



Management of Odontogenic Abscess in Patients with Sickle Cell Anemia: 5 Case Reports

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Authors' contributions

This work was carried out in collaboration between all authors. Author MMA designed the study, Author NBF wrote the protocol and the manuscript. Authors KMD and NKO managed the literature searches. Authors MMA and SR helped with the final editing of the manuscript. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Sickle cell anemia (SCA) is a hereditary disorder characterized by a defect in the red blood cell (RBC) hemoglobin molecule resulting in a diminished ability for the blood to carry oxygen and a tendency for the blood to clump. Clinical manifestations of SCA, apart from the vasoocclusive complications are increasing susceptibility to infection, and chronic anemia from both hemolysis of

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the sickle cells. Five patients with age ranging from 14 to 27 years; presented to the service of Dentistry, Mixed Medicine Center and SS Anemia (MMCSA), Kinshasa/DR. Congo with a history of pain and swelling in the mandible and/or maxillary regions of several days duration. All 5 patients were diagnosed with odontogenic abscesses. Because any infection in a patient with SS disease can become life-threatening, we present these case reports to call attention to this serious problem and share our clinical management and experience, especially with surgeons who practice in areas with limited resources. In addition, we review the signs, symptoms, diagnosis, and surgical management of odontogenic infection.

Conclusion: The diagnosis and the management of odontogenic abscess in the patients with sickle cell anemia require consultation the patient's physician and understanding of the physiopathology as well as the oral clinical manifestations of the disease.

Keywords: Odontogenic abscess; diagnosis; management; sickle cell anemia.

1. INTRODUCTION

Sickle cell anemia is a genetic disease caused by replacement of glutamic acid by valine in position 6 at the N-terminus of the beta-chain of globin, thus resulting in hemoglobin S [1]. The disease is characterized by hemolytic anemia and episodes of severe pain termed sickle cell crisis [2]. Sickle cell anemia occurs primarily in the black population with an incidence of approximately 1/400 in the homozygous state and around 1/10 in the heterozygous state. Persons of Mediterranean, Middle Eastern, and East Indian descent may also be at risk [2,3]. Sickle cell anemia is the most common single gene disorder and haemoglobinopathy found among many populations in the world [4]. In the heterozygous state, the quantity of normal hemoglobin A accounts for 55%–58% of the total hemoglobin and the patient is usually asymptomatic. A sickle cell crisis may take place only in cases with severe stress, severe hypoxia, and severe infection [2]. However, in the homozygous state, the abnormal hemoglobin S levels reach 70%–98%, and the red blood cells may become sickled even under normal oxygen tension. They can be accompanied by joint, muscle, abdominal, chest, and bone pain and possibly low-grade fever, and fatigue with weakness [5].

The most common oral manifestations of sickle cell disease are mucosal pallor, yellow tissue coloration, disorders of enamel and dentin mineralization, changes to the superficial cells of the tongue, multiple caries and periodontal disease that can lead to cellulitis and/or odontogenic abscess [6]. The infection often occurs in poor and marginalized person, with inadequate oral hygiene. It can be accompanied by a fever, a physical asthenia, problems with eating and serious breathing difficulties. Understanding and appreciating this disease of great importance for surgeon, physician and

dentist because of the fact that not only is it commonly seen in clinical practice, but also is challenging and can lead to fatal complication if not treated quickly and appropriately. Our purpose here is to present this case series in order to share our experience with the surgeons practicing in resource-poor settings, especially in relation to clinical assessment and subsequent management.

2. CASE REPORT

Five patients with age ranging from 14 to 27 years and had a mean age of $21.2 \pm$ (SD: 5.16), gender such as demonstrated in (Table 1), reported to the service of Dentistry, Mixed Medicine Center and SS Anemia (MMCSA), Kinshasa/DR. Congo; complaining of swelling in the mandible and/or maxillary for a period of several days and accompanied by joint, muscle, abdomen, chest, and bone pain (Tables 2 and 3). Their medical history, obtained from guardians revealed that the patients were known to have sickle cell disease and all received their care at the same Medical Center. The patients had been hospitalized more than three times and received more than five blood transfusions since being the diagnosis with sickle cell disease. Out of five patients, four of them (cases 1, 2, 3 and 4) were admitted with a provisional diagnosis of sepsis after consultation with a general physician. One patient was admitted directly and hospitalized after consultation by a dentist with a diagnostic of odontogenic abscess. Hemoglobin electrophoresis revealed genotype HbSS and an analysis of the concentration of Hemoglobin determined (Table 1).

On physical examination, the patients were alert, well-oriented and cooperative with normal gait. In the first four patients, the clinical swellings were painful, tender, and fluctuant on palpation indicative of abscess formation. The swelling

noted in patient five was firm, non-fluctuant, and exquisitely tender indicative of cellulitis. The degree of trismus in all of the cases did not permit us to perform a proper intraoral examination (Table 3). In addition to regional maxillofacial pain, bone and joint pain were the most commonly reported severe regional complaints (Table 2). Because of economic reasons, no radiological examination was able to be performed. Four of the cases were treated by surgical incision and drainage followed by extraction of all involved teeth. Resolution of the infection was so slow and the patients were followed for two months. The case of the poorly localized cellulitis was treated medically with empirical antibiotic therapy and extraction of a mandibular second molar. Managing these cases led our team of dentists to promote the prevention of oral infections by teaching good oral hygiene practice (Fig. 1). We stressed the importance of diet control, brushing teeth at least two times a day, and a routine dental examination twice each year.

patient with a sickle hemoglobinopathy must include a thorough history and physical examination. In most instances, these patients are aware of their diagnosis and have experienced multiple hospitalizations, painful crises, severe anemia, multiple blood transfusions and recurrent episode of bacterial pulmonary infections [7]. The report of general manifestations of SCA in the literature [8], was similarly as the present cases reports. A crisis may be precipitated by infection or association with identified etiology and often a history of odontogenic abscess has been found.



Fig. 1. Preventive education oral hygiene

3. DISCUSSION

Sickle cell anemia can manifest at any age but most often appears during the second and third decades of life. The initial evaluation of the

Table 1. Genotype, age, gender, concentration of hemoglobin, hematocrit and blood transfusion history

| Cases | Age (year) | Sex (M or F) | Genotype of Hb | Hb (g/dl) | Ht | Blood transfusion |
|-------|------------|--------------|----------------|-----------|----|-------------------|
| 1 | 14 | M | SS | 6 | 17 | Multiple |
| 2 | 18 | F | SS | 6.9 | 20 | Multiple |
| 3 | 23 | F | SS | 6.3 | 19 | Multiple |
| 4 | 24 | M | SS | 7 | 21 | Multiple |
| 5 | 27 | F | SS | 8 | 23 | Multiple |

Table 2. General clinical complaint

| Cases | Bone pain | Joint pain | Chest pain | Muscle pain | Abdominal pain | Fever |
|-------|-----------|------------|------------|-------------|----------------|-------|
| 1 | +++ | +++ | + | ++ | + | + |
| 2 | ++ | +++ | + | ++ | + | - |
| 3 | +++ | ++ | +++ | + | - | - |
| 4 | +++ | +++ | - | +++ | ++ | +++ |
| 5 | +++ | +++ | ++ | - | +++ | +++ |

Legend: (+++) = severe; (++) = moderate; (+) = middle and (-) nothing

Table 3. Oral and maxillofacial complaints

| Cases | Maxillary swelling | | Mandible swelling | | Teeth pain | | Trismus | Dyspnea | Treatment |
|-------|--------------------|------|-------------------|------|------------|----------|---------|---------|-----------|
| | Right | Left | Right | Left | Maxilla | Mandible | | | |
| 1 | - | + | - | ++ | ++ | +++ | 1.6 mm | - | Sur+Med |
| 2 | ++ | - | ++ | - | + | +++ | 2 mm | + | Sur+Med |
| 3 | - | - | +++ | - | - | +++ | 2 mm | - | Sur+Med |
| 4 | - | - | - | ++ | ++ | ++ | 1.2 mm | - | Sur+Med |
| 5 | +++ | - | +++ | - | +++ | +++ | 2 mm | - | Medical |

Legend: Sur+Med= Surgical and medical treatment

Trismus, dyspnea, and/or odynophagia are common symptoms associated with maxillofacial deep fascial space infections and potentially even more serious in patients with SCA. All of our cases were associated with trismus, but only one dyspnea was noted. Evidence of Hypoxia is a sign of possible imminent airway obstruction and is a medical emergency [4]. Constitutional symptoms such as fever, chills or sweating are associated with infectious diseases but does not necessarily provide any information on its nature and severity. Normally, orthopantomogram (OPG) radiographs are made to better evaluate odontogenic infections. Unfortunately, for some countries in Africa, including the RD. Congo, patients live at a lower socio-economic level making it difficult for them to access and afford basic radiographic imaging. Access to better resources would allow an earlier diagnosis and treatment of facial abscesses [9]. Worsening chest pain evolving in the context of odontogenic abscess is strongly suggestive of mediastina infection [10]. From these case reports, it was difficult to assess the possibility of mediastina infection without a CT scan.

Patients with SCA should receive preventive dental care including oral hygiene instructions, information on a proper diet to help control tooth decay, proper tooth brushing and flossing technique, as well as fluoride gel applications [11]. Surgical treatment should be prompt and aggressive. Our treatment protocol includes a removal of involved teeth and incision and drainage of abscesses under local anesthesia using Lidocaine 2% 1:100,000 epinephrine. The treatment sequence schematically:

- (1) Injection of local anesthesia;
- (2) To drain an intra-oral abscess make a stab incision over the point of greatest fluctuation; however, extra-oral incisions should be made in a natural skin crease or in an area just below the swelling in order to minimize scarring;
- (3) Pus drains;
- (4) Explore the cavity with a small curved hemostat to locate and break any loculus formations;
- (5) Placement of drain;
- (6) Apply dressing (only for extra-oral incisions);
- (7) Drain should left in place for at least each 24 hours;
- (8) Removal of drains when the drainage is completed.

Depending on the case, a rubber drain may be left in place from 6-10 days, [12,13]. Normally, antibiotics are prescribed based on the known flora that most commonly cause oral infections or on the laboratory results of culture and sensitivity tests. Because penicillin is the empirical drug of choice, we choose to administer this drug first unless the patient has a history of penicillin or cephalosporin allergies. However, studies have shown that β -lactamase producing organisms such as Bacteroids are insensitive to penicillin and 30% of the microorganisms are reported as resistant. As odontogenic or orofacial infections are caused by a mixed flora of aerobes and anaerobes, bacterial synergism enhances growth of different types of organisms. Consequently, amoxicillin in combination with clavulanate acid is the better empirical choice of antibiotics, administered by the IV route (2grx3/Day). The choice of these drugs is supported by the fact that they are inexpensive, well tolerated, and widely available. This combination was also administered in others reports by IV route, but with 1gr x3/Day [13-14]. Thus, the dose and duration of treatment depends on the general condition of the patient and also by the resolution of the infection, with on average approximately seven to twelve days.

4. CONCLUSION

Maxillofacial odontogenic abscesses are of concern for surgeons, physicians and dentists because they are commonly seen in clinical practice, challenging to manage, and can lead to fatal complications if not treated timely and in a timely manner. The diagnosis and the management of odontogenic abscess patient with the sickle cell anemia, requires medical consultation and cooperation between dentist and physician. All dental appointments should be short, stress free and long and complicated procedures avoided. Despite the poor economic conditions, there is a place for regular dental SCA patients of Kinshasa city. Through regular follow-up with the dentist and their physician, we can all assure that our patients will be able to smile at future visits.

CONSENT

All authors declare that 'written informed consent was obtained from the patient for publication of this case report and accompanying images.

ETHICAL APPROVAL

All authors hereby declare that all cases have been examined and approved by the appropriate ethics committee and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki

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COMPETING INTERESTS

Authors have declared that no competing interests exist.

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