CASE REPORT

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Multiple pulp necroses in a patient with heterozygous sickle cell disease: A case report

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This study reports several pulp necroses in a patient with sickle cell disease (SCD), a pathology with a high risk of infection which can lead to life-threatening patients. An African male patient with SCD, 31 years old, was received at the municipal oral health center on June 4, 2002, for a pericoronitis of 48 which quickly evolved into cellulitis. The treatment consisted of removing 48. He was seen in 2006 for pain while chewing. Intraoral examination showed dyschromia of 44, 45, 46, and 47. Teeth 46 and 47 presented acute apical periodontitis which were endodontically treated. In 2019, tooth 47 which is painful is removed. X-ray panoramic showed apical translucent images on several teeth. On December 13, 2021, he was received with a depressible palatal swelling due to 12. Endodontic treatment is performed under curative antibiotic therapy. Conclusion: Regular oral monitoring of patients with SCD must be implemented to prevent severe complications with life-threatening.

Keywords: Endodontic treatment; multiple pulp necroses; sickle cell disease.

Introduction

Hemoglobinopathies or sickle cell diseases (SCD) are genetic diseases characterized by an inherited anomaly of hemoglobin. They are divided into two groups: The hemoglobinosis group characterized by structural abnormalities of the globin chain and the thalassemia group characterized by a deficit of one or more hemoglobin chains (1). This deficiency leads to the formation of abnormal hemoglobin S. Transmission is autosomal recessive. Major sickle cell syndromes are represented by SS homozygotes and double heterozygotes SC, SDPunjab, Sß thalassemia, SO-Arab, and SE (2). Nearly 7% of the world population carries a defective globin gene. SCD is the most widespread genetic disease in the world and poses a public health problem with 300–500 thousand homozygous SS newborns (3). In Burkina Faso, the prevalence of the anomaly in the general population is between 5 and 10% (2). Clinical manifestations are related to chronic hemolytic anemia, vaso-occlusive phenomena, and extreme susceptibility to infection (4). Apart from the general manifestations,

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periodontal repercussions linked to circulatory and carious problems have been mentioned. Severe periodontolysis as well as tumors and osteomyelitis in the jaws have also been demonstrated (5). Pulp necrosis has also been associated with SCD. However, very few clinical works on the oral aspects of this disease exist in the literature in Burkina Faso (6). This article reports a case of several pulpal necroses discovered in a patient with SCD following consultation and management of right mandibular cellulitis. Indeed, the high risk of infection in this disease can lead to life-threatening of these patients. The objective of this report is to disseminate widely the impact of this pathology on oral health and to remind the interest for dental surgeons of a careful clinical examination that looks for a history of SCD in the context of low-income countries.

Case Report

This clinical observation reports the case of an African male patient, 31 years old, received at the municipal oral health center (MOHC) on June 4, 2002, for dental pain on the right hemi-mandible in the posterior sector. The clinical examination reveals a good general condition. The medical history consists only of heterozygous SCD. No drug treatment was undertaken by the patient and he had no medical follow-up concerning his SCD. He had not previously consulted or received dental care. Exobuccal examination reports unilateral right submandibular adenopathy. No carious lesion was noted on intraoral examination. The dentition was complete and permanent. However, plaque control was poor with significant tartaric deposits on all the teeth. The 48, chief complaint was not an impacted tooth but presented a pericoronitis. The retroalveolar X-ray that was performed did not reveal any particular information. In view of the SCD mentioned by the patient, antibiotic therapy based on amoxicillin at the rate of 1 g every 12 h was started. A level 1 analgesic, in this case, paracetamol at the rate of 1 g every 6 h was prescribed as well as mouthwash with chlorhexidine. On June 7, 2002, the mucous cap was removed and the patient was seen again for a check-up on June 14, 2002. This check-up showed a significant reduction in symptoms. Two weeks later (June 20, 2002), he presented for consultation with a right genius cellulitis and trismus limiting the mouth opening to 25 mm in amplitude. During the interrogation, he reported having noticed the genius swelling when waking up in the morning. A curative antibiotic therapy based on amoxicillin (2 g/day) and metronidazole (1.5 g/day) was started and tooth 48 was removed the following day. On July 11, 2002, the patient benefited from ultrasonic scaling and polishing. He then lost sight of it until December 20, 2006, when he returned to consultation for severe pain when chewing in the same area. Extraoral examination revealed bilateral subangulo-maxillary adenopathy. Endobuccal examination revealed a complete permanent denture and dyschromia of teeth 47, 46, 45, and 44. Vitality tests were negative and axial percussion was painful on teeth 47 and 46, which presented an acute apical periodontitis. Antibiotic prophylaxis based on amoxicillin (2 g in a single dose 1 h before the procedure) and an analgesic treatment with paracetamol is started. Emergency treatment consisted of carrying out nerve bloc anesthesia supplemented with the vestibular and followed by an access cavity (on 47 and 46) after placement of the rubber dam. This allowed canal debridement with 2.5% sodium hypochlorite abundant rinsing. Temporary medication with calcium hydroxide followed by a tight coronary filling with zinc oxide-eugenol is performed. Both teeth are put under bite and routine endodontic treatment is scheduled later. On January 10, 2007, root canal fillings for 47 and 46 were performed, and a coronary filling with zinc oxide-eugenol was sealed. Once clinical silence was obtained, the definitive restorations were performed 3 weeks later. It was then carried out preventively the endodontic treatment of 45 and 44 which were necrosed. However, once the treatment has started, the patient loses sight of it before the end of the procedure. At this time, the orthopantomogram was not available in the country.

Twelve years later, on June 8, 2019, the patient was seen again for pain in the same area. Tooth 47 which had lost its coronary filling was mobile and painful. Tooth 45 was fractured. Since the orthopantomogram is now available in the country, a panoramic X-ray (Carestream Health Inc. 150 Verona Street Rochester, NY 14608) of the jaws is requested (Fig. 1). Tooth 47 was removed on amoxicillin antibiotic prophylaxis and the patient scheduled to complete the others treatments. In addition, the panoramic X-ray made it possible to note the presence of apical images on 14, 12, 21, 22, 33, 41, and 43. Once again the patient has lost sight of it.

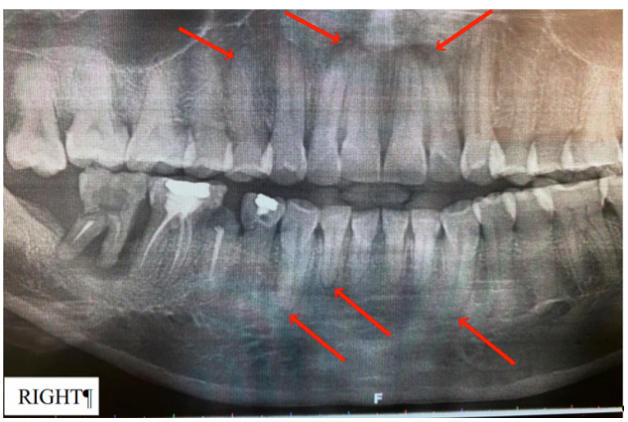

Fig.1. Dental panoramic X-ray from June 08, 2019, showing apical images (multiple necroses) (Dr Kaboré, 2019).

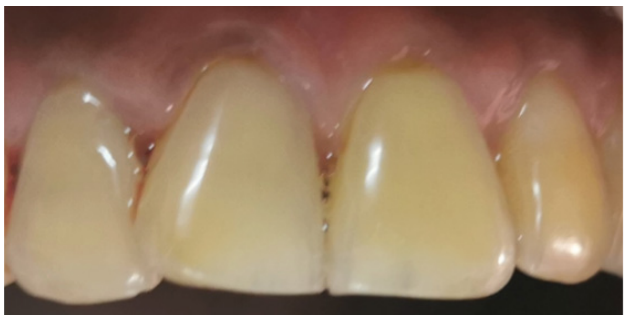

Fig.2. Teeth 12, 11, 21, and 22 discolored (Dr. Kaboré, 2021).

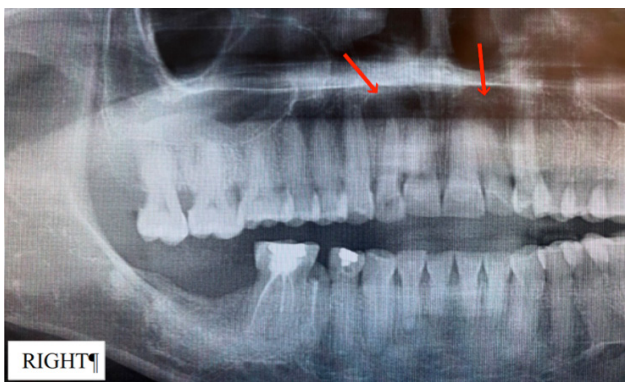

Fig.3. Dental panoramic X-ray from December 14, 2021, showing an increase in the size of the images on the apices of 12 and 22.

Two years later, on December 13, 2021, the patient, now 51 years old, was received urgently for dental pain that did not yield to conventional analgesics. He was directed to the conservative dentistry and endodontics unit of the MOHC. The medical history consists only of heterozygous SCD. The clinical examination reveals an apparently good general condition. During the interrogation, the patient complained of a "masse" in the palatal region, painful, which appeared a week earlier, and causing functional discomfort. No notion of trauma was found. The endobuccal examination reveals a depressible palatal swelling. Teeth 21, 22, 11, and 12 are discolored (Fig. 2) and vitality test is negative. Tooth 12 is painful on percussion. Panoramic radiographic (Carestream Health Inc. 150 Verona Street Rochester, NY 14608) reveals a well-limited maxillary radiolucent image on 12 (Fig. 3). The endodontic treatment of 12 is carried out under antibiotic therapy (amoxicillin 2 g/day during 7 days). Tooth 13, whose root adjoins the cavity, responds positively to the cold vitality test. It will thus be subject to regular monitoring. The patient has again lost sight of it. He presents himself on January 18, 2023, for a systematic visit. He does not complain of any pain or discomfort and we took the opportunity to perform a descaling. The control radiograph (Carestream Health Inc. 150 Verona Street Rochester, NY 14608) shows the beginning of apical healing on 12 (Fig. 4).

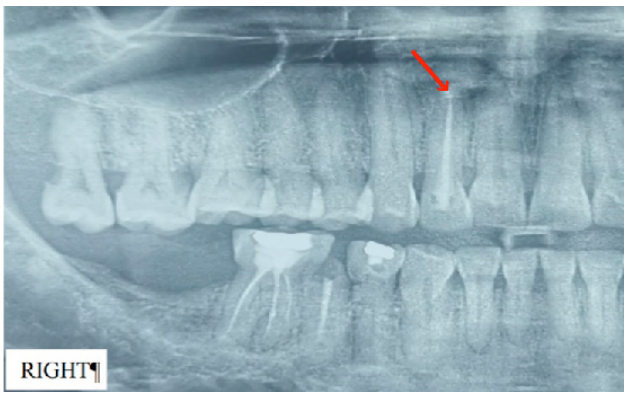

Fig.4. Dental panoramic X-ray of the review showing the treatment of 12 (January 18, 2023) (Dr. Kaboré, 2023).

Discussion

This is only one case of multiple pulp necrosis reported in a patient with SCD. Notwithstanding this, this observation has made it possible to highlight the attention that the dental surgeon must pay to patients with hemoglobinopathies. Indeed, oral health facilities in low-income countries, when they are functional have an increasingly large influx of patients (7). The eagerness of the practitioner could impact the quality of the clinical examination and important medical history in the diagnostic and therapeutic behavior may escape him. SCD is considered one of the most common genetic diseases whose manifestations will affect the patient's quality of life (8). It is characterized by the following main systemic manifestations: Complex pain syndrome, chronic hemolytic anemia, immune deficiency, multisystem damage to major organs, but also various damage to the oral cavity (9,10). The pallor of the mucosa, enamel hypomineralization, periodontolysis, pulpal calcifications, and aseptic pulpal necrosis can be observed (10,11). Chekroun et al. (10) report that the presence of necrotic teeth is 8.33 times higher in a patient with SCD compared to a patient without SCD, due to vascular occlusions of the pulp microcirculation . These vascular occlusions promote the formation of pulpolytes in the pulp tissue. There are many cases of pulp necrosis without any past pain. Pulp necrosis refers to the death of the pulp accompanied by its destruction. Clinically, dyschromia of the crown is observed (12). Thermal tests do not cause a reaction, demonstrating the insensitivity of the tooth (13). The mechanical opening of the pulp chamber is accompanied by a putrid odor when the necrosis is septic. An association between SCD and pulp necrosis in clinically healthy teeth is also demonstrated in the work of Alves et al. (14). A study in Côte d'Ivoire shows a prevalence of 10% of cases of pulp necrosis among 60 SCD patients (15). Rivera-Salinas et al. (16) reported the

same proportions . Andrianjafinoro et al. (17) in a study in Madagascar found a prevalence of 21.7% of sickle cell patients who presented with pulpal necrobioses on intact teeth . Red blood cells containing sickle hemoglobin have the property of polymerizing when they are deoxygenated, giving rise to the formation of fibers that deform the blood cells and give it a sickle appearance (18). As a result, these deformed red blood cells can constitute an embolism and interrupt the blood flow of the capillaries, thus resulting in tissue anoxia, pain, infarction, and necrobiosis. The terminal-type pulpal vascularization penetrates the dental root through the apical foramen which is a constriction of a closed cavity with inextensible dentinal walls. This particular situation promotes pulp necrosis at the slightest vaso-obliteration. SCD increases the risk of infection. Thus, preventive dental care and a healthy lifestyle are important. A biannual consultation with a dental surgeon is essential to detect any latent or at-risk infectious focus. In this report, the patient was lost sight of several times. It is important to raise awareness in concert with hematologists to establish de facto dental clinical follow-up in these patients.

In endodontics, the risk of an expanded infection is the main concern associated with severe SCD (19). Antibiotic therapy should be systematic in the event of suspected infection (20). According to Costa et al. (21), pulp necrosis of permanent teeth with intact crowns in patients with SCD can be considered an indicator of lethality . Various studies report a high prevalence of caries in these patients and recommend awareness campaigns to limit the comorbidity factors (insufficient oral hygiene and snacking on sugary foods) to their state of health already weakened by SCD (22-24).

Conclusion

SCD is a genetic disease that requires particularly complex treatment. While the general lesions are well-known and described, the details of the oral repercussions of the disease are rather unknown. Naturally, disease management focuses on the vital needs of patients, so oral care is often overlooked. However, given the place that oral health plays in the quality of life of these patients, an important place should be given to dental prophylaxis. In addition, the high risk of infection in sickle cell patients requires special precautions such as rigorous anti-infectious prophylaxis and controlled prescription of analgesics. Due to the vaso-occlusive phenomena, the sickle cell sufferer has an extreme susceptibility to infection, which may justify these cases of multiple necroses. Regular oral follow-up of the sickle cell patient should be systematic.

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Informed consent: Written informed consent was obtained from patients who participated in this study.

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