SVOA Dentistry

ISSN:2753-9172

ScienceVolks Open Access

Case Report

Surgical Removal of a Radicular Cyst on a Fused Tooth in a Patient with Down's Syndrome: 14-years Follow-up

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***Received:** February 10, 2022 **Published:** February 18, 2022*

Abstract

Down's Syndrome - also known as trisomy 21 - is one of the genetic anomalies most frequently observed in dental clinic. Patients with Down's Syndrome present several systemic and stomatological alterations. These systemic alterations may influence the dental treatment, requiring some special care. Facing precarious oral hygiene, due to motor coordination restriction, lack of motivation and even lack of professional guidance, a high incidence of caries and periodontal diseases is observed. Subsequently, pulp necrosis and the development of periapical alterations can be observed. The purpose of this article is to present the case of a patient with Down's Syndrome, who presented a fused tooth and radicular cyst. Endodontic treatment was performed and after 12 months, without signs of regression of the cystic lesion, surgical removal of the radicular cyst was performed. The patient has been followed clinically and radiographically for 14 years with no signs of recurrence of the lesion.

***Keywords:** Down Syndrome; Radicular Cyst; Endodontics; Oral Surgery; Special Care in Dentistry.*

Introduction

Down's Syndrome - also known as trisomy 21 - is one of the most frequently observed genetic anomalies in dental clinic. It is estimated that one patient is affected in every 600 live births, totalizing 3% of the world population[1-4].

It was first described by the British physician John Langdon Down in 1866, who identified in children, even sons of European parents, showed similar characteristics to Mongolians. Later, only in 1959, the French pediatrician and geneticist Jérôme Lejeune discovered the genetic anomaly caused by trisomy 21[1-3].

Patients with Down's Syndrome present several systemic and stomatological alterations. These systemic alterations may influence the dental treatment, requiring some special care during dental care. Faced with poor oral hygiene, due to motor coordination restriction, lack of motivation and even lack of professional guidance, a high incidence of caries and periodontal diseases is observed. Subsequently, pulp necrosis and the development of periapical changes can be observed. On the other hand, in view of the presence of diastemas and delayed eruption of teeth, a lower incidence of dental caries is reported[1,2].

The purpose of this article is to present the case of a patient with Down's Syndrome, who presented fused tooth and radicular cyst. Endodontic treatment and surgical removal of the radicular cyst were performed.

Case report

Japanese-descendent female patient, 31 years-old, with Down's Syndrome attended the dental clinic with a complaint of pain.

The patient presented satisfactory gingival condition and oral hygiene. During intraoral clinical examination, tooth 11 showed brownish coloration, suggesting previous endodontic treatment. Tooth 12 presented morphological and size changes. A fistula was observed in the mucogingival line between teeth 12 and 13 (Figure 1). In the palatal region, a gingival increase was observed in the periapical region of tooth 12, with a small ostium and purulent suppuration characterizing a gingival abscess. The patient presented tooth 12 with the coronary canal open for drainage of the purulent suppuration and had been consulted during previous emergency care (Figure 2). The painful symptomatology was consistent with this region.

Fig. 1: *Initial clinical aspects: frontal intrabuccal view.*

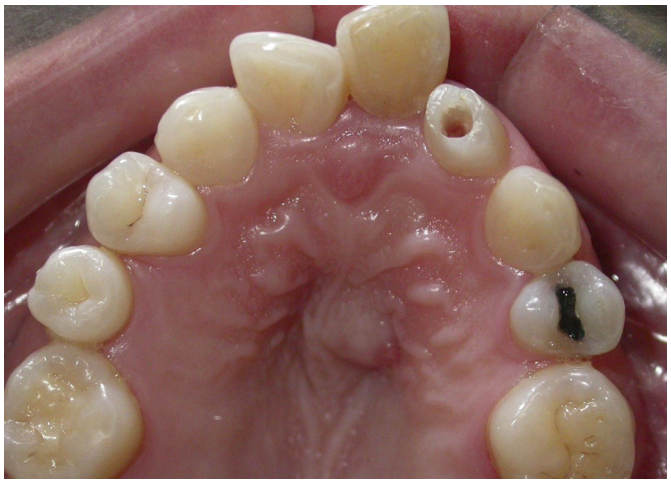

Fig. 2: *Gingival abscess in the periapical region of tooth 12.*

Radiographs showed a radiolucent image suggestive of periapical cystic lesion of teeth 11 and 12. Inside the lesion, a radiopaque image suggestive of endodontic cement extravasated inside the lesion from the filling of tooth 11 was observed. Tooth 12 presented 2 roots and 2 crowns overlapping, indicating the diagnosis of a fused tooth (Figure 3).

The mother of the patient reported endodontic treatment of tooth 11 six years ago and endodontic retreatment two years ago.

The endodontic treatment was performed, in 4 sessions, with 15 days between each one, with difficult access to both root canals (Figure 4). Between each visit, calcium hydroxide (Sealer 26™, Dentsply, São Paulo, Brazil) was inserted as an indwelling medication with the purpose of reducing the periapical lesion. Generally, there was secretion between visits. When the secretion was eliminated and the instrumentation of root canals was completed, it was possible to obturate the canals (Figure 5).

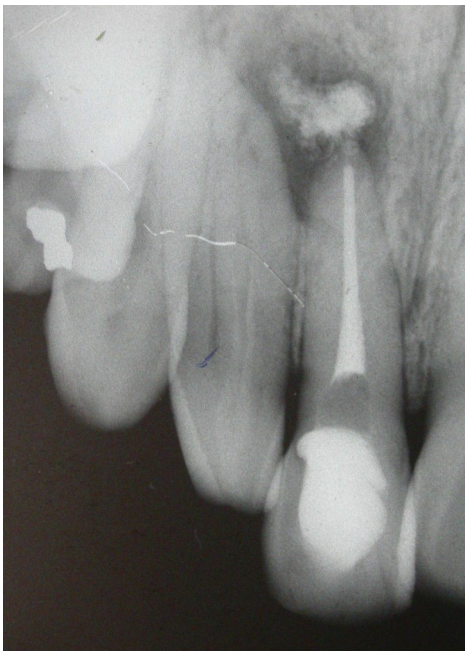

Fig. 3: *Radiolucent image suggestive of periapical cystic lesion of teeth 11 and 12 (fused tooth).*

Fig. 4: *Endodontic treatment of the tooth 12.*

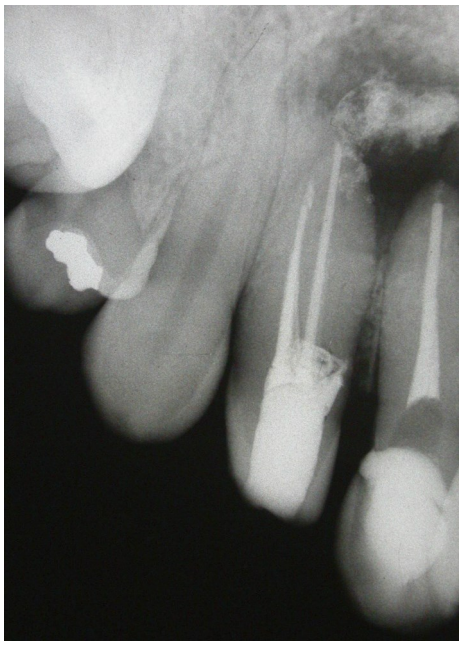

Fig. 5: *Endodontic treatment of the tooth 12 after the obturation of the root canals.*

After 12 months of endodontic treatment, without radiographic evidence of periapical repair, removal of the cystic lesion was recommended. The questions were clarified and the mother of the patient agreed and signed the consent form to perform the treatment. The American Heart Association prophylactic antibiotic protocol was employed, with the administration (orally) of 2g of amoxicillin one hour before the procedure.

Under local anaesthesia, an incision was made in the mucogingival line between teeth 21 and 13 (Figure 6). The mucoperiosteal flap was detached, showing the erosion of the cortical bone over the lesion (Figure 7). The lesion was enucleated and the bone cavity was washed abundantly with saline solution (Figure 8). Complementarily, apicectomy of tooth 12 was performed (Figure 9). Calcium hydroxide powder was inserted into the bone cavity in order to aid disinfection (antibacterial effect due to making the site alkaline) and bone formation (Figure 10). The mucoperiosteal flap was reattached and sutured in position (Figure 11). The patient was given analgesic, anti-inflammatory and antibiotic drugs.

Fig. 6: *Incision made in the mucogingival line between teeth 21 and 13.*

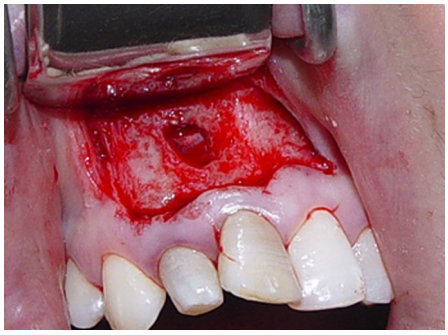

Fig. 7: *Mucoperiosteal flap detached, showing the erosion of the cortical bone over the lesion.*

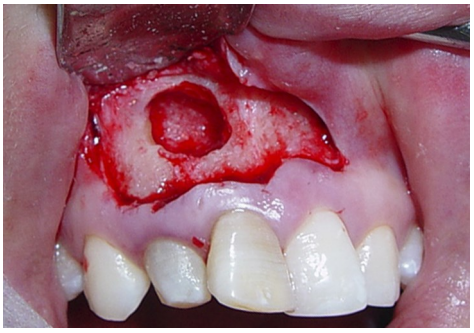

Fig. 8: *Enucleation of the lesion and the bone cavity washed.*

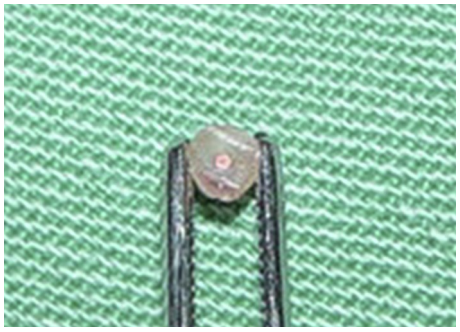

Fig. 9: *Fragment of the root apex removed after apicectomy of tooth 12.*

Fig. 10: *Calcium hydroxide powder inserted into the bone cavity.*

Fig. 11: *Mucoperiosteal flap reattached and sutured.*

The lesion was fixed in 10% formalin and sent to the Laboratory of Surgical Pathology of the School of Dentistry of the University of São Paulo. Histological sections revealed a fragment of cystic capsule formed by dense connective tissue with intense diffuse mononuclear inflammatory infiltrate and congested blood vessels of different sizes. Extravasation of red blood cells and blackish granules compatible with obturating material were also observed. The capsule was lined by stratified pavement epithelium with few layers. Further sections revealed fragment of mature bone tissue permeated by granules of obturating material. The diagnosis was radicular cyst (Figure 12).

After 10 days, the remaining sutures were removed (Figure 13). No complaints or complications were reported.

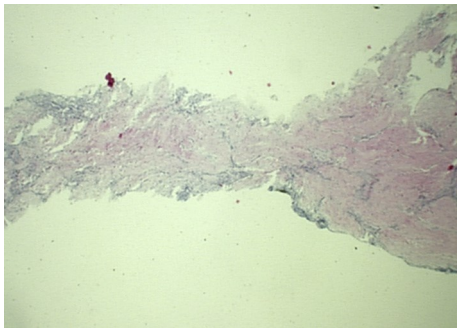

Fig. 12: *Histological aspects of the radicular cyst.*

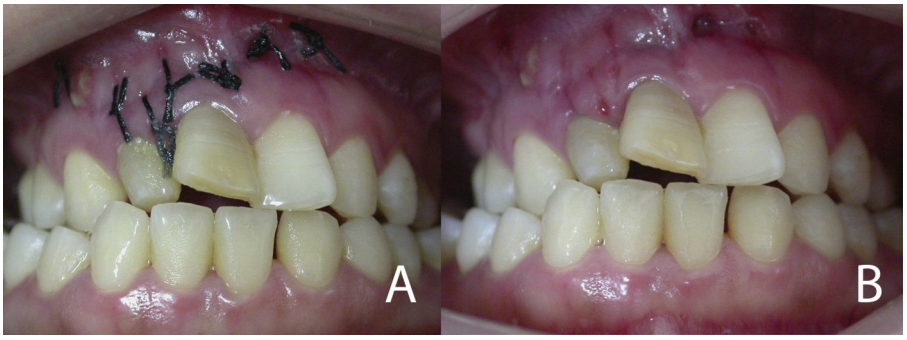

Fig. 13: *Postoperative evaluation (10 days): with remaining sutures (A); after sutures removal (B).*

After 6 months, clinical and radiographic evaluation was performed. A slight increase in radiopacity was observed at the periphery of the lesion, suggesting centripetal bone neoformation (Figure 14). After two years, a greater radiopacity was observed in the region (Figure 15). The patient has been followed-up for 14 years, showing no signs of recurrence. The periapical radiograph showed complete bone repair (Figure 16).

Fig. 14: *Radiographic evaluation after 6 months: slight increase in radiopacity observed at the periphery of the lesion.*

Fig. 15: *Greater radiopacity observed in the region after 2 years.*

Fig. 16: *Radiographic evaluation after 14 years: no signs of recurrence.*

Discussion

Cystic lesions of the bone jaw are frequently observed in dental clinic. The odontogenic cysts present high frequency, ranging from 60% to 94.5%. Most of the times, they are periapical lesions resulting from epithelial remnants stimulated by inflammatory response from pulp necrosis, which in turn are caused by dental caries. Radicular cysts are the most commonly found odontogenic cysts[5-9].

Radicular cysts are usually diagnosed during routine radiographic examination or after acute exacerbation of the infectious process[8,9], as observed in the present case. Other clinical signs such as oedema, mobility, resorption or tooth movement are caused by the slow growth of the cyst[8,9].

The cysts may evolve, regress, or remain stable. Usually, several therapeutic procedures can be performed. Endodontic treatment is instituted when there is a diagnosis of pulp necrosis. Surgical treatment can be performed using enucleation, decompression or marsupialization techniques. Decompression and marsupialization techniques are generally employed in larger cysts, but require strict cooperation from the patient, frequent visits and maintenance of hygiene of the cystic cavity, which sometimes hinders the execution of the procedure. The method of choice for smaller cysts is enucleation of the lesion, with or without the application of graft biomaterials. The simple filling of the bone cavity with blood clot is, most of the times, enough for the bone neoformation of the cavity. An increase in radiopacity suggestive of bone neoformation has been observed after cyst enucleation, on average, after 12 to 24 months. However, this time variation is due to the dimensions and nature of the cystic lesions[6-8,10,11].

Bone repair after enucleation of cystic lesions can occur. It presents some advantages such as practicality of execution, reduction of surgical time, low cost, reduced risk of post-surgical complications when accompanied by grafts[6,11]. In the present case, the patient has been followed up for 14 years without signs of recurrence or symptomatic clinical signs.

Regarding dental care, the patient with Down's Syndrome requires some care. Depending on the cognitive limitations, the treatment must be patient and mild. Due to the reduced immune response and propensity to opportunistic infections, the protocol of prophylactic antibiotic use is recommended (American Heart Association protocol)[1,2], as it was used by us. In some cases, general anesthesia can be employed in a hospital environment, but should be considered when others are inefficient for treatment[1]. Another possibility is the use of inhaled analgesia (gas mixture between nitrous oxide and oxygen)[1].

In the present report, due to the satisfactory cognitive and collaborative response, it was possible to perform all the endodontic and surgical treatments at outpatient level. Additionally, besides the special care during treatment performed in this case, there was, initially, the execution of endodontic treatment in the fused tooth. The dental fusion, known as false twinning or synodonty, is determined by the union between the dentines of one or more teeth, resulting in a reduction in the number of teeth in the dental arch. Fused teeth can present complex internal anatomy and difficulties in endodontic therapy are predictable[12], as observed in the present case.

Conclusions

Patients with Down's Syndrome present several systemic and stomatological alterations, morphological or pathological. Associated with cognitive and behavioral factors, these alterations can make the approach of the dental surgeon and the treatment to be carried out difficult. The knowledge of the dental surgeon and the dental team is imperative for the treatment and adequate conducts. Bone neoformation was observed after enucleation of the cystic lesion presented by the patient. She has been followed up for 14 years with no signs of recurrence.

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Citation: Dias VT, Di Francesco ERS, Shinohara EH, Filho LCM, Pedron IG. "Surgical Removal of a Radicular Cyst on a Fused Tooth in a Patient with Down's Syndrome: 14-years Follow-up". SVOA Dentistry 3:2 (2022) Pages 70-74.