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Case Report

Surgical Removal of an Unusual Radicular Cyst in the Down's Syndrome Adolescent: Case Report with 22-Years Follow-up

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Abstract

Down's Syndrome is one of the most frequent genetic anomalies in dental clinic. Several systemic and stomatological alterations can be observed in patients with Down's Syndrome. Some care during dental treatment may become necessary for patients with Down's Syndrome, influenced by systemic alterations. Additionally, the restriction of motor coordination may hinder oral hygiene, causing high incidence of caries and periodontal diseases. In this perspective, pulp necrosis and the development of periapical alterations may be observed subsequently. The purpose of this article is to present the case of a large radicular cyst in the chin of a patient with Down's Syndrome. The lesion was removed by total enucleation technique. The patient has been followed for 22 years, presenting bone repair and no recurrence of the lesion.

Keywords: *Down's Syndrome; Radicular Cyst; Oral Surgery; Special Care in Dentistry*

Introduction

Trisomy 21, better known as Down's Syndrome, is one of the most frequent genetic anomalies in dental clinic. Approximately 3% of the world population is affected, with an estimated one affected patient per 600 live births [1-5].

It was initially described by British physician John Langdon Down in 1866. Down described the physical characteristics similar to the Mongolian population. Later, in 1959, the French pediatrician and geneticist Jérôme Lejeune reported the genetic abnormality caused by trisomy 21 [1-4].

Several systemic and stomatological alterations can be observed in patients with Down's Syndrome. Table 1 and 2 show, respectively, the main systemic and stomatological alterations frequently observed in patients with Down's Syndrome. Deficient motor coordination may impair oral hygiene, increasing the incidence of carious lesions and periodontal diseases. Consequently, there is a greater risk of pulp necrosis and the development of periapical alterations. Some special care may be required during dental treatment due to systemic alterations [1-3].

Systems	Alterations
Nervous	Impaired coordination and motor function; Dementia analogous to Alzheimer's disease; Delayed communication and speech.
Cardiovascular	Ventricular septal defects; Patent ductus arteriosus; Mitral valve prolapse; Congenital heart disease.
Hematopoietic	Impaired immunity (neutropenia, eosinophilia, lymphopenia, immunoglobulins, impaired chemotaxis); Risk of leukaemia; Risk of hepatitis B.
Musculoskeletal	Generalised muscular hypotonia;
Ophthalmic	Eyelid fissure with high external angles; Wide epicanthium; Convergent strabismus; Nystagmus; Iris hypoplasia; Lens opacifications.
Endocrine	Congenital hypothyroidism;
Respiratory	Respiratory complications (pneumonia, bronchitis); Allergies.
Behavior	Enhanced sexuality; Spontaneity touched; Kindness; Anxiety; Stubbornness.
Phenotypic alterations without pathological significance	Wide, short neck with abundant skin.

Table 1: *Systemic alterations observed in patients with Down's syndrome.*

Regions	Alterations
Palate	Ogival palate Soft palate insufficiency
Perioral muscles	Hypotonic muscles Angle of mouth turned downwards Lower lip everted Oral breathing Sialorrhoea Angular cheilitis
Tongue	Fissured tongue Lingual protrusion Macroglossia Dry tongue
Teeth	Macrodontia or Microdontia Agenesis, Hypodontia, Anodontia or Oligodontia Supernumerary Teeth Diastemas Taurodontism Fusion Morphological alterations, Conoid teeth Hypoplasia and hypocalcification Reduced risk of dental caries Delayed eruption Prolonged retention of deciduous teeth
Periodontium	Periodontal diseases Halitosis
Occlusion	Malocclusion Temporomandibular disorders Bruxism

Table 2: *Stomatological alterations observed in patients with Down's Syndrome.*

Purpose of the Study

The purpose of this article is to present the case of a large radicular cyst in the chin of a patient with Down's Syndrome. The lesion was removed by total enucleation technique.

Case Report

A Caucasian male patient, with Down's Syndrome, 13-years-old (Figure 1), attended the dental clinic, accompanied by his mother, complaining of pain in the region of the lower incisors.

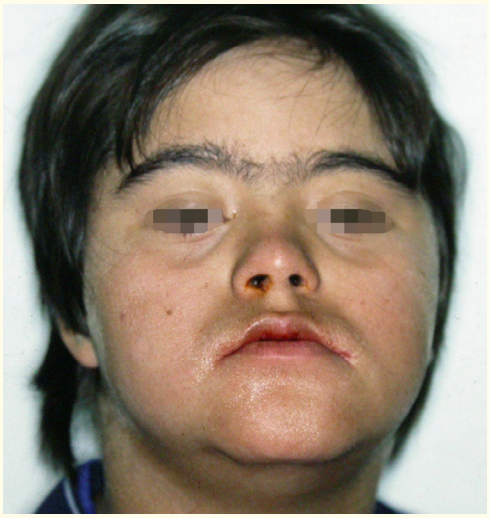

Figure 1: *Patient with Down's Syndrome.*

Clinically, caries in lower incisors, dental absences and malocclusion were observed. No signs of infection or fistula were observed in the mucosa of the lower incisors. However, on palpation, an increase in cortical bone volume was observed (Figure 2). The mother of the patient reported endodontic treatment of the lower incisors 6 months ago, indicated by the presence of a cystic lesion in the region of the chin. Panoramic radiography confirmed the unilocular circumscribed radiolucent image in the periapical region of the lower incisors (Figure 3).

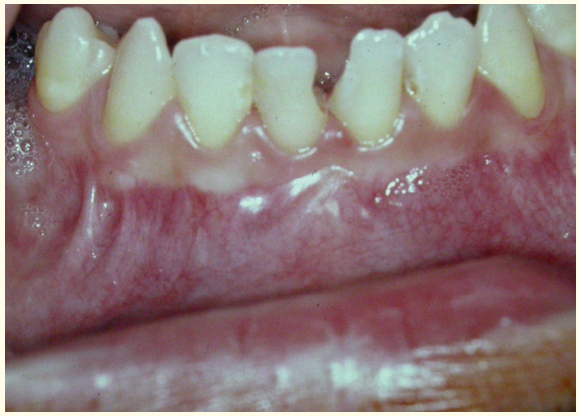

Figure 2: *Increased cortical bone volume in the chin.*

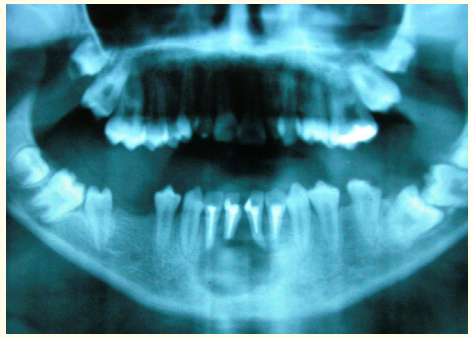

Figure 3: *Unilocular circumscribed radiolucent image in the periapical region of the lower incisors.*

Surgical removal by enucleation technique was indicated. Clarifications were provided to the mother of the patient who, after agreement, signed a consent form for the procedure.

Prior to the surgical procedure, prophylactic antibiotic was prescribed (American Heart Association protocol). Under local anesthesia, intrasulcular and relaxing incisions were made between teeth 33 and 43. The mucoperiosteal flap was detached, exposing the cortical bone, which was already reabsorbed by the lesion (Figure 4). An aspiration puncture was performed, showing a blood collection and revealing the infected condition of the lesion (Figure 5). The lesion was totally enucleated (Figure 6) and the bone cavity was abundantly washed with saline solution (Figure 7). The mucoperiosteal flap was repositioned and sutured (Figure 8). With the purpose of containing the edema, avoiding suture dehiscence and protecting the region of the own patient, a dressing was performed with adhesive tape, which remained for 15 days (Figure 9).

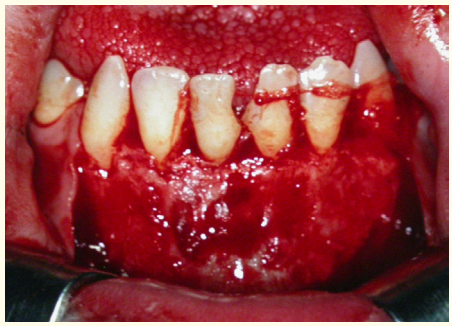

Figure 4: *Mucoperiosteal flap detached, exposing the lesion.*

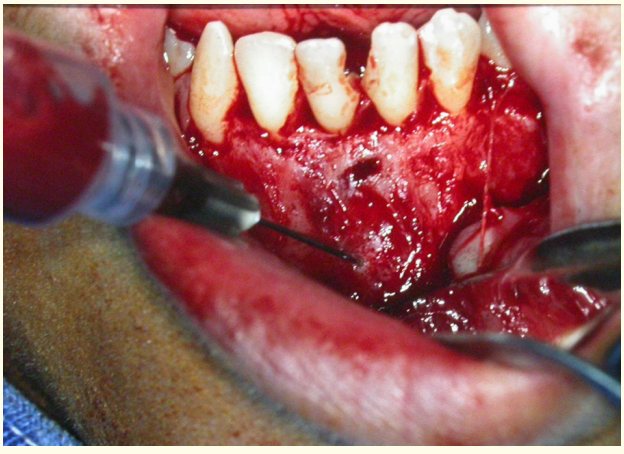

Figure 5: *Aspiration puncture showing a blood collection.*

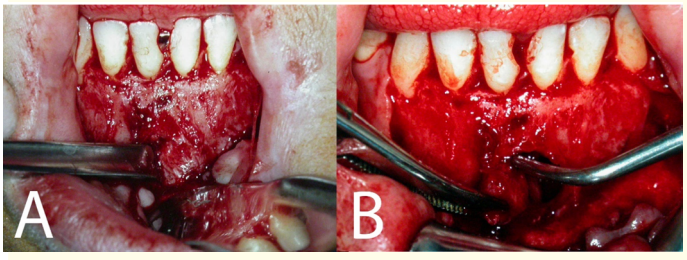

Figure 6: *Enucleation of the lesion (A); removal of the cystic capsule (B).*

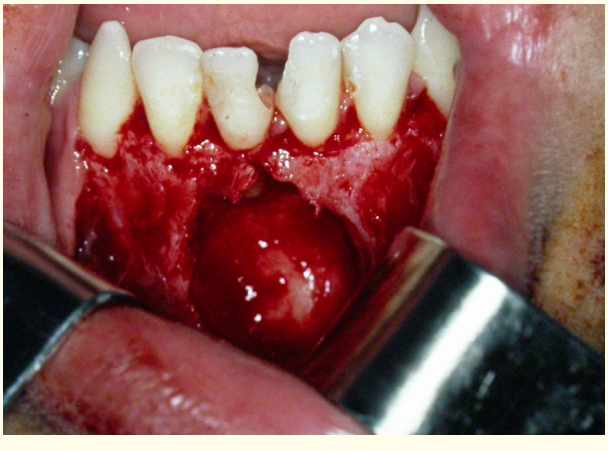

Figure 7: *Bone cavity cleaned.*

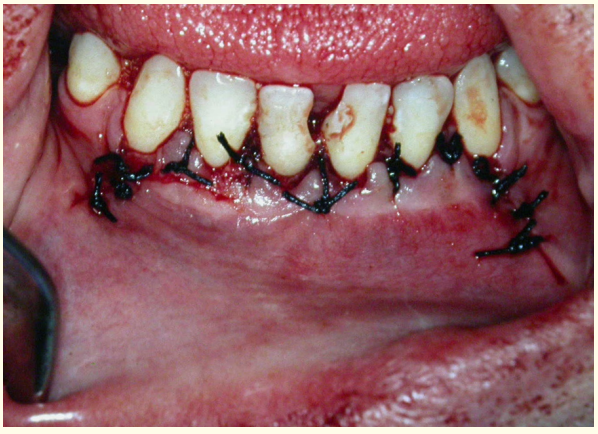

Figure 8: *Mucoperiosteal flap sutured.*

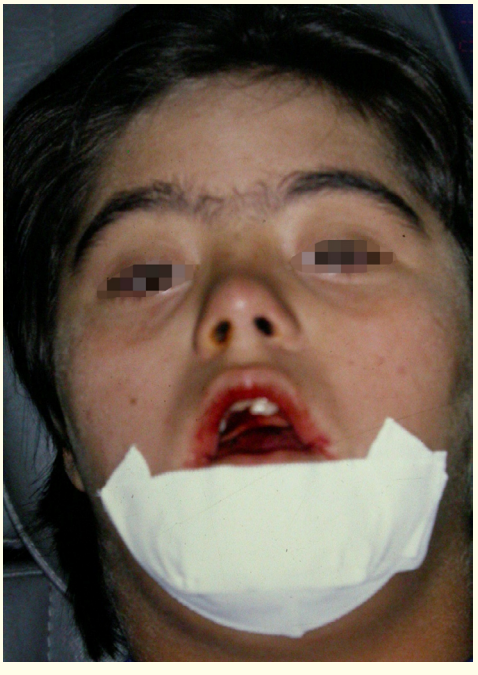

Figure 9: *Dressing with adhesive tape on the chin of the patient.*

The removed fragment (Figure 10) was fixed in 10% formalin and sent to the Laboratory of Surgical Pathology of the School of Dentistry of the University of São Paulo. Histopathological examination revealed a mucosal fragment composed of fibrous connective tissue and a thin lining of typical squamous epithelial tissue. Hyalinized areas, lymphoplasmocytic inflammatory infiltrate, hem-

orrhagic foci and calcification points were observed (Figure 11). The final diagnosis was radicular cyst.

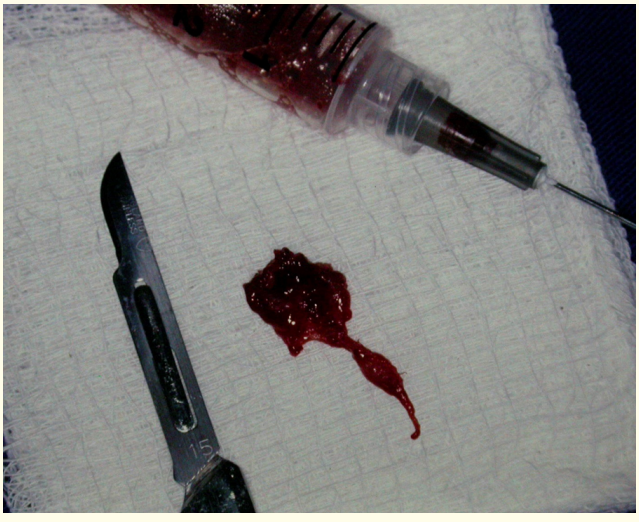

Figure 10: *Fragment removed.*

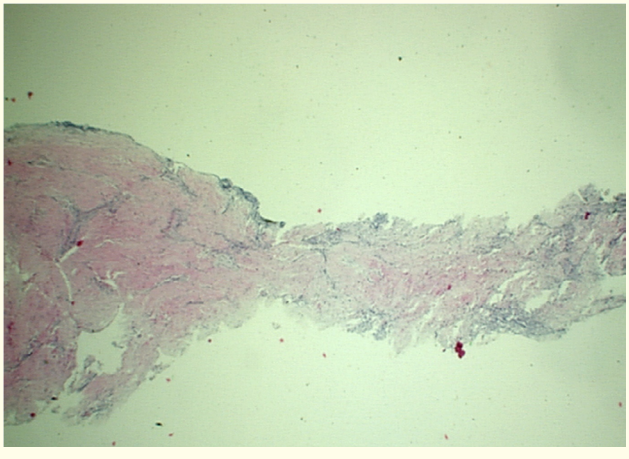

Figure 11: *Histopathological aspects of the radicular cyst.*

After 15 days, the bandage and remaining sutures were removed. No complaints or complications were reported. Satisfactory tissue repair was observed 40 days after surgery. The patient has been followed for 22 years, presenting bone repair and no recurrence of the lesion (Figure 12).

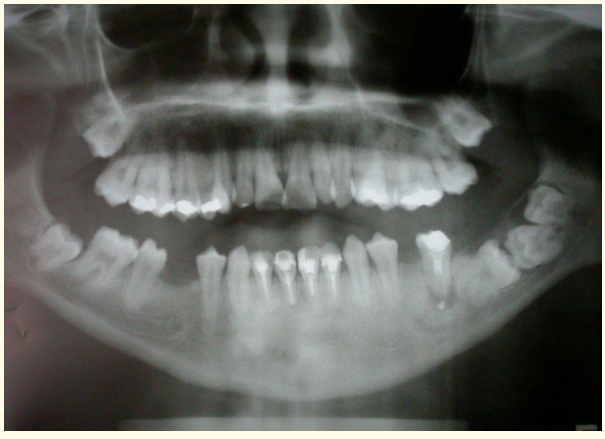

Figure 12: *Bone repair after 22 years of the radicular cyst removal.*

Discussion

Maxillary bone cysts are frequent in dental clinic, and odontogenic cysts are the most prevalent ones, between 60% and 94.5%. Generally, they are lesions developed by the stimulation of periapical epithelial remnants facing the inflammatory response originating from pulp necrosis, probably caused by the evolution of carious lesions. Radicular cysts are the most prevalent odontogenic cysts, in approximately 46.8% of the cases [6-10].

They are usually diagnosed during routine radiographic examination or, upon symptomatic complaint by acute exacerbation of the infectious process. Growth is slow. Mobility, edema, tooth movement or resorption may complete the associated clinical signs [1,9,10].

The cysts may evolve, remain stable or regress. Several treatments can be performed. When pulp necrosis is diagnosed, endodontic treatment should be performed. Surgical treatment can be performed by decompression (or marsupialization) or enucleation techniques. In larger cysts, the decompression and marsupialization techniques are indicated, which aim at reducing the size of the lesion, until subsequent enucleation. However, they require frequent visits for hygiene of the cystic cavity and strict cooperation of the patient, which, in most cases, hinder the success of the techniques. The enucleation technique is indicated for smaller cysts or when total removal of the lesion is really imperative. It is the technique of choice, with or without the use of biomaterials for grafts. After the cystic lesion excision, only the filling of the bone cavity

by the blood clot is enough, most of the times, for bone neoformation, as it was demonstrated in the present case. The increase in radiopacity inside the bone cavity, suggestive of bone neoformation after cyst enucleation, varies, on average, between 12 and 24 months. However, this time varies according to the size and nature of the cystic lesions [1,7-9,11,12]. In the present case, the patient has been followed up for 22 years, without signs of recurrence or symptomatic clinical signs.

Dental treatment to the Down's Syndrome patient requires some special care [1-3]. Table 3 summarized the main care. Treatment should be mild and calm, depending on the cognitive limitations of the patient. Prophylactic antibiotic (American Heart Association protocol) must be used in procedures that involve bleeding, due to the deficient immune response and predisposition to opportunist infections [1-3], as was used by us. Inhaled analgesia (gas mixture between nitrous oxide and oxygen) can be employed in outpatient care. Faced with the cognitive and collaborative limitations of patients, general anesthesia can be used in hospital settings. However, it should be considered when other modalities are inefficient for the treatment [1,2]. In the present report, due to the satisfactory collaborative and cognitive response, it was possible to perform the surgical treatment at outpatient level.

Care in dental treatment
Prophylactic antibiotic (American Heart Association Protocol)
Gentle and calm treatment
General anaesthesia or conscious sedation

Table 3: *Care during dental treatment of patients with Down's Syndrome.*

Conclusion

Several systemic and stomatological, pathological or morphological alterations may affect patients with Down's Syndrome. Among the stomatological alterations, periapical alterations resulting from pulp necrosis are frequently observed, due to the increased incidence of carious lesions. In this perspective, cystic lesions may occur in patients with Down's Syndrome. Depending on the collaborative and cognitive characteristics of the patients, exeresis of the cystic lesion is recommended. In the present case, enucleation of the radicular cyst was performed followed by bone neoformation, followed for 22 years, with no signs of recurrence. The knowledge of dental surgeons about systemic, cognitive and behavioral alterations, which may hinder dental care or require special care, is fundamental.

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