and prevent contamination of bony cavity.

Monthly follow-up was resumed and the evident bone regeneration monitored.

OUTCOME AND FOLLOW-UP

Over the next 1 year the bone showed no evidence of recurrence of the cyst. On the contrary it showed regeneration attaining almost premorbid alveolar height.

The patient was followed up every 6 months to rule out recurrence of cyst. Three year follow-up of the patient till date has shown no incidence of the same (figure 13).

DISCUSSION

OKC continues to be an enigma for effective management. Several schools of thought have been proposed for its effective management but none has proven to be fool-proof. In managing such pathology, emphasis has to be laid

Figure 4 Stent covering both the decompression tubes in place.

Figure 5 Irrigation being carried out through the tubes.

Figure 6 Orthopantomogram after 1 month of decompression and self irrigation.

Figure 7 Orthopantomogram taken after completion of 2 months of decompression and self irrigation.

Figure 8 Orthopantomogram at the end of 4 months postdecompression and self irrigation showing evidence of inferior border recortication.

on elimination of diseased tissue, prevention of recurrence, minimal morbidity and restoration of function and aesthetics.¹ In an endeavour to achieve the above goals, we used a technique described as the Waldron's procedure where a large cyst was first decompressed to allow for cessation of its expansion followed by reduction in size.² Once the

Figure 9 Orthopantomogram at the end of 6 months shows sizeable lingual cortex, thick inferior border and restoration of inferior alveolar canal.

Figure 10 Orthopantomogram showing good bone deposition in all dimensions at the end of 8 months.

Figure 11 Enucleation followed by peripheral osteotomy with chemical cauterisation and apical sealing undertaken at the end of 8 months.

size was reduced to acceptable levels, it was enucleated and the residual bone was treated with a tissue fixative viz. Carnoy's solution. The bone showed good regeneration along with the restoration of size and texture. It was successfully used to anchor endodontically treated teeth that enabled effective preservation of function. It is our opinion that this technique can be used in patients with

Figure 12 Orthopantomogram immediately postoperatively with restored teeth.

Figure 13 Orthopantomogram at the end of 3 years after second surgery showing no evidence of recurrence.

large OKCs at either extremes of age where morbidity of additional donor site, operative time, extraoral scar and reliable bone available for implant placement are defining factors. Moreover, this technique can also be used as a first line of treatment where a radical procedure can be kept as a probable option in cases with recurrence.

Why our technique?

By relieving the pressure within the cystic cavity, one can achieve the following results:

1. Decrease in lesion size due to fall in interleukin-1 level that plays a crucial role in the expansion of keratocysts.³

2. After decompression and marsupialisation, the cyst lining eventually gets replaced by normal oral epithelium and anatomical structures.⁴
3. Minimum morbidity during second surgery.
4. There is an evidence based theory which suggests that keratocyst lining is transformed into non-keratinising epithelium.⁵

Drawbacks of our technique:

- Compliance of the patient.
- Difficult transoral access to the ascending ramus and subcondylar region.
- Limited sample of tissue for initial H/P examination.

Solution:

- Multiple biopsies.
- Lifelong follow-up.

Learning points

- Large multilocular keratocysts can be treated with a conservative approach.
- The only disadvantage being the extended therapeutic time.
- Extensive resection of the mandible with its resultant morbidity can be prevented.
- Radical surgery for OKCs should be, of course, reserved for those cysts that have undergone malignant changes.
- In any case, clinical and radiographic follow-up is mandatory for years after surgery, because recurrence of this lesion may occur even several years later.

Competing interests None.

Patient consent Obtained.

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