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Erupted compound odontoma in patient with epileptic seizures - a case report

Odontoma composto erupcionado em paciente com crises convulsivas – relato de caso

Odontoma compuesto erupcionado en paciente con convulsiones – reporte de caso

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Abstract
Erupted odontomas are rarely found in the oral cavity. To the best of our knowledge there are only a few cases reported of compound erupted odontoma. Also, different pathological conditions have been related to odontoma's aetiology. This article describes a case report with review of a 17-year-old girl in investigation of Dravet syndrome, psychogenic nonepileptic seizures and psychotic disorder in whom it was identified an enamel-like tissue exposed in gingiva. The size of the lesion was around 2 mm and it was located between the right permanent mandibular lateral incisor and canine teeth. Periapical radiography revealed a radiopaque image of irregular tooth-like structures, suggesting the presence of an erupted compound odontoma. The lesion was surgically removed under infiltrative anesthesia and without the need for a flap. Four small tooth-like structures were removed, and the specimens were sent for histopathological analysis, confirming the hypothesis of compound odontoma. The procedure went without complications nor need of patient sedation. Twenty days after the surgery, there was satisfactory healing of the operated region's tissue. Odontoma's aetiology is not very well established yet, so it is unknown whether eventual epileptic seizures that occur during the period of dental formation and thus increase patient risk for accidental dental injuries could favor the development of these lesions.
Keywords: Odontoma; Odontogenic tumors; Seizures; Myoclonic epilepsies.

Resumo

Os odontomas erupcionados são raros na cavidade oral e existem poucos casos relatados de odontomas compostos erupcionados. Além disso, diferentes condições patológicas têm sido relacionadas à etiologia do odontoma. O artigo descreve um relato de caso de uma adolescente de 17 anos em investigação de síndrome de Dravet, crises não epilépticas psicogênicas e transtorno psicótico, em que foi identificado um tecido semelhante ao esmalte exposto na gengiva. O tamanho da lesão era em torno de 2 mm, localizada entre o incisivo lateral inferior permanente direito e o canino. A radiografia periapical revelou uma imagem radiopaca de estruturas semelhantes a dentes irregulares, sugerindo a presença de um odontoma composto erupcionado. A lesão foi retirada cirurgicamente sob anestesia infiltrativa, sem necessidade de retalho. Quatro pequenas estruturas semelhantes a dentes foram removidas e os espécimes foram enviados para análise histopatológica, confirmando a hipótese de odontoma composto. O procedimento transcorreu sem complicações nem necessidade de sedação do paciente. Vinte dias após a cirurgia, houve cicatrização satisfatória dos tecidos da região operada. A etiologia do odontoma ainda não está muito bem estabelecida, portanto não se sabe se eventuais crises epilépticas que ocorrem durante o período de formação dentária e aumentam o risco do paciente de lesões acidentais, poderiam favorecer o desenvolvimento de odontomas.

Palavras-chave: Odontoma; Tumores odontogênicos; Convulsões; Epilepsias mioclônicas.

Resumen

Los odontomas erupcionados son raros en la cavidad oral, hasta donde sabemos hay pocos casos reportados de odontomas erupcionados compuestos. Asimismo, diferentes condiciones patológicas se han relacionado con la etiología del odontoma. El artículo describe un reporte de caso con revisión de una joven de 17 años en investigación de síndrome de Dravet, convulsiones no epilépticas psicógenas y trastorno psicótico, se identificó un tejido similar al esmalte expuesto en la encía. El tamaño de la lesión era de alrededor de 2 mm, localizada entre el incisivo lateral mandibular permanente derecho y los dientes caninos. La radiografía periapical reveló una imagen radiopaca de estructuras similares a dientes irregulares, lo que sugiere la presencia de un odontoma compuesto en erupción. La lesión se extirpó quirúrgicamente bajo anestesia infiltrativa, sin necesidad de colgajo. Se extrajeron cuatro pequeñas estructuras similares a dientes, los especímenes se enviaron para análisis histopatológico, lo que confirmó la hipótesis de odontoma compuesto. El procedimiento se logró sin complicaciones ni necesidad de sedación del paciente. Veinte días después de la cirugía, hubo cicatrización satisfactoria del tejido de la región operada. La etiología del odontoma aún no está muy bien establecida, por lo que no se sabe si eventualmente las crisis epilépticas que ocurren durante el período de formación dental y aumentan el riesgo del paciente de sufrir lesiones dentales accidentales podrían favorecer el desarrollo de estas lesiones.

Palabras clave: Odontoma; Tumores odontogénicos; Convulsiones; Epilepsias mioclónicas.

1. Introduction

The eruption of compound odontomas in oral cavity is rarely found. Odontomas are hamartomas which account for about 22% of the odontogenic tumours. Odontomas have epithelial and mesenchymal origin. They are formed of varying amounts of enamel, dentin, and pulp tissue. Basically, they can be classified as compound and complex odontoma (Satish et al., 2011).

Different pathological conditions have been related to their aetiology such as inflammatory and/or infectious processes, local trauma, mature ameloblasts, dental lamina remnants, odontoblastic hyperactivity, alterations in the genetic component responsible for controlling dental development, hereditary anomalies like Gardner's syndrome and Hermanns syndrome. Nevertheless, the aetiology of odontomas remains unknown (Costa et al.,1990; Raval et al., 2014; Satish et al., 2011). Costa et al., (1990) report a case of permanent tooth retention in mandible caused by a compound odontoma, emphasizing the importance of early diagnosis which provides adequate treatment and limits aesthetic changes.

One of the most severe types of genetic epilepsy is the severe myoclonic epilepsy in infancy, also named Dravet syndrome. It is characterized by a variety of seizures that in most cases are resistant to treatment and affects patients who have significant cognitive impairment (Akiyama et al., 2012; Connolly, 2016). This syndrome is associated with mutations in the alpha 1 subunit gene of the sodium channel in 70-80% of individuals and is therefore considered a channelopathy (Connolly, 2016). The onset of seizures occurs during the first year of life with a mean age of 5-8 months. The first seizure is typically generalized clonic or unilateral. They are often triggered by fever, although photosensitivity may also be the cause (Akiyama et al., 2012).

On the other hand, psychogenic nonepileptic seizure (PNES) is a complex condition that may present physical or neurological symptoms similar to epilepsy, but without the neurological findings responsible for the seizures (Gomes & Gomes, 2015).

The aim of the present study was to report a case of erupted compound odontoma in the mandible of a 17-year-old girl in investigation of Dravet syndrome and psychogenic nonepileptic seizures. The clinical, radiological, and microscopic evaluation, as well as morphology assessment, clinical findings and treatment were described.

2. Case Report

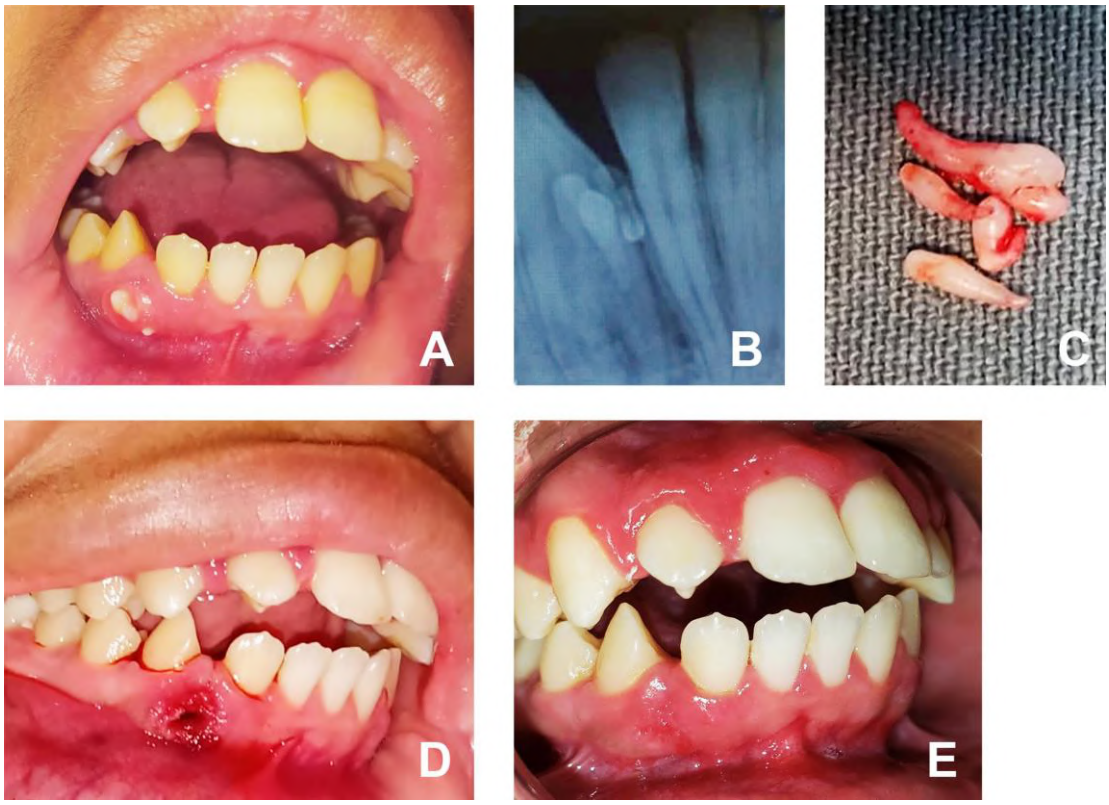
This report was approved by the Research Ethics Committee of the Clinical Hospital. A 17-year-old girl in investigation of Dravet syndrome, psychogenic nonepileptic seizures, mild intellectual disability, and psychotic disorder attended the dental clinic for a first evaluation. She has had seizures since 11 months of age and has been using antiepileptic drugs since. At the age of 16 years old she started to have PNES after more than 4 years without epileptic seizures. During the hospitalization period, the patient had one episode of seizure as well as some episodes of aggression and psychomotor agitation. She also presented mild to moderate developmental delay and intellectual disability.

She reported complaints of dissatisfaction with the aesthetics of her anterior teeth and stated that she was the victim of jokes from her friends. She reported no pain in the oral cavity. The medication she was taking at the time was haloperidol 5 mg, lamotrigine 100 mg, nitrazepam 5 mg, quetiapine 100 mg. On extraoral physical examination, the patient presented normal characteristics. On intraoral examination was observed an enamel-like tissue around 2 mm exposed in gingiva between the right permanent mandibular lateral incisor and canine teeth, an anatomical change in the superior lateral right tooth and an anterior open bite (Figure 1A). A periapical radiography of the mandibular region revealed a radiopaque image of irregular tooth-like structures, suggesting the presence of an erupted compound odontoma (Figure 1B). Considering that the patient was collaborative in all previous appointments for supragingival scraping and oral hygiene guidance, the excision of the lesion was proposed in an outpatient clinic under local anesthesia. The patient and her mother were informed about the lesion and procedure, and the free and informed consent form was signed by the mother.

First, the region was blocked with infiltrative anesthesia (one tube of Citanest 3%) in the mandibular buccal gingival region between the right canine and premolar. The lesion was accessed without the need of a flap, and a tooth-like tissue was removed (Figures 1C and 1D), then the surgical wound was curetted and cleaned with 0.9% saline solution, finally a simple suture was performed with vicryl 4-0.

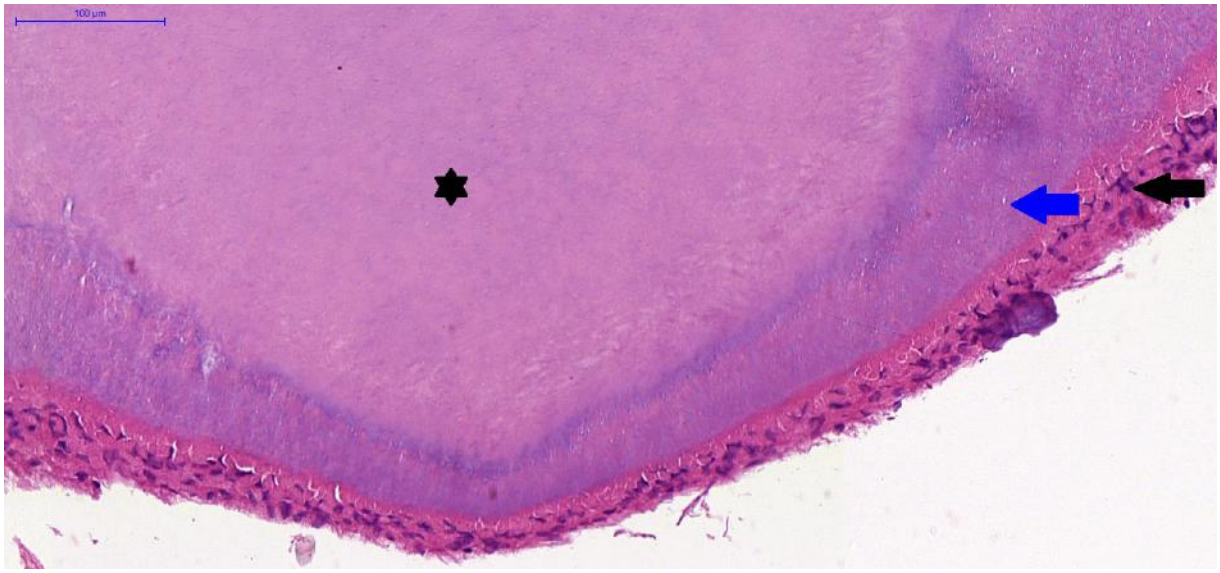
The tissue resembled four small tooth structures, and the specimens were sent for histopathological analysis (Figure 2). According to the pathological report, the diagnosis of compound odontoma was confirmed. The procedure went without complications, episodes of seizures nor need of sedation. The patient was collaborative the whole time. Twenty days after the surgery there was satisfactory healing, and the patient followed her dental and medical treatments (Figure 1E).

Figure 1 – Intraoral aspects. A - Initial clinical aspect; B - Periapical radiography; C - Lesion aspect; D - Surgical wound; E – Healing twenty days after the surgery.



Source: Authors.

Figure 2 - Histopathological image. Follicular epithelium (black arrow); enamel matrix (blue arrow); mineralized dentin (star).



Source: Authors.

3. Discussion

Patients with typical Dravet syndrome have several types of seizures during the course of the disease, including generalized tonic-clonic seizures, generalized clonic seizures, and unilateral alternating clonic seizures. These are the main

types of seizures that occur throughout the life of most patients (Connolly, 2016). Treatment consists of administration of anticonvulsants, frequently employing sodium valproate, clobazam, stiripentol, and topiramate, all of which can be beneficial. In addition, there are non-pharmacological treatments such as stimulation of the vagus nerve, surgical procedures and even the adoption of a ketogenic diet (Jacobsen & Eden, 2008; Connolly, 2016; Ohshita et al., 2019)

PNES can be confused with epilepsy due to similarities in the behavioral manifestations, but the former is not a consequence of abnormal brain discharges and may have a physiological or psychogenic origin (Gomes & Gomes, 2015). In some cases, PNES and epilepsy can coexist in between 8% and 60% of patients (Kutlubaev et al., 2018). During a PNES the electroencephalogram is normal. While some epileptic seizures present a stereotyped pattern of motor movements with loss of consciousness, a PNES varies from one seizure to another without loss of consciousness, although with changes in mental status. This differs from epileptic seizures in which case there is a characteristic electroencephalographic pattern with paroxysmal brain bioelectric changes (Baroni et al., 2016; Kutlubaev et al., 2018; Asadi-Pooya, 2020).

Generally speaking, patients with seizures tend to have poor oral health. This occurs mainly because patients with poorly controlled seizures tend to have a low socioeconomic level, as they suffer from prejudice and face difficulties in entering the job market (Károlyházy et al., 2003; Bryan & Sullivan, 2006). Specific considerations for these patients include management of medication side effects in the oral cavity and rehabilitation of the damage inflicted to the orofacial region's hard and soft tissues due to seizure trauma, particularly in patients suffering from poorly controlled generalized disorders such as tonic-clonic seizures (Jacobsen & Eden, 2008).

Odontomas are the most prevalent odontogenic tumours and mostly observed in the second and third decades of life. There is no observed tendency towards either sex or to any anatomical site in the oral cavity. However, odontomas are most frequently located in the anterior zone of the superior maxilla (50%), followed by the anterior region of the mandible (15%) and have a size inferior to 10 mm (Levi-Duque & Ardila 2019).

Exposure of the odontoma on the oral mucosa is the exception. To the best of our knowledge there are only a few cases reported of compound erupted odontoma. The mechanism of odontoma eruption is different from tooth eruption due to the lack of periodontal ligament and root in this tumour. Therefore, the force required to move the odontoma is not linked to the contractility of fibroblasts, as is the case with teeth. Although there is no root formation in the odontoma, its increase in size can lead to sequestration of the overlying bone and therefore cause occlusal movement or delay in eruption (Verma et al., 2015). Odontomas are benign in nature, but the eruption in the oral cavity can cause pain, inflammation, and infection, in addition to other clinical manifestations (Mehta et al., 2013; Raval et al., 2014; Verma et al., 2015). Similar to the present case, Ohtake K, et al., (1993) report a case of three complex odontomas associated with ossifying fibroma of the right mandible in a 10-year-old boy with epilepsy as only comorbidity.

In patients with seizures, it is important to be aware of the occurrence of dental trauma which is increased in epileptic patients (Moreira Falci et al., 2019; Salas-Puig et al., 2019). In addition, dental appointments should be scheduled for a time when seizures are less likely to occur as to minimize stress and anxiety during the treatment (Károlyházy et al., 2003; Robbins, 2009). If a seizure occurs at the time of dental care, the procedure should be stopped immediately, all instruments should be removed from the oral cavity, the chair should be placed in the supine position and the patient should be stabilized to avoid injury to themselves during the crisis. However, if the seizures continue for more than five minutes, there is difficult breathing, persistence of mental confusion or physical injuries, medical care should be provided (Aragon & Burneo, 2007; Jacobsen & Eden, 2008).

4. Final Considerations

This report of a girl with a history of epileptic seizures presenting compound odontoma highlights the importance of early diagnosis and treatment. Odontoma's aetiology is not very well established yet, so it is unknown whether eventual epileptic seizures that may occur during the period of dental formation and increase patient risk for accidental dental injuries could favour the development of these lesions.

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