

Maxillary Chronic Inflammatory Odontogenic Cyst: A Case Report

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ABSTRACT

Residual cysts are inflammatory odontogenic cysts of the jaw. They are mainly retained radicular cysts after tooth extraction. These lesions are asymptotic and have the potential to damage and infect adjacent bone. In addition, they may resemble more serious lesions and tumors on radiographs. It could be argued that early detection and correct diagnosis are critical for effective treatment of residual cysts. The present case report discusses some technical aspects of surgical treatment of a large maxillary midline residual cystic lesion.

Keywords: Residual cyst, maxillary cyst.

INTRODUCTION

The World Health Organization (WHO) classifies mandibular neoplasms into three categories: odontogenic cysts, nonodontogenic cysts, and pseudocysts. (1-3) The term cyst is defined as "a pathologic cavity with a fluid, semifluid, or gaseous content that does not result from the accumulation of pus. (4) The mandible and maxilla have the highest incidence of cysts in the human body due to the abundance of epithelial remnants in the jaws. (5) Approximately 90% of oral and maxillofacial cysts are odontogenic in origin (2,3,6,7) and are the second most common oral and maxillofacial cysts in adults. (6)

The classification of odontogenic and nonodontogenic cysts is based on the origin of the epithelial lining. (2-4,7,8) Odontogenic cysts derive their epithelium from remnants of tooth-forming tissue. (2-4, 7,8) These include the diminished dental epithelium, the remnants of Malassez and Serres, and others. (4,7) These epithelial remnants may contribute to the formation of residual cystic lesions after extraction of the associated offending tooth (7).

According to the fourth edition of the World Health Organization (WHO) classification, odontogenic cysts can be further subdivided into inflammatory (radicular, residual, and paradental cysts), developmental (dentigerous, odontogenic keratocyst, lateral periodontal, gingival, glandular odontogenic, calcifying odontogenic, and orthokeratinized odontogenic cysts), and neoplastic cysts. (1-3) Inflammatory cysts are the most common type of odontogenic cysts, accounting for 50-75% of all jaw lesions. (2) This type of cyst may be triggered by the proliferation of epithelial debris as a result of inflammation. Apical periodontitis, caused by tooth death and pulp necrosis, is the underlying cause of the inflammation. (2,8) Typically, these cysts are asymptomatic and are discovered incidentally during a routine radiographic examination. (9)

Residual cysts are inflammatory in origin and often arise from a radicular cyst or other inflammatory cyst in the jaw that has grown apically or adjacent to a removed tooth. Residual cysts often have the same histopathologic features as radicular cysts. (8)

Because a variety of jaw cysts resemble each other in their clinical and radiographic manifestations, the diagnosis of odontogenic lesions usually requires a thorough examination of clinical, radiographic, and histopathologic evidence. (5)

However, the identification of residual cysts is still controversial, as some authors suggest that most of them are gradually resolving radicular cysts. (10) Compared to other types of odontogenic cysts, residual cysts have received significantly less attention in the literature. (11) Although these cysts grow slowly and are usually discovered incidentally, they are capable of growing to enormous sizes, displacing surrounding tissues, and transforming into either malignant or benign tumors. (1,7,10).

CASE REPORT

A 46-year-old female patient presented to the Department of Oral and Maxillofacial Surgery, XX University, Faculty of Dentistry. The patient presented with a chief complaint of painless swelling in the right upper maxillary and palatal region, postnasal discharge, and facial asymmetry. She had been confronted with the gradual growth of the cyst for the past 7 months.

The patient's medical history was documented; she did not have any systemic diseases and did not take any medications regularly. The patient stated that she had undergone cyst surgery in the area of her complaint in 2016 and had undergone uncomplicated extraction of many upper right posterior teeth in the past years.

Clinical evaluation of the extraoral features revealed a diffuse swelling of approximately 5 cm \times 3.7 cm in the right middle third of her face, with enlargement of the right nasolabial sulcus and slight deviation of the philtrum and upper lip to the opposite side. The mouth opening seemed sufficient and the skin appeared normal. There was no localized fever, lymphadenopathy, or paresthesias, and the skin around the swelling was pinchable.

An intraoral examination revealed that the canine, premolars, and molars were all absent. A well-defined swelling in the buccal-vestibular and palatal region extended from the first premolar to the maxillary tuberosity. The mucosa above the swelling appeared normal with no discharge or ulceration (Figure 1).



Figure 1. Preoperative intraoral photograph of the lesion

Radiographically, panoramic radiographs and cone beam computed tomography (CBCT) revealed a welldefined radiolucent lesion with irregular borders in the right maxillary sinus region that appeared more blurred than normal with extension into the sinus and edentulous regions, measuring 40.2 mm anteriorposteriorly, 30 mm bucco-palatally, and 40 mm superior-inferiorly. The lesion extended from the first premolar to the area of the maxillary tuberosity. In addition, the lesion displaced adjacent areas, including the buccal cortical plate, lateral nasal wall, and inferior orbital wall/orbital floor (Figure 2).



Figure 2. Preoperative CBCT photograph shows the measurements and extend of the lesion.

Electrical pulp vitality examination showed positive results in the maxillary right and left central incisors. After obtaining the patient's consent, it was decided to perform a biopsy. Aspiration of the first molar area revealed a brownish-red substance that was identified as an infected cystic fluid (Figure 3A). A preliminary diagnosis of an infected, inflammatory odontogenic lesion was made. Marsupialization was performed to reduce the size of the cyst and to relieve the pressure and fluid within the cyst (Figure 3B).

Three months after marsupialization, enucleation was performed under general anesthesia with a mucoperiosteal flap elevated from the central incisor to the maxillary tuberosity. The weak bone covering the lesion was removed and the cystic epithelium was carefully isolated. A beige semisolid content was noted. The maxillary sinus membrane and the epithelium of the cyst were induced simultaneously. The cyst and the maxillary sinus were enucleated (Figure 4). The flap was repositioned and the incision closed with resorbable sutures after irrigation of the cavity and filing of the bone margins.



Figure 3 . a. Image of the aspirated cystic fluid, b. Marsupialisation of the cyst image.



Figure 4. Intra-operative image of the cyst.

Histologic sections of the excised specimen showed ulceration of the surface epithelium. The connective tissue capsule was densely infiltrated with highly active chronic inflammatory cells, blood vessels, and extravasated red blood cells (RBCs). The specimen was identified as a residual cyst.

Five months postoperatively, the patient had restored facial symmetry and no signs of recurrence (Figure 5). The patient was followed intraorally (Figure 6) and radiographically (CBCT and panoramic x-ray) for 4 years after the procedure and no complications were noted (Figure 7 A,B,C,D).



Figure 5. Five months postoperatively.



Figure 6. 4 years postoperative.



Figure 7A. 1st year follow-up.



Figure 7B. 2ed year follow-up.



Figure 7C. 3rd year follow-up.



Figure 7D. 4th year follow-up Panoramic X-ray.

DISCUSSION

Inflammatory lesions are generally the most common type of jaw cyst. This cyst category consists of radicular, residual, and other, with residual cysts being the second most common. The prevalence of residual cysts in the jaw has been observed to range from 2.2% to 18% of all odontogenic cysts. (7)

The etiopathogenesis of radicular & residual cysts is similar. The process begins with the introduction of pathogens from a devitalized tooth into the periapical area of the jaw. If left untreated, this triggers the development of a periapical granuloma composed of activated T cells that produce cytokines. These cytokines interact with the epithelial cells, causing them to proliferate and differentiate into cysts. (8). If a periapical cystic lesion is extracted with improper curettage after removal of the non-vital tooth, it may develop into a progressively enlarged residual cyst, causing severe bone loss. (1)

Residual cysts mainly affect middle-aged individuals in their 3rd decade of life, but they can also occur in the 2nd to 8th decade of life. (12) Males are affected more frequently than females by a ratio of 3:2. (8,12) The maxilla is more commonly affected than the mandible. (66.2%), with the posterior portions most commonly affected. (7,12) The residual cyst formed in the present case was located in the posterior edentulous region of the maxilla, and the patient was a female in her fourth decade, which is consistent with the literature.

It is quite unusual for patients to present intentionally with the sole complaint of a residual lesion, as they are typically asymptomatic and are often detected incidentally during routine clinical and radiographic examinations (8). Although the patient had swelling in the right posterior maxillary edentulous region for six months and had post-nasal discharge, she finally contacted the dental department with a complaint of painless swelling in this case as well.

However, once the cyst becomes secondary infected, patients may experience pain, discomfort and swelling and become aware of the cyst. As this lesion gradually enlarges, it may cause significant tooth dislocation, instability, and bone disintegration at vital sites such as the inferior alveolar canal, maxillary antrum, and sinus cavity.

(7) In the present case, because the cyst filled the maxillary sinus, extended to adjacent structures, and even infected and resorbed the lateral nasal wall, the patient had various symptoms, including postnasal discharge and swelling in the vestibular sulcus.

Residual cysts are subclassifications of inflammatory cysts that often occur periapically and persist after removal of the corresponding (correlated) tooth.(8) In the present case, the patient underwent multiple extractions in the same region of the cyst.

On radiographs, residual cysts appear as unilocular, distinct, radiolucent cysts. They generally appear round or oval in shape, surrounded by a narrow sclerotic margin. They vary in size within the edentulous site of a previously removed tooth. Lesions may degrade over time, resulting in radiolucent zones (dystrophic calcifications) within the lesion cavity. Cystic lesions appear radiolucent when they are in the bone, but when they extend into the sinus, they appear partially radiopaque compared to the sinus cavity, due to the difference in density. There is also a radiopaque line between the sinus cavity and the cyst capsule, which facilitates the diagnosis of cysts. (8, 13) A panoramic radiograph of the cyst in the presented case showed a slightly more blurred radiolucency than the normal sinus appearance.

Radicular and residual cysts have similar histologic features. However, in residual cysts, once the cause of inflammation (i.e., carious tooth) is removed, the inflammatory infiltrate in these cysts diminishes, noninflammatory connective fibrous tissue develops in the cyst walls, and the epithelial lining becomes thinner and more regular. (8)

Histopathologic examination remains the primary approach to diagnosis of these cysts, in addition to radiographic examination. (14) In this case, radiologic and clinical correlation, along with histopathologic examination, become critical in arriving at an appropriate diagnosis.

Treatment approaches that can be performed to manage the residual cyst include enucleation, marsupialization, curettage, decompression, or endoscopic surgery, depending on the size of the cyst, to reduce the size of the cystic lesion by relieving the intraluminal pressure within it. (15) Larger residual cysts, especially those that extend into adjacent anatomic tissues, require periodic management with marsupialization to reduce fluid in the cyst and minimize its size, followed by enucleation of the remaining cyst lining However, the disadvantages of this method are the need for general anesthesia, prolonged duration, two-stage surgical treatment, and most importantly, the risk of proliferation of the cyst epithelium to carcinoma in situ, which is the major disadvantage of marsupialization. (7) The cystic lesion in this case was large and extended to the inferior orbital wall; it was first marsupialized and then enucleated along with the cyst capsule and infected sinus membrane.

Because epithelial remnants that maybe left in the cavity during the surgical excision of the cystic lesion lining, a residual lesion could regenerate and reoccur in the region.(13) Therefore, regular follow-up is necessary to detect any recurrent episodes or malignant developments. (15) The patient was followed up four years after surgery and no complications were noted.

Conclusion

The residual cyst is an inflammatory odontogenic cyst that often goes unnoticed by patients because it remains asymptomatic until it becomes infected. It has similar clinical and pathologic characteristics to more aggressive cysts involving the jaw bone.

Histopathologic evaluation and biopsy are the primary methods for establishing an accurate diagnosis. In addition to radiographic and cytologic evaluations. We concluded that early detection, along with effective and appropriate treatment methods, is essential to prevent residual cysts from enlarging and affecting adjacent structures. In addition, adequate surgical removal of epithelial debris will reduce the recurrence rate of these cysts.

Conflict of Interest

All contributing authors declare that they have no conflicts of interest.

Financial Disclosure

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Informed Consent

Written informed consent was obtained from the patient who participated in this case.

References

- Borah GL, Aziz SR. Tumors of the mandible. In: Mathes SJ, Hentz VR, eds. Plastic Surgery. 2nd ed. vol. 5. Philadelphia: W.B. Saunders; 2006:189– 215.
- 2. Wright, J.M.; Vered, M. Update from the 4th edition of the World Health Organization classification of head and neck tumours: Odontogenic and maxillofacial bone tumors. Head Neck Pathol. 2017, 11, 68–77. [Google Scholar] [CrossRef][Green Version]
- 3. Soluk-Tekkeşin, M.; Wright, J.M. The World Health Organization Classification of odontogenic lesions: A summary of the changes of the 2017

(4th) Edition. Turk Patoloji Derg. 2018, 34. [Google Scholar] [CrossRef] [PubMed]

- Smith RA. Jaw Cysts. In: Lawlwani AK, ed. Current Diagnosis and Treatment Otolaryngology Head and Neck Surgery. 3rd ed. New York: McGraw-Hill; 2008: 374–388.
- Fomete B, Osunde OD, Ogbeifun J, Agbara R, Ononiwu CN. A 10-Year Retrospective Analysis of 64 Cases of Cystic Lesions of the Oral and Maxillofacial Region in a Nigerian Tertiary Hospital. Oman Med J. 2016; 31: 434–438. https://doi.org/10.5001/omj.2016.87 PMID: 27974959
- 6. Jones AV, Franklin CD. An analysis of oral and maxillofacial pathology found in adults over a 30-year period. J Oral PatholMed 2006; 35: 392e401.
- 7. Tsvetanov TS. Residual cysts: A brief literature review. Int J Med and Dent Sci. 2016; 5: 1341– 1346.
- 8. Shear M, Speight P. Cysts of the Oral and Maxillofacial Regions. 4th Ed. Oxford: Blackwell and Munksgaard. 2007: 123–140.
- Sagit, M.; Guler, S.; Tasdemir, A.; Somdas, M.A. Large radicular cyst in the maxillary sinus. J. Craniofac. Surg. 2011, 22, e64–e65. [Google Scholar] [CrossRef]
- 10. Lee J, Costandi J, Mandel L. The residual radicular cyst. N Y State Dent J. 2014;80(4):38–40.
- High AS, Hirschmann PN. Age changes in residual radicular cysts. J Oral Pathol. 1986;15:524–8.
- Blois M, de Souza F, Kuhn A, Zanin R. Giant radicular cyst of the maxilla: enucleation and follow-up. Int J Oral Maxillofac Surg. 2019;48:226–7.
- 13. Jamdade A, Nair GR, Kapoor M, Sharma N, Kundendu A. Localization of a peripheral residual cyst: diagnostic role of CT scan. Case Rep Dent. 2012;2012:760571.
- 14. Boffano P, Gallesio C. Exposed inferior alveolar neurovascular bundle during surgical removal of a residual cyst. J Craniofac Surg. 2010; 21: 270–273. https://doi.org/10.1097/SCS.0b013e3181c5a42b PMID: 20098197
- 15. Kammer PV, Mello FW, Rivero ER. Comparative analysis between developmental and inflammatory odontogenic cysts: retrospective study and literature review. Oral Maxillofac Surg. 2020;24(1):73–84.