

CASE REPORT

Noninvasive Treatment Choice for an Aged Down Syndrome Patient Presenting a Residual Periapical Cyst

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ABSTRACT

Aim: This is the first report to illustrate the marsupialization as an effective treatment for a Down Syndrome (DS) patient presenting a residual periapical cyst.

Background: These cysts occur within the alveolar ridge, usually at the local site of a previously extracted tooth that did not received proper curettage; usually the surgical excision of a cyst and also the vigorous curettage of a socket is very simple, if not for the fact that mentally disabled patients require rapid and non-stressful procedures.

Case description: The 54-year-old DS patient represented herein received a minimally invasive marsupialization under local anesthesia. Due to the large extent of the lesion, the acrylic resin drain was maintained for 30 days. Through the following period, a daily irrigation of the cystic cavity with saline solution was carried out to prevent a secondary infection within the cystic cavity. A follow-up of 16 months showed no signs of recurrence.

Conclusion: Marsupialization of residual periapical cyst is completely effective and safe, even for a DS patient that is considered to be at an advanced age.

Clinical significance: Marsupialization poses as a minimally invasive choice for mentally disabled patients, even when presenting advanced ages; treatment success was stated by the easy clinical conduct, uneventful postoperative situation and the lack of recurrence along 16 months of follow-up.

Keywords: Down Syndrome, Aged, Marsupialization, Residual Periapical Cyst.

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INTRODUCTION

To the best of our knowledge, this is the first report of a residual periapical cyst in a patient with Down Syndrome (DS). The complexity of oral health needs of individuals that present intellectual disabilities may be related to the inability to receive appropriate professional care,¹ and although there is no evidence of a higher prevalence of periapical cysts in DS patients, the clinical conduct to be chosen is of major importance and may significantly contribute to the oral health maintenance of these patients.

Actually, the oral care of DS patients should be logically based on general health concepts;² moreover, dentists should be aware of the patients' systemic conditions in order to treat them accordingly. In that way, when correctly trained, dentists can provide dental care to DS patients,² and this case report illustrates how a non-invasive approach such as the cyst marsupialization (already highly accepted for the general population)^{3,4} can be completely successful in a DS individual.

The patient here presented is 54 years old, and although this advanced age is still considered to be unusual for DS patients,⁵ several medical conducts have been improved and the DS life expectation continuously rises.⁶ Although DS is constantly related to immunological, hematological and esophageal abnormalities,² DS patients can be generally treated for dental conditions in an ambulatory level, as long as procedures that involve manipulation of the gingival tissue and/or periapical region are preceded by the antibiotic prophylaxis recommended by the American Heart Association (AHA).⁷

CASE DESCRIPTION

This case report obtained Ethical clearance by acquiring a written informed consent from the patient's legally responsible person. A 54-year-old white female diagnosed with Down Syndrome (DS) was referred to the Stomatology Clinic of our institution by her sister with a complaint of swelling at the left posterior maxillary region, which she had first noticed 21 days before and was not related to any local trauma. The patient did not report any painful symptomatology and her medical history was unremarkable, which is a rare condition for a middle age patient with DS^{2,6}; her sister also could not describe when the local missing teeth were extracted.

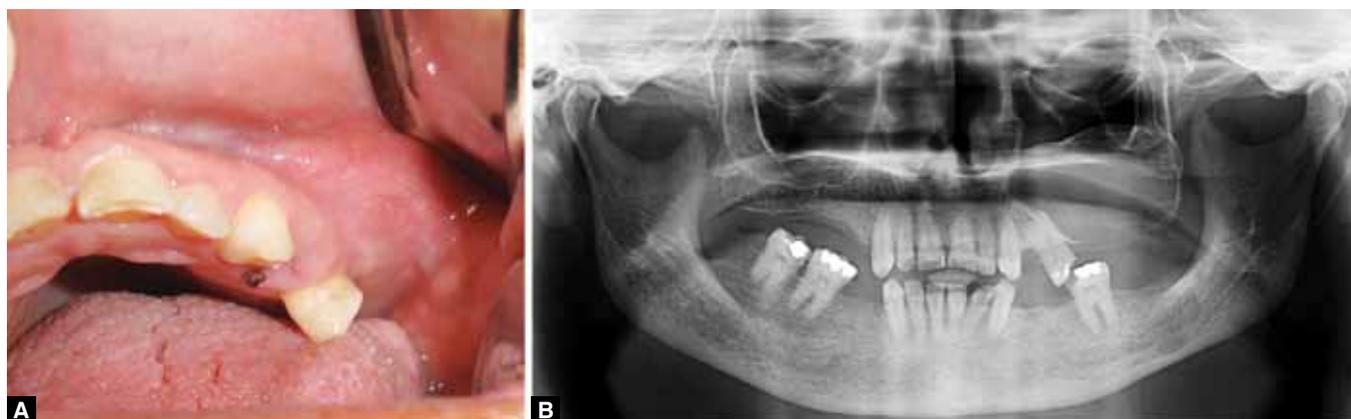
Extraoral examination showed typical features of a DS individual, including short stature, depressed nasal bridge, hypertelorism, flat face and large lips. Facial asymmetry caused by swelling in the lower third of the face on the affected side was also observed. The intraoral examination revealed oral manifestations of DS, such as macroglossia and atresic palate, and a wide swelling at the ridge of the left maxillary first premolar to first molar region with buccopalatal expansion covered by normal mucosa (Fig. 1A). The left maxillary second premolar was responsive to vitality pulp testing. Radiographic examination showed a large well-defined unilocular radiolucency at the left posterior maxillary alveolar region below the maxillary sinus. The lesion caused displacement of the roots of the adjacent premolar and no signs of root resorption were noted (Fig. 1B).

The differential diagnosis included Residual Periapical Cyst and Keratocystic Odontogenic Tumor. Hematological tests were requested and the results were within the normal limits. The proposed treatment was the cystic decompression by the marsupialization while excising the overlying

mucosa, opening a window (1 cm in diameter) into the cystic cavity and suturing an acrylic resin drain to the oral mucosa (Fig. 2) under local anesthesia. Prior to the surgical intervention the patient received the demanded antibiotic prophylaxis.⁷

The specimen was fixated in 10% formaldehyde and sent for histologic examination. Microscopically, the lesion was characterized by a cystic cavity lined by an atrophic stratified squamous epithelium exhibiting a flat epithelial/capsule boundary. The cystic wall consisted of fibrous connective tissue presenting a discrete inflammatory infiltrate containing lymphocytes, plasma cells and histiocytes (Fig. 3). The histopathological features were consistent with the diagnosis of Residual Periapical Cyst.

Due to the large extent of the lesion, the acrylic resin drain was maintained for 30 days. Through the following period, a daily irrigation of the cystic cavity with saline solution was carried out to prevent a secondary infection within the cystic cavity; antibiotic therapy consisted on



Figs 1A and B: (A) Clinical appearance of the lesion comprehending a significant swelling recovered by intact mucosa at the left posterior side of the oral cavity and (B) radiographic examination showing a large well-defined unilocular radiolucency at the left posterior maxillary alveolar region below the maxillary sinus



Fig. 2: Immediate postoperative situation illustrating the acrylic resin drain placed for the cyst decompression

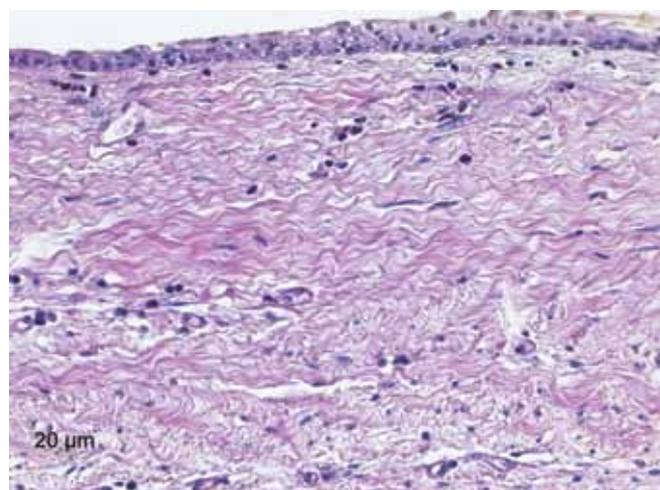


Fig. 3: Histological appearance of the lesion showing a cystic capsule constituted by connective tissue lined by an atrophic stratified squamous epithelium (Hematoxylin and Eosin at 400X)



Fig. 4: Panoramic radiograph showing no signs of lesion recurrence in a 16-month follow-up

500 mg of amoxicillin at every 8 hours, for 7 days. At the patient's 16 months follow-up, the lesion clearly resolved and the patient presents no signs of recurrence (Fig. 4).

DISCUSSION

As suggested by previous researches, individuals that present intellectual disabilities are inclined to show poor oral hygiene and are more susceptible to periodontal diseases and even caries.⁸ Nevertheless, there are few reports, if any, of periapical cysts in DS patients. Keratocystic Odontogenic Tumors, on the other hand, may be correlated to other syndromes such as the Nevroid Basal Cell Carcinoma Syndrome or Ehlers–Danlos Syndrome,^{9–11} though the histopathologic features excluded the possibility of this lesion.

A periapical or radicular cyst appears typically as a rounded radiolucency that encircles the apical part of any tooth, while the residual cyst shows the same radiographic features, although it is not associated with a tooth apex. When a tooth is surgically extracted, a careful curettage must follow to exclude the possibility of an upcoming residual cyst; however, long-taking procedures may not be possible when dealing with a DS patient.

While life expectation of DS patients is constantly rising,⁶ these slow-growing radicular or residual periapical cysts^{12,13} may become a reality. Actually, residual cysts are even more slow-growing than their tooth-associated counterpart.¹³ In addition, different growing rates are expected for these lesions and are usually intersected by non-proliferative periods; periapical cysts that are lined by atrophic epithelium and that present a flat epithelial/capsule borderline, as the one here reported, were related to a reduced growth.¹⁴

Among the systemic abnormalities that a DS individual can present are the cardiovascular abnormalities, including unrepaired cyanotic congenital heart disease;² individuals that present this condition may become cyanotic, hypoxic and even present predisposition to postoperative hemorrhage.¹⁵ In that way, dentists should avoid stressful situations at most

when treating patients with cyanotic heart diseases;¹⁵ that is why marsupialization serves as an elective choice for DS individuals presenting maxillary cysts.

In fact, marsupialization is a widespread technique that has proven efficacy for the treatment of several cysts.^{3,4,16} While treating a DS patient, on the other hand, this was a novel approach; however totally safe and minimally invasive, which is precious for mentally disabled patients that often require sedation.^{2,17} This case report was also privileged to demonstrate a DS patient normally recovering after the surgical procedure; the antibiotic coverage predicted by AHA⁷ should always be respected and the patient recuperation should be followed closely, once DS patients tend to present defects in natural killer, B and T cell functions.^{18,19}

CONCLUSION

Residual periapical cyst may appear as the consequence of an undertreated radicular cyst, which is understandable due to the difficulty to perform surgical therapies on DS patients. Nonetheless, marsupialization is completely effective and safe, even for a DS patient that is considered to be at an advanced age.

CLINICAL SIGNIFICANCE

Marsupialization poses as a minimally invasive choice for mentally disabled patients, even when presenting advanced ages; treatment success was stated by the easy clinical conduct, uneventful postoperative situation and the lack of recurrence along 16 months of follow-up.

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