Arch Health Invest (2025)14(12):2733-2738
http://doi.org/10.21270/archi.v14i12.6656
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Clinical case

Multidisciplinary Approach for Dental Management of Pediatric Patient with DiGeorge Syndrome and Tetralogy of Fallot: A Case Report

Abordagem Multidisciplinar para Manejo Odontológico de um Paciente Pediátrico com Síndrome de DiGeorge e Tetralogia de Fallot: Relato de Caso

Abordaje Multidisciplinario para el Manejo Dental de un Paciente Pediátrico con Síndrome de DiGeorge y Tetralogía de Fallot: Reporte de un Caso

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Abstract
Patients with congenital heart disease (CHD), particularly those with associated syndromes such as DiGeorge Syndrome (DGS), present unique challenges in dental management due to systemic and oral health complexities. DGS is a genetic condition that affects multiple systems, including craniofacial and dental structures, often leading to enamel defects and an increased risk of dental caries. Tetralogy of Fallot (ToF), a common CHD, further compounds these challenges due to the heightened risk of bacterial endocarditis associated with oral infections. This case report details the dental management of a 7-year-old boy with both DGS and repaired ToF. The patient was presented with severe dental caries, poor oral hygiene, and non-collaborative behavior, necessitating treatment under general anesthesia. A multidisciplinary team, including cardiologists, anesthesiologists, and oral surgeons, coordinated care to ensure a safe and effective approach. Multiple extractions were performed under antibiotic prophylaxis to minimize the risk of infection and systemic complications. Post-operative recovery was uneventful, and follow-up revealed satisfactory healing and improved oral care adherence. This report highlights the importance of a multidisciplinary strategy in managing dental treatment for medically complex pediatric patients. Early diagnosis, preventive measures, and collaboration among healthcare providers are crucial to optimizing oral and systemic health outcomes while minimizing risks.
Descriptors: Pediatric Dentistry; Multidisciplinary; Tetralogy of Fallot; Digeorge Syndrome; Heart Defects, Congenital.
Resumo
Pacientes com cardiopatia congênita (CC), particularmente aqueles com síndromes associadas, como a Síndrome de DiGeorge (SDG), apresentam desafios singulares no manejo odontológico devido às complexidades sistêmicas e orais. A SDG é uma condição genética que afeta múltiplos sistemas, incluindo estruturas craniofaciais e dentárias, frequentemente resultando em defeitos de esmalte e aumento do risco de cárie dentária. A Tetralogia de Fallot (TF), uma cardiopatia congênita comum, agrava ainda mais essas dificuldades em razão do risco elevado de endocardite bacteriana associada a infecções orais. O presente relato descreve o manejo odontológico de um paciente do sexo masculino, de 7 anos de idade, portador de SDG e TF previamente corrigida. O paciente apresentava cárie dentária extensa, higiene oral deficiente e comportamento pouco colaborativo, o que tornou necessário o tratamento sob anestesia geral. Uma equipe multidisciplinar, composta por cardiologistas, anestesiologistas e cirurgiões bucomaxilofaciais, coordenou o cuidado a fim de garantir uma abordagem segura e eficaz. Exodontias múltiplas foram realizadas sob profilaxia antibiótica para reduzir o risco de infecção e de complicações sistêmicas. A recuperação pós-operatória ocorreu sem intercorrências, e o acompanhamento demonstrou cicatrização satisfatória e melhor adesão aos cuidados orais. Este relato ressalta a importância de uma estratégia multidisciplinar no tratamento odontológico de pacientes pediátricos com condições médicas complexas. O diagnóstico precoce, as medidas preventivas e a colaboração entre profissionais de saúde são fundamentais para otimizar os desfechos orais e sistêmicos, minimizando riscos.
Descritores: Odontopediatria; Tetralogia de Fallot; Síndrome de Digeorge; Cardiopatias Congênitas.
Resumen
Los pacientes con cardiopatías congénitas (CHD, por sus siglas en inglés), particularmente aquellos con síndromes asociados como el síndrome de DiGeorge (DGS), presentan desafíos únicos en el manejo odontológico debido a las complejidades de la salud sistémica y oral. El DGS es una condición genética que afecta múltiples sistemas, incluyendo las estructuras craneofaciales y dentales, lo que a menudo conduce a defectos del esmalte y a un mayor riesgo de caries dental. La Tetralogía de Fallot (ToF), una CHD común, agrava aún más estos desafíos debido al mayor riesgo de endocarditis bacteriana asociada con infecciones orales. Este informe de caso detalla el manejo odontológico de un niño de 7 años con DGS y ToF reparada. El paciente presentaba caries dentales severas, mala higiene oral y un comportamiento poco colaborativo, lo que requirió tratamiento bajo anestesia general. Un equipo multidisciplinario, que incluía cardiólogos, anestesiólogos y cirujanos orales, coordinó la atención para garantizar un enfoque seguro y eficaz. Se realizaron múltiples extracciones bajo profilaxis antibiótica para minimizar el riesgo de infección y complicaciones sistémicas. La recuperación postoperatoria fue sin incidentes, y el seguimiento mostró una cicatrización satisfactoria y una mejor adherencia al cuidado oral. Este informe destaca la importancia de una estrategia multidisciplinaria en el manejo del tratamiento odontológico para pacientes pediátricos médicamente complejos. El diagnóstico temprano, las medidas preventivas y la colaboración entre los proveedores de atención médica son fundamentales para optimizar los resultados en la salud oral y sistémica mientras se minimizan los riesgos.
Descriptores: Odontología Pediátrica; Tetralogía de Fallot; Síndrome de DiGeorge; Cardiopatía Congénita.

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INTRODUCTION

The dental management of patients with congenital heart disease is an increasingly significant area of interest in the healthcare field, given the complexity of this condition[1]. Patients with genetic disorders often present specific oral manifestations and a higher predisposition to dental problems, while those with congenital heart disease (CHD) are at an increased risk of bacterial endocarditis, a potentially fatal condition that can be precipitated by oral infections[1]. The dental care provider plays a crucial role in educating these patients and guardians about the importance of good oral hygiene for both oral and overall health. In most cases, these patients often present with dental problems that, due to their systemic medical conditions, can be challenging to manage both medically and dentally[2].

The DiGeorge Syndrome (DGS), also known as Chromosome22q11.2 deletion syndrome (22q11.2 DS) is a multifaceted syndrome, caused by the impaired development of structures arising from the third and fourth pharyngeal pouches during the germinal stage[3-5]. Individuals with DGS may present vast clinical features such as hypoparathyroidism, hypocalcemia, thymic hypoplasia (or, in rare cases, thymic agenesis), conotruncal heart defects, facial dysmorphism, and palatoschisis. This complex phenotype exhibits considerable variability, affecting craniofacial, neurological, cognitive, behavioral, ocular, speech and hearing, musculoskeletal systems, and internal organs[4,5]. Dental and craniofacial characteristics are common in most individuals with 22q11.2DS[6]. Individuals with this syndrome display various dental characteristics, including abnormalities in tooth shape, eruption, and number, as well as enamel defects such as hypomineralisation and hypoplasia. These oral manifestations also include a high prevalence of dental caries[6]. Given these features, dentists must be aware of the dental aspects of this condition to refer patients to appropriate specialists when necessary. Early detection enables dentists to initiate preventive programs and reduce the risk of dental diseases[6]. In counterpart, it is not uncommon to find patients with less severe symptoms, but added up these symptoms cause substantial disability[7]. The incidence of DGS is approximately 1:4000 live births according to previous studies stage[3,4,5,7,8]. Although some cases result from autosomal dominant inheritance, 90% of DiGeorge syndrome cases are *de novo* mutations.

Tetralogy of Fallot (ToF) is the most prevalent type of CHD, occurring in approximately 3,000 per one million births. The etiology of ToF remains unclear, but it has been associated to defects that occur in intrauterine life, from week 3 to 8[9,10]. It is characterized by four clinical components, 1) perimembranous ventricular septal defect, 2) right ventricular outflow tract obstruction, 3) an overriding aortic root, and 4) right ventricular hypertrophy[10]. These defects collectively reduce the oxygen level in the blood, leading to a bluish discoloration (cyanosis) when oxygen-poor blood is circulated throughout the body, particularly affecting the fingers, toes, and lips. Children with ToF often experience easy fatigability, shortness of breath, and hyperpnea due to hypoxia[10].

Children with Tetralogy of Fallot (ToF), similar to those with DiGeorge Syndrome (DSG), are predisposed to developing potentially fatal complications, such as bacterial endocarditis. Within the scope of dentistry, bacterial endocarditis is strongly associated with dental infections, which can be worsened by poor dental hygiene, in association with enamel development defects, commonly seen in children with ToF[11]. In addition, there is an increased risk of developing caries, particularly in primary dentition due the medications for heart diseases that contain high concentrations of sugar. Preventive measures include dietary counseling, fluoride treatment, application of pit and fissure sealants, and instruction on oral hygiene practices to the parents[12].

Approximately 21% of Tof have been associated with other syndromes, like Down syndrome, Noonan syndrome and DiGeorge syndrome. Fortunately, due to medical advance in the last decades, >80% of affected children reach adulthood. However, with respect to oral health, these children have a negative impact [8,12]. Studies on the prevalence of dental caries in children with CHD have shown variable results[13]. Hallet et al.[14] reported higher levels of caries in affected children compared to their healthy siblings. Similar findings were observed in studies conducted with preschool participants in Sweden and Brazil[15,16]. Additionally, significantly higher levels of salivary Streptococcus mutans and caries rates were found in affected patients in Iran[17]. In counterpart, a scoping review revealed that patients with CHD often experience a higher incidence of untreated caries, delays in receiving treatment, and a lower standard of dental care, which may lead to multiple extractions of primary teeth, possibly requiring the use of general anesthesia[9]. Therefore, it is crucial that these patients are seen regularly by the dentist in order to prevent such conditions[12]. Considering that patients with congenital heart diseases (CHD) and Tetralogy of Fallot (ToF) may present significant dental development defects, which facilitate and/or exacerbate the occurrence of oral problems, and that these oral problems can severely compromise the overall health status of the patient, a multidisciplinary approach is essential to ensure successful treatment. Collaboration among cardiologists, dentists, pediatricians, and other

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healthcare professionals is fundamental in developing an integrated and effective treatment plan. This approach not only aims to prevent and adequately treat oral diseases but also ensures continuous monitoring of the patient's overall health, promoting early and effective interventions that can prevent more severe complications and improve the quality of life for these individuals Thus, the aim of this article is to report a multidisciplinary approach for dental management of a pediatric patient diagnosed with DiGeorge Syndrome and Tetralogy of Fallot in a hospital setting.

CASE REPORT

A 7-year-old boy was referred to our oral and maxillofacial surgery service by his dentist for multiple teeth extractions due to extremely poor oral health condition. According to the dentist, the patient was uncollaborative and would not allow the procedure to be made at clinical practice. The patient medical history included a diagnosis of Tetralogy of Fallot, with anterior crossing of the pulmonary arteries and DiGeorge Syndrome. A previous medical report indicated that in 2017, the patient underwent complete repair of Tetralogy of Fallot (ventricular septoplasty with a bovine pericardial patch, right ventricular myectomy, commissurotomy of a bicuspidized pulmonary valve with a large anterior leaflet and a smaller posterior leaflet, while preserving the pulmonary annulus). During the surgery, the absence of the thymus was noted, a characteristic feature of DiGeorge Syndrome. The procedure was performed without complications, with satisfactory clinical progress and good acceptance of diet. A follow-up echocardiogram performed 8 days post-surgery yielded satisfactory results. The patient was discharged in good general condition, with a regular heart rhythm, cardiac auscultation revealing normal heart sounds in two phases with a systolic murmur at the high left sternal border graded 2+/4+, clear pulmonary auscultation, and an unremarkable abdomen. The prescribed medications included furosemide at 2 mg/kg/day, propranolol at 1 mg/kg/day, and Noripurum drops. In addition, the patient was cleared by his cardiologist with no contraindications for undergoing the dental surgical procedure, however the patient should receive antibiotic prophylaxis 2 hours before the surgical procedure, with a dose of 50 mg/kg of amoxicillin.

On extra oral examination, there was no alterations. Intraoral, an extremely poor oral hygiene was observed, and large caries lesions were present throughout the mouth. It was clear that extractions of multiple teeth in all quadrants would be necessary (Figure 1). Despite these findings, there was no report of oral pain at the admission. The panoramic radiograph showed several extremely carious teeth, the presence of root remnants, and developing permanent tooth buds, with no abnormalities detected (Figure 2). Based on these findings and given the non-collaborative profile of the patient and his medical condition, treatment under general anesthesia was planned, and a consent for the procedure was signed by the mother.

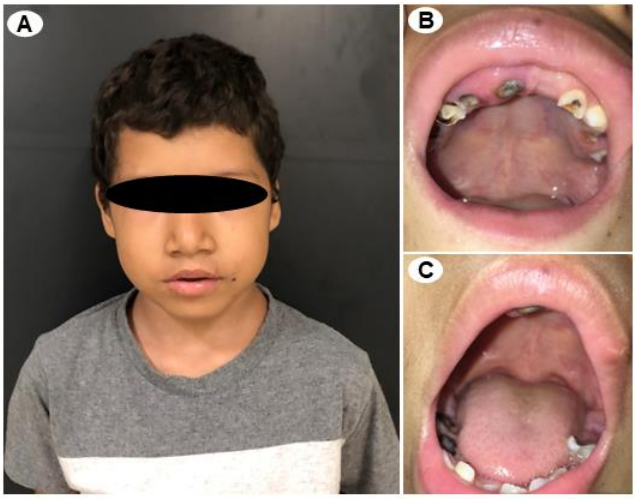

Figure 1A: Frontal photograph of the patient in a natural head position. No alterations or edema are observed on the face. Figure 1B: Intraoral view of the maxillary arch and the mandibular arch (Figure 1C), highlighting the presence of extensive carious lesions and retained roots, associated with poor oral hygiene.

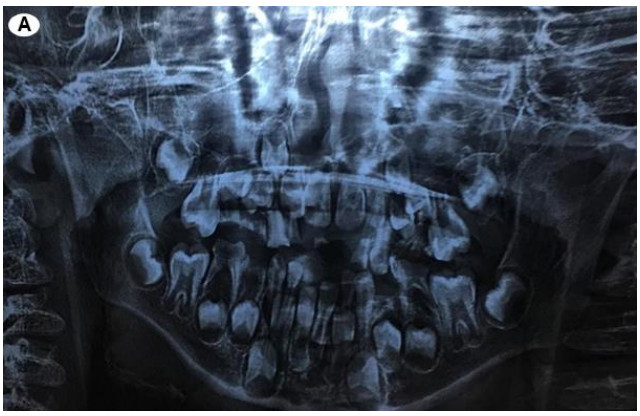

Figure 2: Panoramic radiograph of the patient, showing coronal destruction of the deciduous incisors, canines, and molars, with no evidence of periapical and/or furcation lesions.

After the initial clinical examination, the patient was evaluated by the hospital anesthesiologist and cleared as ASA 2 and score lee 0,9%. The electrocardiogram revealed a pathognomonic profile of a patient with Tetralogy of Fallot. Chest X-ray revealed clear costophrenic angles.

The patient was subjected to the surgical procedure under general anesthesia. Previously, antibiotic prophylaxis was administered 2 hours before the surgical procedure as recommended by the cardiologist. Intraoral and extraoral antisepsis was performed with 2% chlorhexidine. Local infiltration with 2% lidocaine and 1:200,000 epinephrine was administered in all quadrants of

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the mouth. All carious dental elements (51, 52, 53, 54, 55, 62, 64, 65, 75, 84, and 85) were extracted without complications. Resorbable sutures were placed, and hemostasis was obtained. After the procedure was completed, the anesthesia was reversed, and the patient was extubated without complications.

Recovery from anesthesia proceeded without adverse events. The patient remained in the post-anesthesia care unit until he was wide awake and diet was offered. After acceptance of diet, he was moved to the pediatric unit overnight for monitoring and was discharged home the next morning. A prescription was given for a regimen of 250mg penicillin 6/6 hours for seven days, and a follow-up appointment was scheduled in one week for reevaluation.

During the follow-up appointment, the patient was found to be in good health. Upon intraoral clinical examination, the regions where extractions were performed were healing normally. No infectious focus was observed. The mother reported being more attentive to the child's oral hygiene care, as reinforced by the team. Subsequently, the patient was discharged from the care of the oral and maxillofacial surgery team to undergo further cardiac surgical intervention under the supervision of the cardiology medical team, which has overseen his case since the initial Tetralogy of Fallot correction procedure.

DISCUSSION

This case report highlights the complexity and inherent challenges of managing pediatric dental patients with severe systemic comorbidities, such as Tetralogy of Fallot and DiGeorge Syndrome. Both conditions have significant implications for general and oral health, requiring an interdisciplinary and highly personalized approach. Patients with congenital heart diseases are at an elevated risk of developing dental caries, particularly severe early childhood caries. This heightened risk is attributed to the frequent consumption of sweetened medications, which elevates the intraoral bacterial count and consequently raises the likelihood of concurrent bacteremia during dental procedures[18]. Additionally, these patients are more susceptible to caries due to increased plaque retention on the enamel's developmental defects, which arise from systemic disturbances during amelogenesis[19,20]. Establishing regular dental visits early in childhood, followed by consistent follow-up appointments, is crucial in such cases. These proactive measures play a key role in implementing preventive strategies to address potential dental issues as the child grows[21].

The DGS has a profound impact not only on affected individuals but also on their families, society, and healthcare providers worldwide. Despite its estimated frequency, around 1:3000-1:6000 live births, the number of reported cases in literature is disproportionately low, raising concerns about newborns awaiting genetic diagnosis and children who remain undiagnosed[22]. This diagnostic delay can lead to adverse outcomes, since DSG is a major cause of morbidity and mortality, contributing to birth defects, complications in childhood and adulthood[23].

Congenital heart disease is identified in approximately 60–80% of children with DGS. The most common cardiac anomalies in these patients are conotruncal defects, which include Tetralogy of Fallot, pulmonary atresia with ventricular septal defect, truncus arteriosus, interrupted aortic arch type B, conoventricular and/or atrial septal defects, and aortic arch anomalies[24,25]. The syndrome is associated with unfavorable early perioperative outcomes, which may subsequently lead to worse cognitive and neuropsychiatric outcomes[26]. Additionally, major comorbid conditions such as hypocalcemia, thyroid disorders, autoimmune diseases, behavioral problems, and neurodevelopmental disabilities further exacerbate the adverse prognosis[27].

In the present case, the patient was diagnosed right after birth with DGS and ToF and underwent complete repair of ToF, which was essential to his good prognosis. A comprehensive assessment of the patient's medical history and symptoms is critical, necessitating effective communication with a multidisciplinary team, including cardiologist, pediatric dentist and pediatrician. This multidisciplinary approach reduces the risk of adverse effects, as it aids the dentist in developing an appropriate oral care plan while enabling the cardiologist to schedule procedures and mitigate dentally related complications[8].

Given the complexity of managing a patient with DSG and ToF, it is imperative that the anesthesiologist possesses thorough knowledge and expertise in the patient's care[8]. In the reported case, the decision to perform treatment under general anesthesia was based on the need to ensure the safety and effectiveness of the dental procedures, considering the patient's clinical and behavioral limitations. This decision underscores the importance of minimizing physical and psychological stress, as well as reducing the risk of potentially life-threatening cardiac complications. Therefore, a pre-anesthetic evaluation was conducted to assess the risks associated with administering general anesthesia for the surgical center approach. The patient was cleared by the anesthesiologist as ASA II with a Lee score of 0.9%, the latter being used to predict the cardiac risk index[8]. In the evaluation by the pediatric

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cardiologist, the ECG revealed pathognomonic signs of Tetralogy of Fallot, with an ejection fraction of 65.6%. These thorough assessments, combined with multidisciplinary collaboration, were critical to safely and effectively managing the patient's complex medical conditions.

As previously mentioned, the incidence of endocarditis is significantly higher in this patient population, making pivotal the administration of antibiotic prophylaxis prior to the procedure. However, it is crucial to prioritize the prevention of infection and its potential complications. Therefore, definitive treatments, such as extractions, may be preferable to procedures like pulpotomy, as they more effectively address the underlying risks associated with infection in these high-risk patients[8,28]. In the present case, the patient was going to be subjected to another corrective cardiac surgery and we chose to do the multiple extractions to eliminate the possibility of new infections, once the patient had a non-collaborative behavior.

Although this case reported a repaired defect of ToF with no signs of cyanosis, the patient was scheduled for an additional cardiac surgery following the completion of oral health management. This underscores the importance of a comprehensive evaluation of the patient's medical condition, both pre- and post-heart defect repair, to ensure the safest and most effective interventions. Nonetheless, for patients with unrepaired ToF, additional precautions are necessary to mitigate the risk of severe cyanotic episodes. The patient's medical condition, both before and after the heart defect repair, should be carefully evaluated to determine appropriate interventions. A multidisciplinary approach that includes close collaboration between cardiologists, anesthesiologists, and dental professionals is critical for tailoring care to the unique needs of patients with repaired or unrepaired congenital heart defects.

CONCLUSION

In summary, to provide the necessary treatment to patients with CHD it is mandatory to include consultations with physicians and cardiologists to reduce the risk of serious systemic complications while ensuring the successful completion of required dental treatment.

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CONFLICT OF INTERESTS

The authors declare no conflict of interest.

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Received 01/09/2025
Accepted 31/12/2025