Case Report

Surgical Ciliated Cyst of the Posterior Maxilla in an Old Male, Mimicking Residual Cyst or Odontogenic Keratocyst: A Case Report

Maryam Peighoun[1], Sahand Samieirad [2*], Noushin Mohtasham[2], Elahe Tohidi[2], Shahin Moeini[3]

1. Department of Endodontics, School of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran.
2. Oral and Maxillofacial Diseases Research Center, Mashhad University of medical sciences, Mashhad, Iran.
3. Department of Oral and Maxillofacial Radiology, School of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran.

***Corresponding Author:**

Sahand Samieirad

Oral and maxillofacial Surgery Department, Mashhad Dental School, Mashhad University of Medical Sciences, Mashhad, Iran.

Tel.: +9851-38829501-15
Email: samieerads@mums.ac.ir

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ABSTRACT

Surgical ciliated cysts of the maxilla arise from respiratory epithelium that lodges in the maxilla after radical maxillary or sinus surgery. This case report was conducted in 2020 in Mashhad Dental School, Mashhad University of Medical Science, Mashhad, Iran. We present here an unusual case of surgical ciliated cyst of a 73-year-old adult male patient with a history of nasal polyp surgery 40 years ago. Early diagnosis due to CBCT and Clinical examination was a residual cyst or OKC (odontogenic keratocyst). The histopathology result of this lesion surprised us after the excisional biopsy and enucleation. The clinical examinations and radiographs of the jaw lesions may lead the maxillofacial surgeons to misdiagnosis and mistreatment. Therefore, it is obligatory to notice all aspects of these lesions carefully.

KEYWORDS
Surgical ciliated cyst; Maxilla; Enucleation

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INTRODUCTION

The maxillary cysts with pseudostratified columnar epithelium linings such as respiratory epithelium one may mimic diverse pathologic conditions. These lesions can lead to the delay in the correct diagnosis, thus might challenge both the clinician and pathologist. These lesions include mucocele of the maxillary sinus and surgical ciliated cyst[1].

Surgical ciliated cysts of the maxilla might arise from respiratory epithelium invasion in the maxilla after radical maxillary or maxillary sinus surgery[2]. At first, surgical ciliated cyst was described in 1927 in Japan[3]. This lesion is more frequent in Japanese people; however, it occurs in other races from the age of 5 to 49 years[4,5].

This lesion is rare but locally aggressive and can be a complication after surgery in the maxillary sinus, midface osteotomies, traumatic tooth extraction, orthognathic surgery and maxillary fractures[6]. Surgical ciliated cyst most common in the posterior maxilla[7].

Hereby, we present an unusual case of surgical ciliated cyst of a 73-year-

old adult male patient mimicking residual cyst or OKC, with a history of nasal polyp surgery for 40 years ago.

CASE PRESENTATION

An old man of age of 73 year old, referred to the Oral and Maxillofacial Department of Mashhad Dental School, Mashhad University Of Medical Sciences, Iran, in April 2020. His chief complaint was pain in the left maxilla. The patient had a history of nasal polyp surgery 40 years ago.

In extraoral and intraoral examination, the patient had no lymphadenopathy and swelling. All laboratory test results were normal and he had no history of systemic disease. In clinical examination, neither evidence of swelling nor pus was observed.

CBCT-based findings revealed a cystic-like lesion in the posterior left edentulous maxilla near the maxillary sinus, the same as a residual cyst or an odontogenic keratocyst. Moreover, in the coronal view, destruction of the buccal cortex and thinning of the lingual cortex and the invasion of lesion to the maxillary sinus were obvious (Figure 1).

All procedures performed in this study involving the human participant were following the ethical standards of our institutional Ethics Committee, Mashhad University of Medical Sciences, Mashhad, Iran. Due to the CBCT and clinical examination, our early diagnosis was residual cyst or OKC.

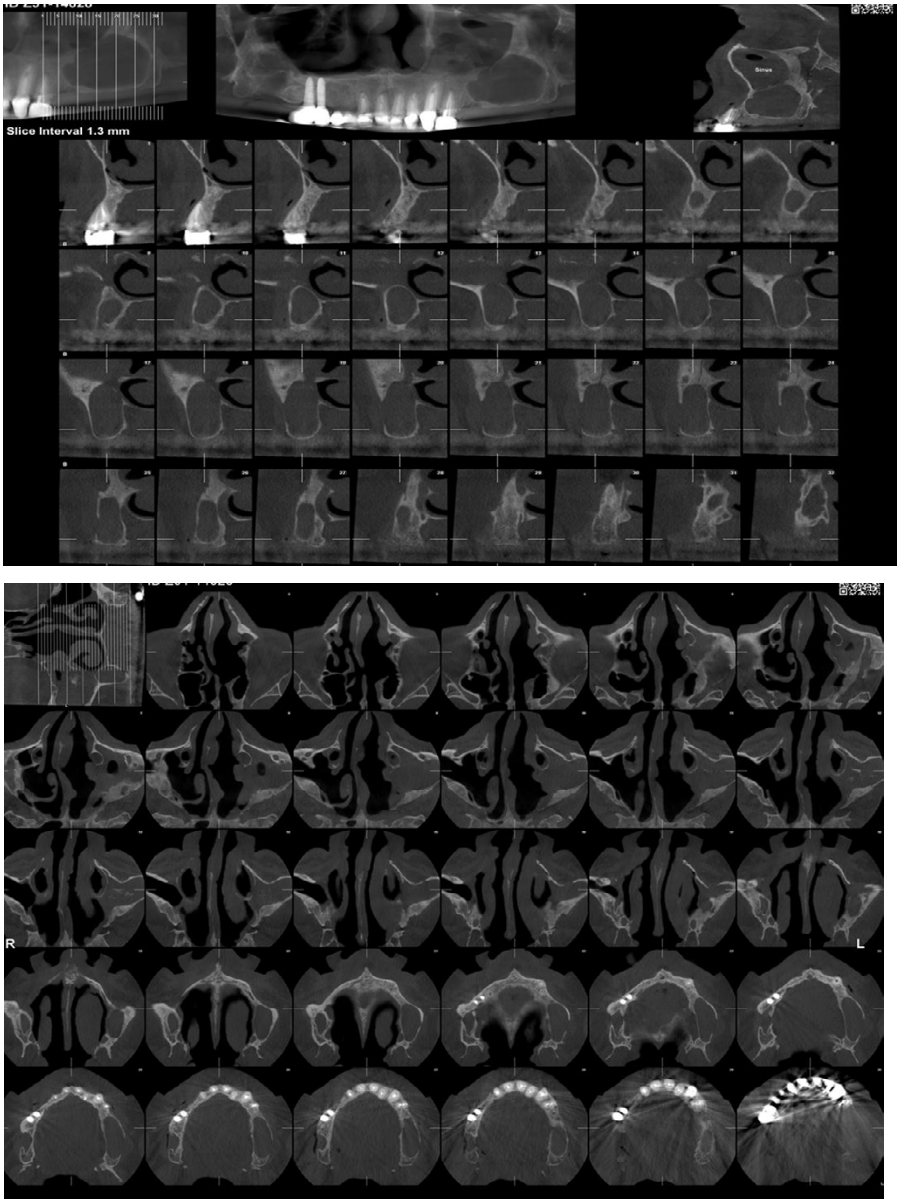

Figure 1: CBCT view showed cystic lesion near the maxillary sinus, with destruction of buccal cortex and thinning of lingual cortex

After obtaining informed consent from the patient, the excisional biopsy was performed to determine the nature of the lesion. In the surgical procedure, the authors decided to enucleate this lesion. The surgeon performed the surgical procedure under local anesthesia (2% lidocaine with 1:100000 epinephrine).

Afterward, an intraoral crestal incision was made in the left maxillary ridge with two vertical incisions in the posterior and anterior region being made to access the lesion (Figure 2).

The cyst was completely removed with a curette from bone. Then the enucleation of the lesion with the peripheral osteotomy was accomplished (Figure 3). The cancellous bone block allograft was placed at the surgical defect to reconstruct the region and prepare for future implants (Figure 4). Finally, the single interrupted sutures with vicryl 3/0 (Supa, Tehran, Iran) was used for watertight and tension-free closure (Figure 5).

The lesion was sent for a pathologic evaluation. In the histopathological feature of our case, the

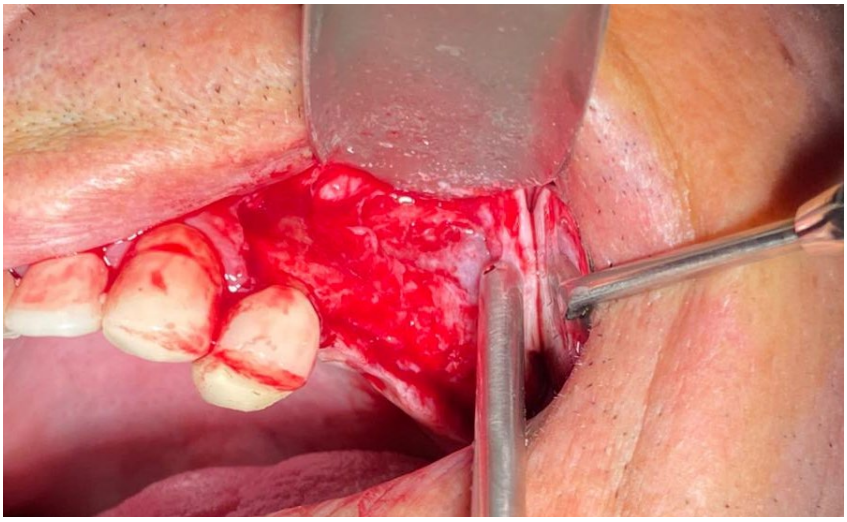

Figure 2: The intraoral incision was made to access the lesion

Figure 3: Enucleation of the lesion

fragments of an inflammatory cystic lesion were observed, covered by a false cylindrical ciliated epithelium. Due to this histopathology feature, the surgical ciliated cyst was confirmed as our definitive diagnosis (Figure 6). The patient had no infection or recurrence in the 6-month follow-ups.

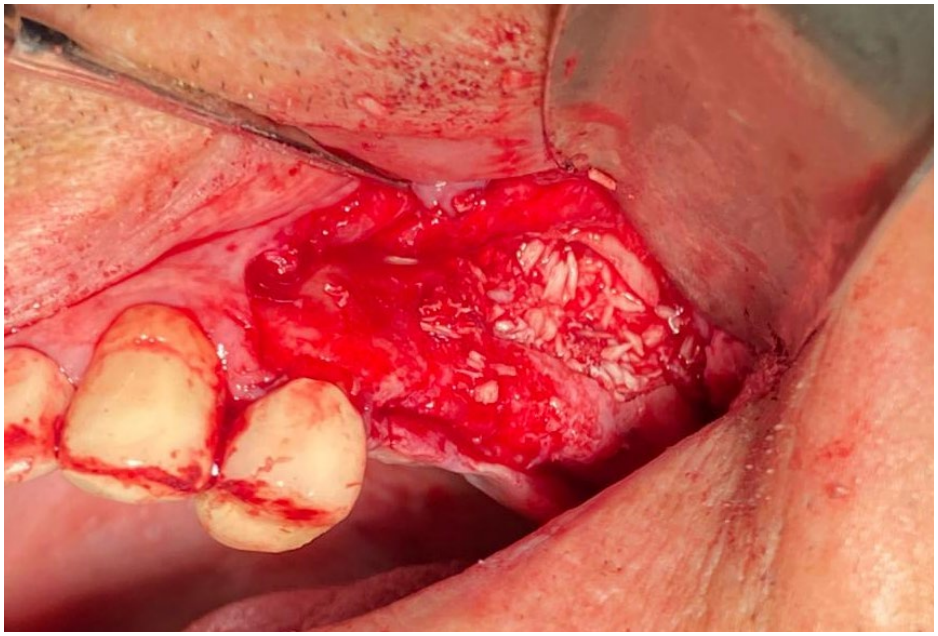

Figure 4: The cancellous bone block allograft was placed at the surgical site to prepare for future implants

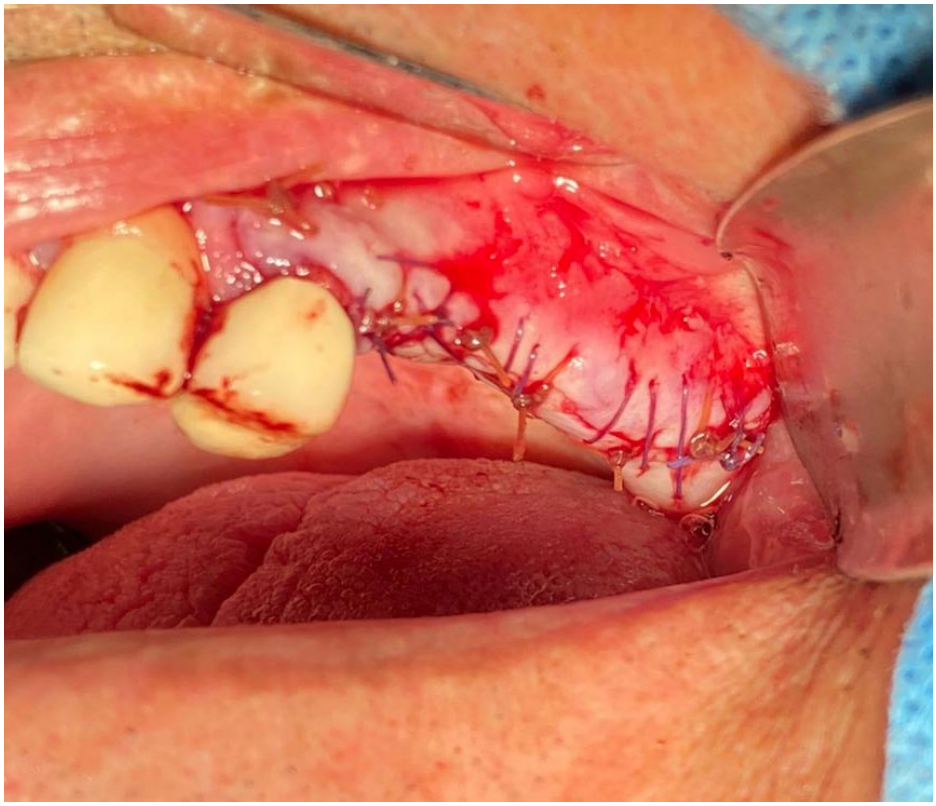

Figure 5: Tension free closure

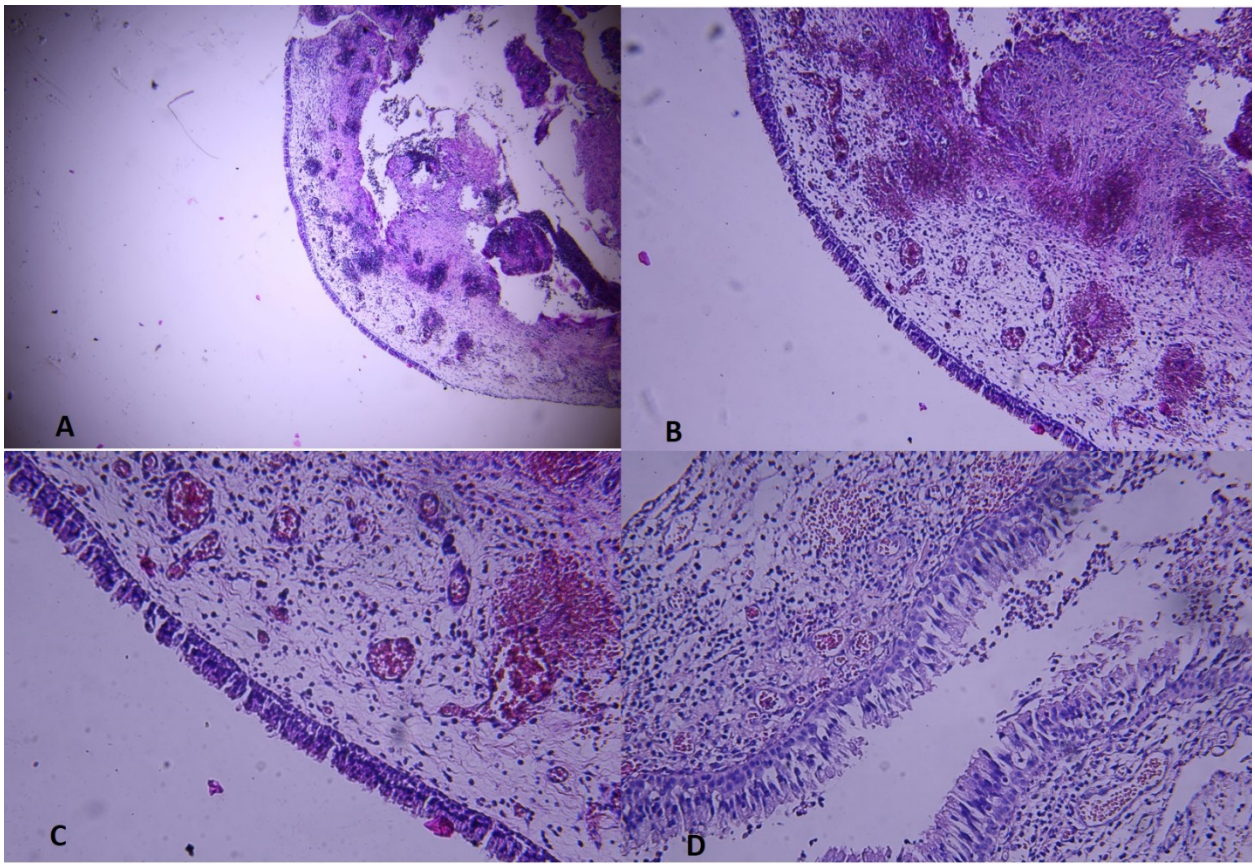

Figure 6: Histopathologic feature (H&E staining). The Fragments of an inflammatory cystic lesion were covered by a false cylindrical ciliated epithelium. A) 40X magnification, B) 100X magnification, C) 200X magnification, and D) 400X magnification view of optical microscope

DISCUSSION

The remarkable point of this case report was that the differential diagnosis was residual cyst or OKC (odontogenic keratocyst) for this case, due to radiographic view and clinical examination. However, the surgical ciliated cyst was surprisingly confirmed as our definitive diagnosis, according to histopathologic view.

The review of the literature showed the age range of 15 to 49 for surgical ciliated cyst[5]. Nevertheless, our case was a 74 yr old male. The surgical ciliated cyst cases commonly present swelling or pain of the cheek and the mucogingival fold, in the clinical feature[8]. In our case, the clinical observation was normal, and the patient had only pain in the left maxilla.

Three cases of a maxillary cyst lined were reported by ciliated epithelium. A history of radical maxillary surgery with the potential to entrap sinus epithelium in the maxilla was reported in each case[3]. Our patient had a history of nasal polyp surgery 40 years ago.

The pathophysiology of this lesion is the entrapment of the maxillary sinus mucosa in the wound by a surgical procedure (e.g. Caldwell-Luc procedure, Le Fort I osteotomy), which leads to the inflammatory process that induces the cystic changes of the trapped respiratory mucosa, and the expansion of the cyst by the osmotic difference from the surrounding tissue[9]. The differential diagnosis of surgical ciliated cysts from other cysts with similar clinical features such as mucous retention cysts, odontogenic cysts, simple bone cysts, or residual cysts is very important[10,11].

Radiographic feature of surgical ciliated cyst is a well-defined unilocular radiolucency closely associated with the maxillary sinus. The cyst may be enclosed by a zone of sclerosis. When the cyst expands, the adjacent sinus wall may become thin and finally perforated. Although it is usually unilocular, multilocular variants have also been reported[11]. In our report a cystic-like lesion with the destruction of the buccal cortex and thinning of the lingual cortex was obvious.

In histological features of case[1], the connective tissue of the cyst wall mainly contains an intense chronic inflammatory infiltrate of lymphocytes and

plasma cells. Some dystrophic calcifications were also detected. The cystic content was hemorrhagic. Furthermore, it contained a various amounts of inflammatory cells such as neutrophils.

In a treatment plan of the surgical ciliated cyst, 110 cases were reported. The authors advised enucleation or marsupialization of the cyst. In our case, we enucleated the lesion with the excisional biopsy[5].

A study with 60 patients was reported who presented surgical ciliated cysts and reported a recurrence rate of 20% with a mean follow-up period from the initial operation of 4.7 years. The treatment of choice for our patient was enucleation, and after 6 months of follow-up, there has been no recurrence[12].

CONCLUSION

The clinical examinations and radiographs of the jaw lesions may lead the maxillofacial surgeons to misdiagnosis and mistreatment. Therefore, it is obligatory to notice all aspects of these lesions carefully to obtain the correct diagnosis.

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CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

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