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CASE REPORT

Oral management in a male pediatric patient with Doose Syndrome. Case Report

Manejo estomatológico en paciente pediátrico masculino con Síndrome de Doose. Reporte de Caso

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ABSTRACT

Introduction: Doose syndrome, also known as myoclonus-astatic syndrome, is a rare form of epilepsy that usually presents in childhood and is characterized by myoclonic and drop or startled seizures, along with delayed psychomotor development. Dental management in patients with Doose syndrome requires a comprehensive approach, taking into account both the medical and psychological and social implications of the patient. This can influence the patient's ability to maintain postural stability, which could increase the risk of falls or accidents during dental treatment, as well as anticonvulsant treatment, which is common in these patients, since many of these drugs can have side effects related to oral health, such as xerostomia (dry mouth), which can predispose to dental caries and oral infections.
Clinical Case: a four-year-old male patient, diagnosed with Doose syndrome, presents to the pediatric dentist's office with the purpose of receiving dental treatment and rehabilitation. The patient was seen after being previously referred to a neurology specialist, and a comprehensive treatment plan encompassing various strategies was implemented. This approach consisted of performing necessary surgical procedures and providing continuous follow-up for a period of 12 months, taking into account both his physical health and his psychological and social status. Thanks to this collaborative effort, the factors that could trigger seizure episodes were significantly reduced. As a result, a notable decrease in plaque accumulation was observed, the patient's oral cavity remained caries-free, and he better adapted to dental visits.
Conclusion: the patient's rehabilitation was successful without any complications throughout the process. The procedure related to his management was subjected to further analysis, in which behavioral control measures were implemented, and was performed under local anesthesia. A notable improvement in both the patient's oral and general health was observed, evidenced by the increase in his weight and height during the follow-up visit, which took place six months after the initial evaluation.

Keywords: Pediatric dentistry; Neurology; Seizures; Epilepsy.

RESUMEN

Introducción: el síndrome de Doose, también conocido como síndrome de mioclonías-astáticas, es una forma rara de epilepsia que se presenta generalmente en la infancia y se caracteriza por crisis mioclónicas y de caída o atónitas, junto con un desarrollo psicomotor retrasado. El manejo odontológico en pacientes con síndrome de Doose requiere un enfoque integral, tomando en cuenta tanto las implicaciones médicas como las psicológicas y sociales del paciente. Esto puede influir en la capacidad del paciente para mantener la estabilidad postural, lo que podría aumentar el riesgo de caídas o accidentes durante el tratamiento odontológico, así como también el tratamiento anticonvulsivo, que es común en estos pacientes, ya que

muchos de estos fármacos pueden tener efectos secundarios relacionados con la salud bucal, como xerostomía (boca seca), que puede predisponer a la caries dental y a infecciones orales.
Caso Clínico: un paciente masculino de cuatro años de edad, que ha sido diagnosticado con el Síndrome de Doose, se presenta en la consulta odontopediatra con el propósito de recibir tratamiento y rehabilitación dental. El paciente fue atendido después de haber sido referido previamente a un especialista en neurología, y se implementó un tratamiento integral que abarcó diversas estrategias. Este enfoque consistió en llevar a cabo procedimientos operatorios necesarios y proporcionar un seguimiento continuo durante un periodo de 12 meses, teniendo en cuenta tanto su salud física como su estado psicológico y social. Gracias a este esfuerzo conjunto, se logró reducir significativamente los factores que podían desencadenar episodios de crisis convulsivas. Como resultado, se observó una notable disminución en la acumulación de placa dentobacteriana, se mantuvo cavidad bucal del paciente libre de caries, y se facilitó una mejor adaptación a las visitas al dentista.
Conclusión: la rehabilitación del paciente se llevó a cabo de manera exitosa y sin la aparición de ninguna complicación durante todo el proceso. El procedimiento relacionado con su manejo fue sometido a un nuevo análisis, en el cual se implementaron medidas para controlar su conducta, y se realizó bajo condiciones de anestesia local. Se observó una notable mejora en la salud tanto bucal como general del paciente, evidenciada por el aumento en su peso y altura durante la consulta de control que se realizó seis meses después de la evaluación inicial.

Palabras clave: Odontopediatría; Neurología; Convulsiones; Epilepsia.

INTRODUCTION

Doose syndrome is a rare neurological disorder that mainly affects young children. It is characterized by epileptic seizures that include myoclonus (sudden, rapid muscle movements) and sudden falls due to atonic seizures (loss of muscle tone). This syndrome is considered a difficult-to-manage form of epilepsy and is associated with neuropsychological developmental delay, which affects patients' quality of life.[1]

Although Doose syndrome does not have a clearly identified genetic cause, a predisposition is identified in mutations of certain genes, but most cases do not have a directly established genetic cause.[2]

The prevalence of this specific condition is estimated to be between 1 and 2 percent of all cases of epilepsy in children, with a ratio of approximately 2,7 males to 3,1 females affected.[3]

The World Health Organisation recognizes epilepsy as a significant health problem affecting around 50 000 000 people, with nearly 80 % of patients living in low- and middle-income countries. The annual incidence in developed countries is approximately 50 per 100 000 inhabitants of the general population, with children being particularly prone.[4]

The condition typically appears in children between the ages of 2 and 5, peaking around 3 to 4 years of age. It is important to note that this phenomenon usually occurs in infants with normal and expected development up to that point. Twenty percent of individuals undergoing medical treatment experience episodes of simple febrile seizures before the appropriate care protocol is initiated. Generalized tonic-clonic seizures, which are a type of seizure that affects the entire brain, are often the first type of epileptic seizure to occur in many patients with epilepsy. Over time, which can range from a few days to several months, additional episodes of seizures may occur, which may be myoclonic, astatic, or even a combination of both, along with episodes of atypical absences.[4] These events occur with increasing frequency until a critical point is reached, described as a 'stormy phase.' MA seizures are characterized by the onset of myoclonic jerks, which are sudden muscle contractions, specifically in the proximal muscles of the body. These contractions are followed by a phase in which the muscles lack tone or atonia, temporarily losing their normal tension.[5] In certain patients, brief tonic muscle contractions may be observed, manifesting as seizures. Children often tend to experience episodes of a seizure state that does not involve convulsions, manifesting through episodes of drowsiness, changes in gait, and erratic myoclonic seizures. These episodes can last for a period ranging from several hours to even entire days.

This syndrome is considered a difficult-to-manage form of epilepsy and is associated with neuropsychological developmental delay, affecting patients' quality of life.

The typical onset of Doose syndrome occurs between the ages of 1 and 5, although the age of onset may vary, and it presents gradually.[6]

The first signs usually include psychomotor development disorders, with delays in motor control and language.

Myoclonic seizures are one of the most common symptoms. They are characterized by sudden, jerky muscle movements that are usually short-lived and mainly affect the arms and head.[3]

Atonic or drop seizures are another characteristic finding and are caused by a temporary loss of muscle tone, which can lead to unexpected falls.[7]

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Various factors, such as stress, fever, sleep deprivation, or metabolic factors, can trigger the severity and frequency of seizures.[8]

Cognitive delay (social and emotional behavior) is common for neuropsychological developmental disorders.[9]

The diagnosis of Doose syndrome is clinical and based on observing symptoms that appear progressively throughout the medical history, corroborating the diagnosis with various tests and complementary examinations.[7]

In most cases, the underlying cause, also known as the etiology, remains unidentified, and it is suspected that a polygenic inheritance component may be involved in its origin. The causes that have been identified as monogenic include various genetic variants, among which are those corresponding to the SLC6A1 gene located in the 3p25.3 region of the chromosome, variants of the CHD2 gene located in 15q26.1, and the AP2M1 gene located in 10q23.2.

Although some children show a clinical course that could be considered favorable, it is essential to note that epileptic seizures may not respond adequately to anticonvulsant drug treatments, commonly known as AEDs, initially. The prescription of broad-spectrum antiepileptic drugs is strongly recommended, as the clinical situation of many patients often requires the implementation of a treatment strategy combining two or three different drugs. Recommended drugs include sodium valproate, which can be very effective, and its use in combination with lamotrigine, where a synergistic effect is observed that can enhance therapeutic results. In addition, other drugs that may be considered in this approach are levetiracetam and ethosuximide, especially in cases where myoclonic seizures are particularly prominent and concerning. There are additional options that can be considered, including drugs such as zonisamide, topiramate, clobazam, and clonazepam. The ketogenic diet is the most effective therapeutic intervention for the treatment of myoclonic-astatic epilepsy, achieving a reduction of more than 50 % in seizure frequency in around 50 % to 90 % of patients who follow it. This type of diet is especially recommended in cases where first-line antiepileptic drugs, commonly referred to as MACs, have failed to provide the necessary control over the disease.

The management of Doose syndrome focuses mainly on controlling seizures and improving the patient's quality of life. As this is a complex disorder to control, treatment often involves a combination of pharmacological therapies (antiepileptic drugs associated with xerostomia) and non-pharmacological therapies (educational, occupational, physiotherapy, and speech support therapies).[10]

Additional considerations and continuous monitoring for safety must be applied for the psychosocial intervention of pediatric patients.[3]

The prognosis for patients with Doose syndrome is variable; however, in the long term, it will depend on several factors, such as seizure control, early intervention, and educational support.

CASE PRESENTATION

A male patient, 4 years old, was diagnosed with Doose syndrome (myoclonic-astatic epilepsy) when he was 2 years and 6 months old. He presents psychomotor retardation and myoclonic and atonic seizures, which are partially controlled with anticonvulsant treatment (valproate and lamotrigine). The child is nonverbal, has difficulty coordinating movements, and shows limited motor skills.

Reason for consultation: The mother came to the dental clinic because she was concerned about advanced tooth decay in several of the child's deciduous teeth and oral hygiene problems due to the child's limited ability to perform dental hygiene independently.

The child was admitted to the pediatric dentistry service, where a physical examination was performed. There were no apparent signs of pathology in the skin or integuments at the level of the stomatognathic system.

Patient interview (medical and dental history): Clinical examination, Assessment of diagnostic records (study models, photographs, X-rays such as orthopantomograms).

Vital signs: Heart rate 87 beats per minute, oxygen saturation 97 % with O2 21 %, blood pressure 110/72 mmHg, respiratory rate 20 per minute, and axillary temperature 36,8 °C.

An asthenic patient with an Angle Class II intermaxillary relationship, clinical and radiographic examination revealed developing primary dentition, a bilateral mesial terminal step, bilateral Class I canines, a diastema arch (Baumé Type I), rampant caries, bacterial plaque, gingivitis, and xerostomia.

Medical conditions (neurological): Doose syndrome.
Allergic history: none reported
Surgical history: none reported
Family history: none reported
Diet: 3 times a day
Food intolerances: none
Bowel movements: 2 times a day
Urination: 4 times a day
Sleep: 8 hours per day

The analysis and evaluation of oral health revealed anterior open bite, early childhood caries, gingivitis, a

significant level of xerostomia, angular cheilitis on the left side, and mouth breathing (figures 1-2 A-B).

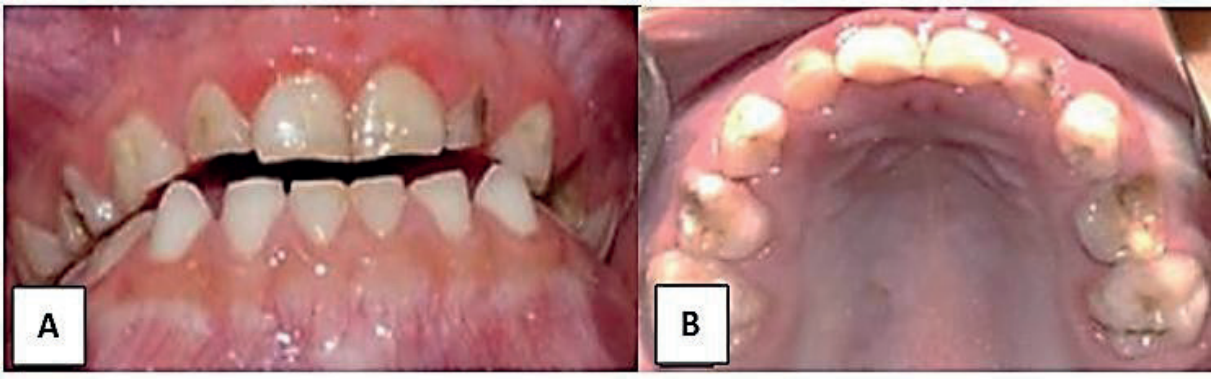

Figure 1. Oral health analysis and assessment

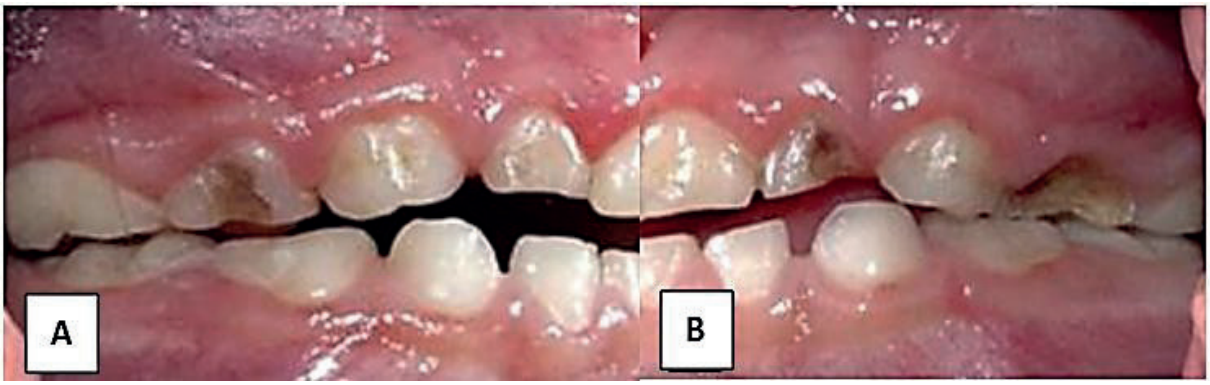

Figure 2. Oral health analysis and assessment

The treating pediatric neurologist requested A report and authorization to receive adequate dental care. The report stated that the patient was receiving pharmacological treatment with clobazam at a dose of 5 mg every 12 hours, valproic acid at a dose of 1,2 ml every 8 hours, and topiramate at a dose of 50 mg every 12 hours. This medication regimen has caused side effects such as xerostomia (dry mouth syndrome), gingivitis, and dental caries.

To provide adequate care, the process was carried out in stages:

First stage—preventive: The patient's behavior was managed using strategies such as desensitization and triple E. Once the patient had been conditioned, efforts were made to reinforce proper toothbrushing techniques and implement healthy habits with the parents.

After the diagnosis and treatment plan, prophylaxis was carried out with a pumice stone, a toothbrush, a prophylactic cup, and fluoridation.

Second stage – Rehabilitation: caries inactivation, restorations, pulpectomies, and pulpotomies were performed.

During each appointment, meticulous biofilm control was carried out, and the prophylaxis procedure was performed to ensure the patient's dental health. An anesthetic with a vasoconstrictor, such as lidocaine in a concentration of 2 %, was used and performed in an environment of absolute isolation to ensure maximum effectiveness and safety during the procedure.

DISCUSSION

This case presents the multiple oral complications in a pediatric patient with myoclonic-astatic epilepsy (Doose syndrome), in whom neurological, pharmacological, and functional factors coexist that affect the maintenance of adequate oral health. The use of broad-spectrum anticonvulsants and barriers to daily hygiene aggravates the condition.

Dental caries

The findings of rampant caries are consistent with those reported in pediatric populations with epilepsy, where the prevalence of caries is higher due to the use of liquid medications containing sucrose and difficulties in oral hygiene. Goyal et al. demonstrated that epileptic children taking liquid medications have a higher prevalence of caries (76,1 %) compared to those who do not (55,6 %).[12] Similarly, Yeung et al. found that children

treated with multiple antiepileptic drugs had a higher prevalence of caries and worse gingival conditions than those on monotherapy. Morgan et al. confirmed these observations, showing a higher caries index in primary dentition and greater gingival problems in epileptic children compared to healthy controls.[13]

Xerostomia

In the present case, xerostomia was a relevant clinical manifestation, probably induced by the pharmacological regimen based on valproate, clobazam, and topiramate. The literature shows that this side effect may be directly related to the prolonged use of antiepileptic drugs, negatively affecting the oral health of pediatric patients.[14,15]

Gidal describes how physiological changes affecting drug absorption, including xerostomia, are familiar with antiepileptic drugs and should be considered for their impact on oral administration and mucosal health.[16] Although his study focuses on older adults, the mechanisms involved can be extrapolated to the pediatric population under chronic therapy.[17]

Torres and Elizabeth also warn that the adverse effects of antiepileptic drugs include xerostomia, ulcerations, glossitis, and an increased incidence of caries, which must be carefully considered during the dental treatment of these patients.[16]

Bereda also points out that, among the typical adverse reactions to antiepileptic drugs, xerostomia appears as a significant consequence that can alter the patient's quality of life and long-term oral health.

These findings support the need for active surveillance and preventive management of dry mouth in pediatric patients with epilepsy to mitigate oral complications such as caries, infections, and functional discomfort.

These findings reaffirm the need to monitor not only the neurological condition but also the oral effects associated with prolonged treatment to prevent complications such as rampant caries, which compromise both the oral health and quality of life of pediatric patients with epilepsy.

Drugs

The patient's drug regimen, based on valproate, clobazam, and topiramate, has been documented in the literature as being associated with various adverse oral effects. Prolonged exposure to antiepileptic drugs is related not only to the systemic impacts but also to oral manifestations that can compromise the quality of life of pediatric patients.[17]

Ghafoor et al. conducted a study of 150 epileptic children. They found that gingival growth was the most common side effect associated with the use of antiepileptic drugs, along with soft tissue lesions such as lip and cheek biting, as well as dental fractures resulting from trauma during seizures.[18]

Joshi et al. compared oral hygiene and the presence of gingival enlargement in epileptic and healthy children, finding that medicated epileptic children had poorer oral hygiene and a higher prevalence of gingival hyperplasia, with valproate being the drug most commonly associated with this finding.[19]

Lundström et al. analyzed the effect of carbamazepine and phenytoin on the oral health of pediatric patients. They concluded that those treated with phenytoin had greater gingival probing depths, lower salivary secretion rates, and lower buffering capacity, which increased susceptibility to periodontal disease and caries.[9]

These studies highlight the need for continuous monitoring of the oral environment in patients undergoing prolonged antiepileptic treatment and individualized preventive and therapeutic dental strategies.

Diet

Although the patient in question does not follow a ketogenic diet, this type of regimen is frequently used as an alternative therapy in refractory epilepsies such as Doose syndrome. This diet, high in fat and low in carbohydrates, induces a state of ketosis that has been linked to a decrease in the frequency of epileptic seizures. However, its impact on oral health must be carefully monitored.[18]

Sharma and Mathur highlighted that the biochemical changes induced by the ketogenic diet can negatively affect oral health by altering the oral pH and saliva composition, increasing the risk of caries and tooth erosion.[20]

Complementarily, Coppola et al. reviewed the available evidence and emphasized that, although the ketogenic diet offers anticonvulsant benefits, it can also have side effects that include digestive disorders, nutritional deficiencies, and dental problems if its implementation is not monitored correctly.[21]

Weijenberg et al., in an observational study of children who started a liquid ketogenic diet, confirmed its clinical efficacy but warned of the need for constant monitoring to ensure complete nutritional intake and prevent adverse effects, including those related to oral health.[22] These findings reinforce the importance of integrating dentists into the multidisciplinary team that manages epileptic patients on a ketogenic diet to prevent dental demineralization, oral dysbiosis, and caries.

CONCLUSION

The elements analyzed in this clinical case highlight the importance of a detailed medical history, especially

in pediatric patients with complex neurological conditions such as Doose syndrome. This approach allows not only for the thorough recording of the patient's systemic status but also for the establishment of clinical correlations between the types of epileptic seizures, their possible triggers, and the associated oral manifestations.

The comprehensive dental approach allowed us to identify risk factors such as chronic medication with antiepileptics (valproate, clobazam, and topiramate), whose impact on oral health has been widely documented in the scientific literature due to their association with gingival hyperplasia, xerostomia, reduced salivary pH and, consequently, increased susceptibility to caries and periodontal disease. These adverse effects highlight the need for continuous monitoring by the dentist during the treatment of epileptic patients.

Likewise, the importance of considering diet in these patients was evident. Although the patient evaluated does not follow a ketogenic diet, this therapeutic alternative is common in refractory epilepsies such as Doose's.

Implementing it can alter the oral balance, increasing the risk of dental erosion; therefore, interdisciplinary monitoring is recommended. The stomatological management was successful, characterized by the absence of seizures during treatment, a positive adaptation of the patient to dental consultations, and a notable improvement in his oral health, evidenced by the lack of carious lesions and a healthy periodontal and mucosal status.

These results reinforce the effectiveness of a preventive and personalized approach and highlight the importance of collaboration between dentists, neurologists, nutritionists, and carers to achieve comprehensive and safe control in pediatric patients with epilepsy.

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CONSENT

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CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

AUTHOR CONTRIBUTION

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