

CASE REPORT

Orofacial features and dental management of a pediatric patient with moyamoya disease—A one year follow up

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Key Clinical Message

Moyamoya disease (MMD) is a chronic cerebrovascular disorder increasing stroke risk. This study presents a four-year-old MMD patient, discussing her multidisciplinary care plan including dental, behavioral, and medical management. It emphasizes the need for awareness among oral physicians and suggests further research due to the rising prevalence of MMD.

Abstract

Moyamoya disease (MMD) is a chronic cerebrovascular disorder that narrows major blood vessels in the brain, reduces blood flow, and increases the risk of stroke. The purposes of this study were to: (1) present the case of a four-year-old patient diagnosed with MMD; (2) discuss important aspects of her multidisciplinary care plan involving dental, behavioral, and medical management; and (3) suggest approaches and recommendations for health care providers. A four-year-old Middle Eastern female diagnosed with MMD, neurogenic bladder, chronic constipation, hyperactivity, and delayed milestones presented to the dental clinic with her parents seeking dental care. The child was referred by a pediatric neurologist for dental care. In the present case, there were no specific oral manifestations. However, oral physicians should be aware of possible behavioral management modalities when providing dental care to pediatric patients, including those diagnosed with MMD.

KEYWORDS

Case report, Dental care, Moyamoya disease, Pediatric dentistry

1 | INTRODUCTION

Moyamoya disease (MMD) is a progressive cerebrovascular condition of unclear etiology involving the intracranial internal carotid arteries and their proximal branches, leading to reduced blood flow in major blood vessels and predisposing affected patients to stroke.¹ In 1957, MMD was first described as “a hypoplasia of

bilateral internal carotid arteries”; this led to the appearance of diagnostic criteria for the identification of dilated collateral vessels in the angiograph.² The condition was termed *moyamoya* by a Japanese scientist. It means “something hazy like a puff of smoke,” which describes the appearance of the vessels on an angiograph.³ This disease originated in Asia, particularly in females.⁴ In Japan, a prevalence of three cases per 100,000 was

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reported.⁴ However, in Europe, a prevalence of 0.3 cases per 100,000 (1/10 of Japan) was reported,⁵ and a lower incidence among Americans (0.086 per 100,000) was reported.⁶ Recently, the incidence in East Asia increased to 6.03 per 100,000.⁷ When MMD is associated with any of the syndromic conditions, such as trisomy 21, sickle cell disease, neurofibromatosis type 1, and cranial therapeutic irradiation, it is referred to as Moyamoya syndrome.¹ To date, no previous study has focused on the worldwide prevalence or epidemiology of Moyamoya disease. Also, there is a lack of data on the disease prevalence in the Middle East region and Saudi Arabia.

The dental literature is limited regarding the management of pediatric patients with MMD. Only 15 scientific studies on MMD cases have been published in the Middle East: five in Saudi Arabia,^{8–12} five in Kuwait,^{13–17} two in Lebanon,^{18,19} one in Qatar,²⁰ one in Bahrain,²¹ and one in Egypt.²² The present study is the first to examine the dental management of MMD in the Middle East. Here, a case of a 4-year-old female with MMD is presented, and her physical and dental characteristics are described. The results of several investigations identified the patient's need for dental surgery.

Prevention of oral disease and early dental management is crucial for children with chronic medical conditions. Some conditions increase susceptibility to oral disease, while other conditions can be exacerbated by the discomfort and anxiety typically associated with dental procedures, which can further complicate their medical status. Prompt and regular dental care helps prevent serious oral diseases, minimizes the risk of infections that can affect the systemic condition, and ensures that any dental complications are managed effectively.²³ This study aimed to achieve the following: (1) present the case of a child diagnosed with MMD; (2) discuss important aspects of her multidisciplinary care plan involving dental, behavioral, and medical management; and (3) suggest approaches and recommendations for healthcare providers.

2 | CASE HISTORY

Verbal and written informed consent to the patient's inclusion in the study was obtained from her parents. They were informed about the purpose of the report, nature of the information to be disclosed, and the intended use of any accompanying images.

This patient was born in at King Abdulaziz University Hospital (KAUH), Jeddah, Saudi Arabia, by emergency cesarean section delivery at 39 weeks and 4 days with normal birth weight. Her physical examination at birth was normal, and the patient was discharged following a normal protocol.

At the age of 22 months, the patient presented to KAUH because the parents were concerned that she was not up to the milestones for her age, had clenched fists, and was unable to sit up until 11 months of age. The physical examination revealed that the patient was dysmorphic, with microphthalmia, hypertelorism, and low-set ears. Motor dysfunction and developmental delays were also observed. Mild myotonic muscular dystrophy has been suggested as a primitive diagnosis. Magnetic resonance imaging (MRI) revealed a prominent and tortuous pial vasculature in addition to a dural arteriovenous (AV) fistula. However, no intervention except supportive management was provided, and a genetic study was recommended. The family's psychosocial history and relevant genetic information yielded inconclusive findings.

Episodes of urinary retention and *E. coli* urinary tract infection (UTI) recurred several times over a period of 15 months, and multiple catheterizations and bladder ultrasounds were performed. Physicians were concerned that a neurogenic bladder was involved. Based on the neurologist's recommendations, the patient was admitted for cerebral angiography under intravenous sedation. The angiogram revealed dilated and extensive lenticulostriate and thalamostriate collaterals in the basilar cistern below the basal ganglia and thalamic regions (i.e., "puff of smoke"), with no evidence of aneurysmal dilatation or arteriovenous malformation. The neuroradiologist then raised concerns regarding MMD.

To confirm this diagnosis, the patient was admitted for brain computed tomography (CT) (Figure 1). The CT presented evidence of decreased cerebral blood flow and volume, with corresponding increases in time to peak (TTP) and mean transit time (MTT). Further clinical and electroencephalogram (EEG) correlations have been suggested.

2.1 | Investigations and treatment

At the age of four years, the patient presented to the Pediatric Dentistry Department at King Abdulaziz University Dental Hospital (KAUDH); her parents complained of multiple tooth cavitations with no pain. Upon intraoral clinical examination, carious lesions were detected in all primary molars. The patient was asymptomatic, with no clinical signs or history of abscess formation. However, the parents were directed to preventive dental treatment before the onset of dental pain. A medical consultation was obtained, and there were no contraindications for any dental intervention, including the need for general anesthesia (GA).

Because of her complex medical condition and acute stress response, the patient underwent dental treatment in an operating room (OR) setting under GA. This treatment

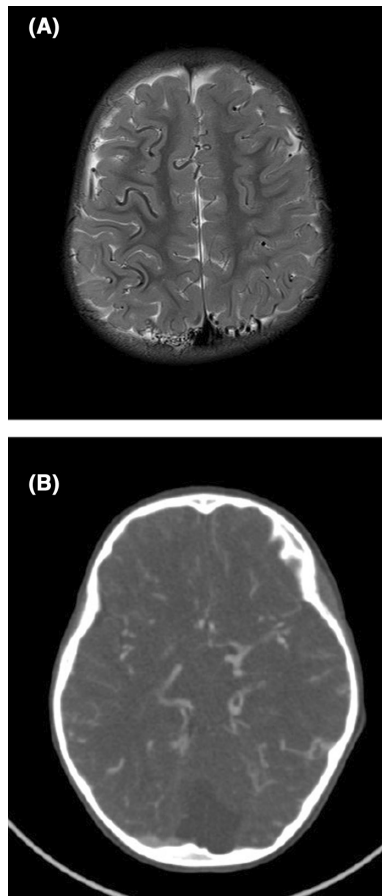


FIGURE 1 Magnetic resonance imaging (MRI) and brain computed tomography (CT) of a pediatric patient diagnosed with Moyamoya disease (A). The MRI showed that tortuosity and enlargement of the pial vessels were predominantly distributed around the posterior aspect of the superior sagittal sinus, particularly along the right cerebral hemisphere. Some tortuous vessels were also noted within the basal cisterns and posterior fossa. Prominent leptomeningeal vessels along both cerebral hemispheres, with mild prominent cortical vessels. Bilateral periventricular mild white matter volume loss with posterior predominance. Secondary elongation and distortion of the posterior horns were observed (B). CT perfusion revealed decreased cerebral blood flow.

modality was chosen because it is the safest and most controlled environment for children who demonstrate a risk of ischemic or hemorrhagic events.²⁴ Pre-operative laboratory investigations included a complete blood count (CBC), prothrombin time (PT), partial thromboplastin time (PTT), international normalized ratio (INR), urea and electrolytes (U&E), and blood typing. All parameters were within the accepted ranges to operate, the patient was brought to the OR at KAUH, and GA was initiated. Intra oral radiographs and photographs were taken (Figure 2).

The following treatment was performed under rubber dam isolation: teeth prophylaxis, mineral trioxide aggregate (MTA) pulpotomy; stainless-steel crowns on the

primary upper right first molar, primary upper left first molar, and primary lower left first molar; indirect pulp capping and stainless-steel crowns on the primary upper right second molar, primary upper left second molar, primary lower left second molar, primary lower right first molar, and primary lower right second molar (Figure 3). On the remaining teeth, five percent sodium fluoride varnish was applied.

The patient did not experience any perioperative complications, and her vital signs remained stable. The patient was discharged on the same day. Ten days postoperatively, the patient returned to the dental clinic with typical manifestations of hand-foot-and-mouth disease, including maculopapular or papulovesicular rashes on the hands and soles of the feet, and painful oral ulcerations. Hand-foot-and-mouth disease is caused by human enteroviruses and coxsackieviruses in children younger than 10 years of age.²⁵ It is usually transmitted via fecal-oral, oral-oral, and respiratory droplet contact.²⁵ The parents of the patient were instructed to follow a palliative treatment protocol, including hydration and pain relief, as needed. Oral hyaluronic acid topical gel was suggested for use on oral sores.

The patient was seen at a three-months, six-month, nine-months, and one-year follow-up appointments. She showed marked improvement in her walking and speech abilities. The parents stated that she is now enrolled in a specialized day care center for speech therapy and behavior modification. The patient's medical history was reviewed, and no changes were reported in the past six months. However, the dose of the prophylactic antibiotic was adjusted by her pediatrician to accord with her growth and weight gain. The clinical oral examination revealed no signs of infection or inflammation, the occlusion was stable, and oral hygiene was impeccable. No pain was detected upon percussion or palpation. Bitewing radiographs showed good bone healing with no signs of infection (Figure 4). Table 1 presents the timeline of medical and dental milestones for the presented case.

3 | RESULTS

Overall, the parents were very pleased with the results of the patient's dental treatment, and they were grateful for the care she had received. They appreciated the significant amount of communication between the medical and dental teams in helping them navigate the most suitable treatment plan for her case. They noted the genuine concern shown by the healthcare professionals, who not only addressed her physical needs but also alleviated the emotional burden that accompanied her medical condition. They highlighted that the collaborative effort had made them feel supported and understood, including the

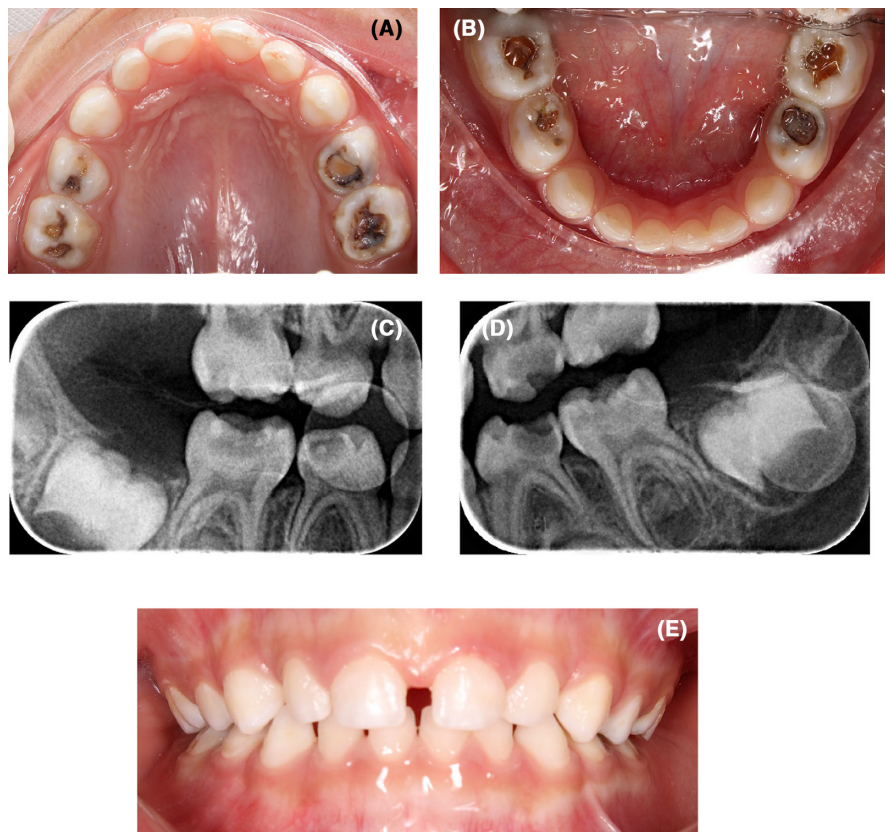


FIGURE 2 Clinical and radiographic presentation of the patient's oral cavity before full mouth rehabilitation: (A) and (B) Clinical presentation of the upper and lower views of the dental arches; (C) and (D) Radiographic bitewings presenting the upper and lower posterior teeth; (E) Frontal view of the patient's teeth.

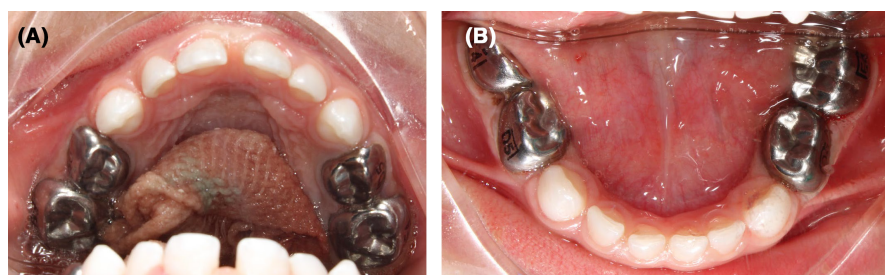


FIGURE 3 Clinical views of the patient's teeth following full mouth rehabilitation: (A) upper dental arch and (B) lower dental arch.

personalized attention during consultations and the empathy shown during challenging moments.

4 | DISCUSSION

This case report presented a case of a four-year-old Middle Eastern female patient with a progressive cerebrovascular disorder and a clinical diagnosis of Moyamoya disease based on the pathognomonic feature of a “puff of smoke” detected by a brain CT scan. The patient had a history of recurrent UTIs and a neurogenic bladder. When first presented at the dental clinics, she was still wearing diapers. She presented with hyperactivity and delayed milestones, including speech and sitting ability, which are compatible with some of the early symptoms described in the literature, including cognitive impairment and mobility disturbances.^{26,27}

According to the literature, almost all reported cases concerning the dental care of patients with MMD have employed pharmacological methods, specifically sedation and general anesthesia.^{28,29} However, in one reported case, the child displayed the potential for cooperation and was treated through the application of non-pharmacological behavior management strategies, including systematic desensitization, the “tell-show-do” technique, and live modeling. In addition, efforts were made to minimize the length of the procedure as much as possible.³⁰ In children with MMD, implementing pain management strategies, such as sedation during the perioperative period, can reduce the occurrence of postoperative ischemia and accelerate the recovery process.³¹ Anesthesia should be carefully administered by ensuring stable hemodynamics and preventing both cerebral and peripheral vasoconstriction. Hypotension may result in ischemia, whereas hypertension can lead to intracranial hemorrhage.³¹ [Table 2](#)

FIGURE 4 Bitewing radiographs showed good bone healing with no signs of infection at (A) and (B) six-month follow-up visit and (C) and (D) one year follow-up visit.

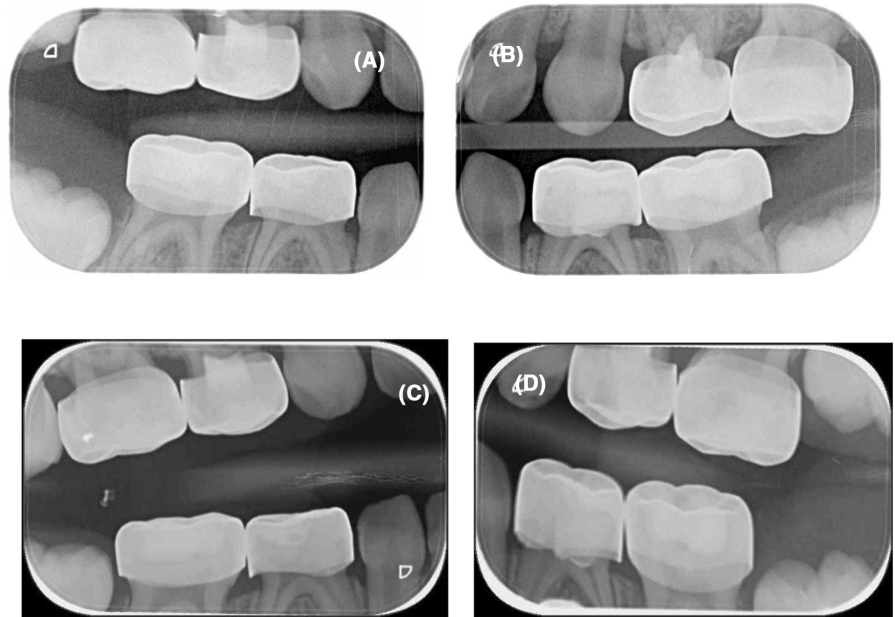


TABLE 1 Timeline of medical and dental milestones for the presented case with MMD.

	Medical status	Dental Status
2019	Patient was born, went through physical examination, and discharged normally	No dental visit yet
2021	Parents noticed delay in development which was confirmed by the physician	No dental visit yet
2022	Patient had recurrent UTI and diagnosed later with neurogenic bladder	No dental visit yet
2023	Patient was seen by a neurologist which recommended brain CT and MRI and diagnosed with MMD	First dental visit and full mouth rehabilitation under general anesthesia
2024	Follow up neurology visit	Follow up dental visit

compares oral findings and dental management in pediatric patients with MMD in the literature.

In the present case, pre-anesthesia blood parameters were assessed (CBC, PT, PTT, INR, U&E, and blood typing) which all were within normal ranges. Oral midazolam was administered to the patient prior to transferring her to the operating room. General anesthetic medications were delivered through the feet, as the veins of the hands were inaccessible. It is noteworthy that in dental treatment for patients with MMD, it is essential to avoid potential pain and stress resulting from dental caries; pain, stress, and hyperventilation could aggravate stroke.³⁵

The prognosis of children with MMD is contingent upon several factors, such as the speed and extent of vascular blockage, the patient's capacity to establish functional collateral circulation, the age at which symptoms first appear, the severity of initial neurological impairments and resulting disability, and the level of infarction observed on CT or MRI scans during the initial presentation.³⁶ Typically, a neurological condition at the time of treatment, rather than the patient's age, serves as a more

predictive indicator of long-term outcomes.²⁴ However, children with MMD usually have a higher rate of completed stroke because, unlike adults, they may not be able to communicate the early signs of stroke.³⁷

Currently, no medical intervention has been found to halt or reverse the advancement of Moyamoya syndrome. However, evidence supports the utilization of two categories of medications to decelerate disease progression: anticoagulants/antiplatelet agents and vasodilators.^{24,36} In severe cases, conservative medical management is ineffective in managing the condition, such as the implementation of surgical revascularization interventions to prevent disease progression.²⁴ Because our patient was asymptomatic, she had not undergone medical or surgical treatment for MMD.

In the present case, a team of pediatricians, Pediatric dentists, urologists, nephrologists, gastroenterologists, neuroradiologists, speech therapists, and neurologists collaborated to manage the patient. Effective communication and prompt treatment of a patient's underlying health condition are crucial, with a simultaneous emphasis on

TABLE 2 Oral findings and dental management reported in the literature of pediatric dental patients with MMD.

Author, year	Country	Age (in years), gender	Oral findings	Behavior management technique used
Eslava, 2021 ³²	Syria	10, male	<ul style="list-style-type: none"> Dental malposition and cavities 	<ul style="list-style-type: none"> Not mentioned
Rahul, 2021 ³⁰	India	5, female	<ul style="list-style-type: none"> Early childhood caries Retained maxillary primary incisors with palatally erupting maxillary permanent central incisors Increased overjet with class II molar relationship 	<ul style="list-style-type: none"> Systematic desensitization Tell–show–do technique Live modeling
Goswami, 2020 ³³	India	12, male	<ul style="list-style-type: none"> Broken upper anterior teeth due to dental trauma 	<ul style="list-style-type: none"> Basic behavior management techniques in the dental clinic
Van Camp, 2019 ²⁸	Belgium	15, female	<ul style="list-style-type: none"> Agenesis of the left incisor Fully impacted wisdom teeth 	<ul style="list-style-type: none"> Dental treatment under conscious intravenous sedation with midazolam and fentanyl
Alkeshan, 2019 ³⁴	South Korea	4, male	<ul style="list-style-type: none"> Early childhood caries 	<ul style="list-style-type: none"> Full mouth rehabilitation under general anesthesia
Ko, 2018 ²⁹	USA	2, female	<ul style="list-style-type: none"> Early childhood caries 	<ul style="list-style-type: none"> Full mouth rehabilitation under general anesthesia
Bo, 2017 ²⁴	USA	8, male	<ul style="list-style-type: none"> Multiple carious primary and permanent teeth Chronic draining dental infection Anterior crossbite Crowding in both arches 	<ul style="list-style-type: none"> Dental treatment initially in outpatient setting under local anesthesia, followed by completion of treatment under general anesthesia due to moderate level of anxiety displayed by the patient

preventing dental disease.²⁹ In delivering dental care to children with MMD, effective pain and anxiety management is crucial to avoid the risk of stroke.²⁹

This study demonstrates the importance of effective communication among medical healthcare professionals to ensure holistic patient care, including dental treatment. Thorough medical knowledge is recommended for pediatric dentists who manage patients with special healthcare needs, which also facilitates communication with the hospital's medical staff.

The strength of this case report is that it contributes to the existing medical and dental literature by shedding light on a rare disease that has been inadequately documented. Based on a meticulous examination of the patient's history, diagnostic procedures, and treatment outcomes, this report provides a comprehensive overview that enhances the understanding of the orofacial features and dental management of pediatric patients with MMD. This report not only contributes valuable insights for clinicians and researchers but also serves as a reference point for healthcare practitioners who encounter similar cases in the future.

It is essential to acknowledge certain limitations that may affect the generalizability of the findings. First, the limited number of published cases and variable disease presentations restricted the ability to formulate a protocol for the dental management of patients with MMD.

Second, the retrospective nature of this report implies a reliance on historical medical records, potentially leading to incomplete or missing information.

Future studies should focus on exploring the genetic and environmental elements that influence the development of Moyamoya angiopathy, which could potentially facilitate the creation of novel treatment approaches for MMD.³⁸

Children with Moyamoya disease have no specific orofacial features, yet they require a unique management protocol applied by a multidisciplinary team of healthcare professionals. Effective communication between the patient's pediatrician, neurologist, anesthesiologist, and pediatric dentist is essential for the delivery of safe and efficient comprehensive care. Emphasis must be placed on the prevention of dental disease and the reduction of pain and anxiety. The treatment approaches recommended for healthcare providers who encounter patients with conditions such as MMD are based on the findings of this case study.

Long-term dental care strategies for patients with MMD should focus on comprehensive and individualized care plans that address their unique needs. Owing to the increased risk of stroke and other neurological complications, minimizing dental infections and inflammation is essential. Regular dental follow-up visits should emphasize meticulous oral hygiene practices, such as brushing with fluoridated toothpaste, flossing, and the use of antiseptic mouthwashes

if needed. Behavioral management techniques to reduce pain and anxiety should be utilized to ensure that dental visits do not exacerbate their condition. Alongside with anticipatory guidance and support for both patients and caregivers to help in maintaining optimal oral health.

AUTHOR CONTRIBUTIONS

Salma G. Alharbi: Data curation; investigation; methodology; writing – original draft; writing – review and editing. **Ghalia Y. Bhadila:** Conceptualization; investigation; methodology; supervision; writing – original draft; writing – review and editing. **Abdullah Almushayt:** Conceptualization; supervision; writing – review and editing.

FUNDING INFORMATION

The study was independently conducted without external financial support from any specific funding agency or organization.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data supporting the reported results can be found in the hospital records.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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How to cite this article: Alharbi SG, Bhadila GY, Almushayt A. Orofacial features and dental management of a pediatric patient with moyamoya disease—A one year follow up. *Clin Case Rep.* 2024;12:e9502. doi:[10.1002/ccr3.9502](https://doi.org/10.1002/ccr3.9502)