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## CASE REPORT

### Surgical Management of Mandibular Recurrent Odontogenic Keratocyst- A Case Report

Sukhvinder Singh Rana<sup>1</sup>, Neera Ohri<sup>2</sup>, Aman Abrol<sup>3</sup>

<sup>1,2</sup>Senior Resident, Department of Dentistry, Dr. Rajendar Prasad Government Medical College (RPGMC) Tanda, Himachal Pradesh, India

<sup>3</sup>M.D.S Department of Dentistry, Dr. Rajendar Prasad Government Medical College (RPGMC) Tanda, Himachal Pradesh, India

#### ABSTRACT:

Odontogenic keratocysts is a developmental odontogenic cyst are known for their propensity to recur after surgical treatment. The unique histopathological appearance and tendency to recurrence has made its management controversial. This article described the management of a case of recurrent odontogenic keratocyst with aggressive approach.

**Key words:** Odontogenic Keratocyst, developmental odontogenic cyst, recurrence.

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**Corresponding author:** Dr. Sukhvinder Singh Rana, Senior Resident, Department of Dentistry, Dr. Rajendar Prasad Government Medical College (RPGMC) Tanda, Himachal Pradesh, India

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#### INTRODUCTION

An odontogenic keratocyst (OKC) is a benign but locally aggressive developmental odontogenic cyst. It was firstly identified and described in 1876 and further it was classified by Phillipsen in 1956<sup>1</sup>.

The reported frequency of OKC has been reported to vary from 3% to 11% of odontogenic cysts.<sup>2</sup> The odontogenic keratocysts is a developmental odontogenic cyst which is believed to arise from cell rests of dental lamina. This type of cysts may be located anywhere in the tooth-bearing portions of the jaws; mostly in the posterior region of mandible and ascending ramus.

These lesion occurs in any age but mostly seen during the 2<sup>nd</sup> to 4<sup>th</sup> decades of life with a slight males predilection.<sup>3</sup> It appear as an unilocular or a multilocular radiolucent area with scalloped margins in radiograph. This lesion is involved with unerupted tooth in 25% to 40% of cases. The OKC is considered as one of the most aggressive odontogenic cysts with high recurrence rate and its tendency to involve the adjacent tissue.<sup>2</sup>

In 1963, Pindborg and Hansen, described seven histological criteria of the odontogenic keratocysts.<sup>4</sup> 1) A thin, uniform lining epithelium with no or very less evidence of rete ridges. 2) Cuboidal or columnar basal layer and often in a palisaded arrangement. 3) Thin spinous cell layer is 8-4 cells in thickness. 4) The spinous cell layer often shows intracellular edema. 5) Keratinization is usually parakeratinized, but it may be orthokeratinized. 6) Thin corrugated layer of keratin. 7) Thin and usually uninflamed fibrous cell.

#### CASE REPORT

A male patient of 52 years old reported to us with chief complaint of pain in right lower side of face and difficulty in mouth opening from last 3 month. History of present illness revealed that the patient had experienced swelling and pain in same region with difficulty of mouth opening in August in 2002 for which he was diagnosed as a case of Odontogenic Keratocyst right side of mandible after an incisional biopsy from left angle region. He was then

operated for surgical removal of cyst by enucleation using an extraoral approach. The patient was asymptomatic for 10 years. In June 2012 he experienced sudden pain in right side of the mandible. The pain was spontaneous, mimicking toothache and localized to the right lower facial region. This was followed by trismus. Pain relieved by analgesic. On extra oral examination, there was tenderness on the angle region of Mandible on right side

Intraoral examination revealed foul odour with missing 26, 27, 47, 48 and alveolar mucosal dehiscence w.r.t 47, 48 region with creamy white discharge. The patient's general condition was good. All hematological tests were within normal limits.

An Orthopantomogram(OPG) shows a large unilocular radiolucent area with scalloped margin involving the body region of mandible extending from distal aspect of lower right premolar up to the entire ramus region except the condylar region and coronoid process.( Fig 1a)



Fig 1a- OPG showing large unilocular radiolucent lesion

Aspiration of cyst revealed white cheesy material. An incisional biopsy were undertaken under local anaesthesia from 47, 48 region and sent for histopathological examination. It was reported as an odontogenic keratocysts, the lining of which was parakeratinized.(Fig.1b)

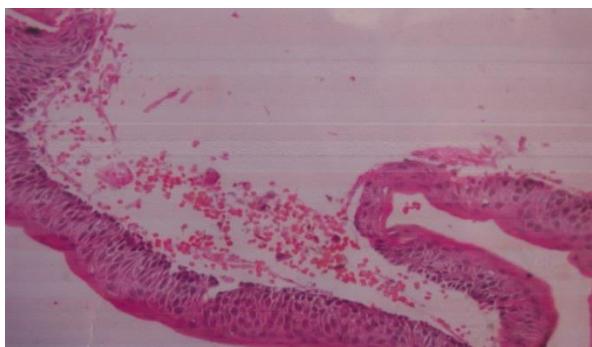


Fig 1b.Photomicrograph of the biopsy specimen that showing palisaded basal cell layer (8-10 cell layer thickness) with polarized stained nuclei, and parakeratinized surface with corrugated appearance.(original magnification 40X)

Computed Tomography (CT) scan was taken for localizing the lesion and to assess its precise extent and formulating the treatment plan.(Fig 1c)

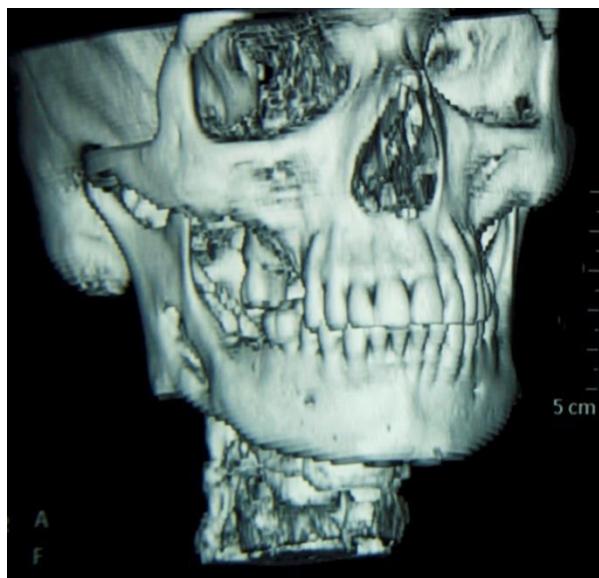


Fig 1c. Computed Tomography (CT) scan

As it was a case of recurrent OKC surgery was carried out to remove the lesion along with one centimeter of healthy bone. This treatment plan required removal of the ramus of the mandible except posterior border of ramus, coronoid process and condylar region with body region distal to 44, in toto. Surgery was performed under general anaesthesia, submandibular approach was used to expose the lesion. Resection of mandible distal to 44 done with the help of bur and osteotome, but the posterior border of ramus, coronoid process and condylar region was preserved.(Fig.1d ) IMF was done with help of arch bars and tie wires followed by adaptation of AO reconstruction plate.(Fig.1e)

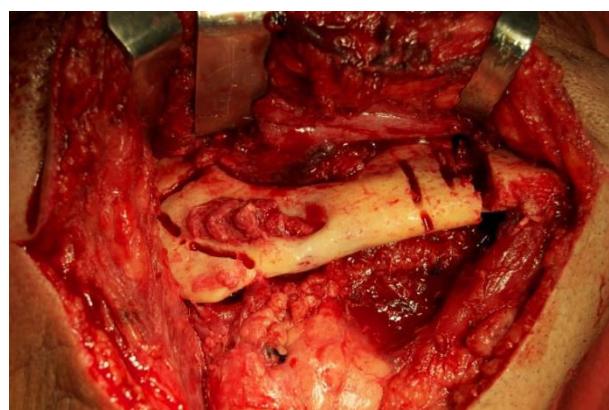


Fig 1d. resection of mandible

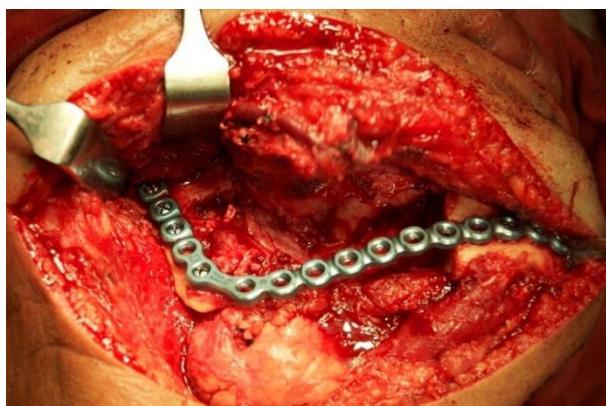


Fig 1e.reconstruction plate placed

## DISCUSSION

The treatment of OKCs remains controversial.<sup>2</sup> Different types of treatments are there to treat OKC both conservative and as well as aggressive. In conservative management we have enucleation of cyst, curettage, marsupialization and decompression. In aggressive management, enucleation with chemical curettage or resection of cyst with or without loss of jaw continuity is treatment of choice.<sup>1</sup> Recurrence potential makes OKC an interesting lesion and so conservative treatment of keratocyst of the jaws may encounter a major problem to the operating surgeon.

The OKC is thought to recur because of incomplete removal/residual cystic lesion gives rise to new cyst formation (microcysts /daughter-cysts/ epithelial islands). The OKC may recur due to epithelial offshoots of the basal layer of the oral epithelium. Continuous recurrence of new OKC is seen in patients with basal cell nevus syndrome.<sup>5</sup> OKCs are characterized by unique and distinct histological features, an aggressive biological behavior, and a high recurrence rate.<sup>6</sup> The rate of recurrence vary enormously, from a maximum of 62% to a minimum of 0%.<sup>6,7,8</sup> Because of the high recurrence, accepted treatment is complete surgical removal of cyst, with meticulous curettage of the adjacent tissues.<sup>8</sup> There are 2 categories of OKC: orthokeratocyst and parakeratocyst.<sup>9</sup> The parakeratinized and orthokeratinized OKCs were significantly different in molecular as well as the recurrence rate. The parakeratinized OKCs had a higher recurrence rate than the orthokeratinized OKCs.<sup>2,9,10</sup> A polarized layer of basal cells with corrugated hypercellular epithelial lining are found in parakeratinized OKCs but not in the orthokeratinized type. So these feature make highly active epithelium in parakeratinized OKCs and may explain the high recurrence rate.<sup>10</sup> Because of this parakeratinized variant of OKCs need more aggressive treatment than simple enucleation has been advised.<sup>11</sup> Recurrence of OKCs of mandibular molar region occurs more often as compare to the other sites.<sup>12,13</sup> OKCs treated with simple conservative approach like enucleation had a significantly higher recurrence rate than those treated with surgical methods.<sup>2</sup>

The lesion described in this report is a parakeratotic type of OKC located on right mandibular molar area recurred after 10 years of enucleation. OKC if do recurred they can be treated with surgical approach reduce or minimize the recurrence rate. Many reputable authors have suggested resection of the jaw with surgical margins free of cystic growth is treatment of choice for OKC.<sup>14</sup>

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