Surgical Management of Recurrent Odontogenic Keratocyst

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Abstract The odontogenic keratocyst is a distinct entity arising from odontogenic epithelium. The unique histopathological appearance and the propensity for recurrence has made its management controversial in terms of the conservatism to be followed. This article describes the management of a case of recurrent odontogenic keratocyst with an aggressive therapy being adopted due to the nature of the pathology.

Keywords Keratocyst · Recurrence · Carnoy's solution

Introduction

The term 'odontogenic keratocyst' (OKC) was introduced in 1956 by Philipsen. The odontogenic keratocyst is a distinctive developmental odontogenic cyst typically occurring in the jaws, with a predilection for the posterior body and ascending ramus of the mandible. The odontogenic keratocyst is considered to be distinctive not only because of its unique histopathologic appearance but also because of its potential for local recurrence and association with the nevoid basal cell carcinoma syndrome. Two distinct varieties have been described histologically

orthokeratinized and parakeratinized. The orthokeratinized variant has a limited growth potential, low recurrence rate and no association with nevoid basal cell carcinoma syndrome and therefore should be distinguished from the parakeratinized variant. The treatment of the OKC remains controversial. Treatment is generally classified as conservative or aggressive. Conservative treatment generally includes simple enucleation, with or without curettage or marsupialization. Aggressive treatment generally includes peripheral osteotomy along with chemical curettage with Carnoy's solution or resection. Some surgeons believe that the cyst can be properly treated with enucleation if the lesion is removed intact. However, complete removal of the OKC can be difficult because of the thin friable epithelial lining, limited surgical access, danger of cortical perforation, and the desire to pre-serve adjacent vital structures.

Case Report

A 29-year-old patient reported to the Department of Maxillofacial Surgery, AFMC Pune with chief complaint of painless swelling in the left midface along with difficulty in opening mouth. History of present illness revealed that the patient had experienced difficulty in mouth opening and pain at the pre auricular region in November 1995 for which he reported to a hospital at Kolkata, where he was diagnosed as a case of OKC left side of mandible after an incisional biopsy from the left ramal region. He was then operated for surgical removal of cyst by enucleation using an intraoral approach. The patient was asymptomatic for 7 years; in 2002 he experienced sudden pain in preauricular region and was operated again for the recurrence of cystic lesion of left side of mandible.

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Fig. 1 Showing preoperative tomogram of the patient

Personal history revealed absence of any deleterious oral habits. On general physical examination, the patient was moderately built and nourished, with normal gait and posture and well oriented to time and place. Extra-oral examination revealed an obvious facial asymmetry due to the presence of a 3 cm × 5 cm single, diffuse, ovoid swelling in the left pre-auricular region extending from tragus to 3 cm behind left zygomatic prominence and superior attachment of ear to lower border of mandible. Mouth opening was 26 mm with deviation of chin to the left on opening. On palpation swelling was soft non tender and fluctuant; no sensory neural deficit was present. Condylar movements were palpable bilaterally. Intra-oral examination revealed the presence of a 3 cm × 2 cm soft, sessile tender, fluctuant swelling around anterior border of ramus. No discharging sinus was seen intraorally. Occlusion was stable but dental midline had shifted to the right side. Biopsy of the lesion was done which confirmed the diagnosis of OKC. Orthopantomogram and CT scan were taken for localizing the lesion and to assess its precise extent and formulating the treatment plan. Orthopantomogram (Fig. 1) showed a multilocular radiolucency involving the ramus of mandible extending from the left angle to coronoid and condylar process. CT scan revealed a well defined expansile cystic lesion in the left ramus of the mandible measuring 22 × 29 × 35 mm in size, thinning both the lingual and buccal cortices with no relation to any tooth.

The lesion being a recurrent one, surgery was carried out removing 1 cm of healthy margins along with the lesion. This treatment plan required removal of the ramus of the mandible proximal to 38 region along with coronoid and condylar process in toto.

Surgery was performed under general anesthesia, with nasotracheal intubation. Submandibular approach was used to expose the lesion as far as the condyle (Fig. 2). Resection of ascending ramus proximal to 38 along with condylar and coronoid process was carried out using piezoelectric saw and osteotomes. Intermaxillary fixation was done with help of archbars and tie wires followed by

Fig. 2 The lesion perforating buccal cortex

Fig. 3 Postoperative tomogram showing condylar prosthesis in place

adaptation of titanium reconstruction plate with condylar implant in the defect region. This was fixed using two bicortical screws (Fig. 3), since the patient had two prior episodes of recurrence it was decided not to go for primary reconstruction using autogenous bone. Haemostasis was achieved, suction drain was secured in place and closure was done using vicryl and silk sutures. Postoperatively the suction drain was removed after 2 days and postoperative healing was uneventful. Postoperative occlusion was stable. Maximum interincisal opening was 34 mm was achieved.

The patient was subsequently followed up for 2 years at 6 months intervals. Clinically there was no further functional deterioration, radiography there was no evidence of lytic changes in the glenoid fossa or mandible in relation to the reconstruction plate.

Discussion

Recurrence following the surgical treatment of keratocysts of the jaws may present a major problem to the oral surgeon [1]. Recurrence of odontogenic keratocysts has been

attributed to several mechanisms, Woolgar et al. [2] described three different theories. The first involves incomplete removal of the original cyst lining. The second involves growth of a new OKC from small satellite cysts or odontogenic epithelial rests left behind by the surgical treatment. The third involves the development of an unrelated OKC in an adjacent region of the jaws that is interpreted as a recurrence. It has been suggested that recurrence is a consequence of microcysts in the mucosa overlying the recurrent lesions [3]. Attempts have been made to reduce this high recurrence rate by improved surgical techniques, such as removal of adjacent mucosa, smoothening of the osseous wall of the cystic cavity, resection of neighbouring parts of the mandible, tanning of the epithelial lining of the cyst with Carnoy's solution and marsupialization [4–6].

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