Case Series of Patients with Haemophilia Managed in a Paediatric Dental Clinic at Lagos University Teaching Hospital

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ABSTRACT

Background: Bleeding disorders are a group of disorders in which blood does not clot properly due to defects in the blood vessels, coagulation system, or platelets. Haemophilia is one of the most frequently occurring inherited bleeding disorders. Initial recognition of such bleeding disorders and their possible effects plays a significant role in reducing potential complications. This case series aims to provide knowledge on the dental management of children with haemophilia.

Case Presentation: We report two patients with haemophilia who were managed in the Dental clinic by a team comprising fourth-year residents in Paediatric Dentistry and a consultant in Paediatric Dentistry. One of the cases was a known haemophiliac.

The first patient was a 10-year-old male who presented on account of gingival bleeding of one week's duration. The bleeding was from a pedunculated, well-circumscribed mass in the region of the lower left first molar. An impression of pyogenic granuloma was made. Excisional biopsy of the lesion was done under local anaesthesia, and the patient was discharged home afterwards with no postoperative complications.

The second patient was a 6-year-old male who presented to the paediatric dental clinic on account of bleeding from the tongue following trauma. There was a positive history of excessive post circumcision bleeding. The patient was referred to the paediatric haematology clinic for review after requesting for haematological investigations. Investigations revealed a haematocrit of 21%. Transfusion with whole blood was given, laceration was re-sutured, and patient was placed on intravenous tranexamic acid for five days. He was later discharged in a stable condition to be followed up in the clinic.

Conclusion: The management of patients with bleeding disorders depends on the severity of the condition and the invasiveness of the planned dental procedure. The goal is to minimize the challenge to the patient by restoring the haemostatic system to acceptable levels and maintaining haemostasis by local and adjunctive methods.

Keywords: Haemophilia, oral bleeding, haemostatic control, dental management

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INTRODUCTION

Haemophilia is an X-linked hereditary bleeding disorder characterized by a defect or deficiencies of coagulation factors VIII (Haemophilia A), IX (Haemophilia B, also known as Christmas disease) or XI(Haemophilia C).[1] These factors play a crucial role in the intrinsic phase of blood coagulation, with severe deficiency defined as plasma activity of <1 IU/dL (normal range: 50-100 IU/dL), moderate deficiency ranging from 2-5 IU/dL, and mild deficiency between 6-40 IU/dL.[2]

Globally, over 350,000 individuals are affected by haemophilia, with 70% having a positive family history.[3] However, in approximately 30% of cases, the disorder arises from spontaneous mutations or unrecognized maternal carrier status.[4]

Oral bleeding is a common complication in individuals with haemophilia, with reported rates averaging 29.1 episodes per year requiring factor replacement in patients with Factor VIII deficiency, of which 9% involve oral structures[5]. The most frequently affected sites include the labial frenum (60%), tongue (23%), buccal mucosa (17%), gingiva and palate (1%). Haemophilia-associated oral haemorrhage often results from traumatic injury, with severity correlated to the degree of factor deficiency.[5]

Kaneda et al,[6] further reported the distribution of oral haemorrhage as follows: gingiva (64%), dental pulp (13%), tongue (7.5%), lip (7%), palate (2%), and buccal mucosa (1%). While minor oral bleeds can often be managed with local haemostatic measures, major bleeding necessitates parenteral factor replacement therapy.

In paediatric dentistry, bleeding episodes may be triggered by physiological exfoliation of primary teeth, poor oral hygiene, and iatrogenic factors such as inferior alveolar nerve block administration. Additionally, invasive procedures, including tooth extractions, pose significant bleeding risks[5]. Effective management of haemophilics in dental practice requires a comprehensive understanding of the disorder, genetic counseling and a multidisciplinary treatment approach tailored to the severity of the condition and the planned dental intervention. The primary goal of dental care is to restore haemostatic balance and minimize the risk of bleeding complications through a combination of systemic and adjunctive local haemostatic strategies.[7]

This report presents two clinical cases of individuals with haemophilia who sought dental treatment. The challenges encountered and the treatment protocols implemented are discussed to provide insight into best practices for managing haemophilics in a dental setting.

METHODS

This case series includes non-consecutive cases in a single centre. The location of the health facility was the Lagos University Teaching Hospital, Idi-Araba, Nigeria, which is an Academic setting. Ethical approval was obtained from the institution's Health Research Ethics Committee with registration number ADM/DSCST/HREC/APP/7450. Informed consent and assent were obtained accordingly.

CASE REPORT 1

A 10-year-old male child from Nigeria presented to the Paediatric Dental Clinic at Lagos University Teaching Hospital with a one-week history of gum bleeding. The patient's mother reported the complaint. The mother had been placing cotton wool soaked in adrenaline packs to relieve the bleeding the entire week before presentation, but the bleeding persisted. The child was then taken to the Paediatric emergency. At presentation, haematocrit was 18% and the patient was admitted and worked up for blood transfusion. The patient is a known haemophilic and had been previously hospitalized nine months before the current presentation due to an injury unrelated to the head and neck region. Diagnosis was made at the haematology clinic based on findings from factor assay and clotting profile. Available tests in the facility include Factor VIII and IX assays and clotting profile. Factor concentrates are supplied by an international non-governmental organization. However, access is restricted to registered patients. History taken from the mother revealed that pregnancy was planned, with the mother seeking antenatal care from the fourth month of gestation, and there were no significant complications during the pregnancy. The child was delivered at term via spontaneous vaginal delivery. Immunization was completed according to the standard schedule. There was no family history of bleeding disorders or any other systemic illnesses. There were no known drug allergies. The patient was the third of four children, currently in junior secondary school (JSS1), and reported a daily consumption of refined carbohydrates.

Intraoral examination revealed a well-circumscribed, pedunculated mass located in the region of the left lower first molar. The mass was non-tender, and there was active bleeding from the gum, characterized by bright red blood. Although the bleeding was minimal, it required frequent changes

of the cotton wool pack. The bleeding episodes were effectively controlled with pressure packs soaked in adrenaline. A multidisciplinary management approach was employed, and the patient had blood transfusion, and factor VIII concentrate was administered. After appropriate treatment, an excisional biopsy of the lesion was performed under local anesthesia. A provisional diagnosis of pyogenic granuloma was considered, but laboratory confirmation was not possible as the patient's father was unable to afford the additional costs. The patient was discharged three days postoperatively, with instructions and a planned follow-up for further management.

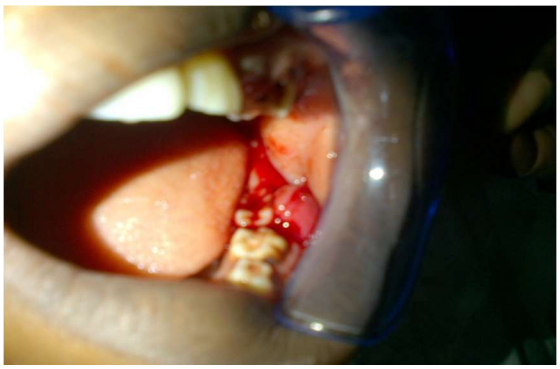

FIGURE 1: Preoperative clinical picture of a pedunculated mass located in the region of the lower left first molar

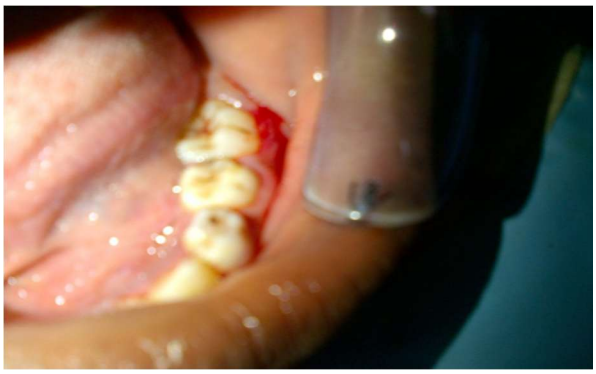

FIGURE 2: Postoperative clinical picture of satisfactory healing of gingivae around the lower left first molar

CASE REPORT 2

A 6-year-old male presented to Paediatric Dental Clinic at Lagos University Teaching Hospital (LUTH) with a complaint of persistent bleeding from the dorsum of the tongue, which occurred following a traumatic injury. The patient had a notable history of excessive post-circumcision bleeding and a similar episode of oral haemorrhage, both of which were managed with suturing at a local health center.

On examination, the patient appeared mildly pale and was still actively bleeding from the site of the laceration. Suturing was performed under local anesthesia, and haemostasis was successfully achieved. Given the patient's history of abnormal bleeding, he was referred to the Pediatric Hematology Clinic for further evaluation and investigations. Available tests in the facility include Factor VIII and IX assays and clotting profile.

After a day, the patient was readmitted to the Children's Emergency Room (CHER) due to recurrent bleeding from the same laceration. Laboratory result revealed a haematocrit of 21%. Although clotting profile assays were requested, they were delayed due to financial constraints. In response to the acute bleeding, the patient was transfused with whole blood, and the laceration was re-sutured. Additionally, the patient was started on intravenous tranexamic acid 15mg/kg 8hourly, for five days to aid in bleeding control. Following stabilization, the patient was discharged with post -operative instructions and plans for follow-up at the clinic.

FIGURE 3: Preoperative clinical picture of laceration on dorsum of tongue

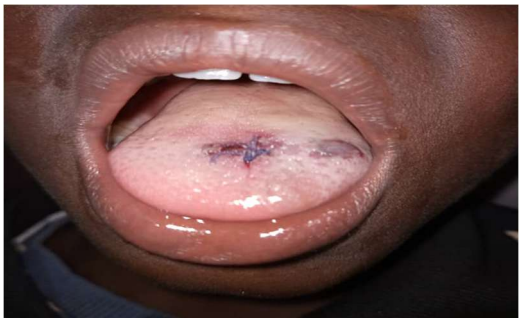

FIGURE 4: Postoperative clinical picture of suture of laceration on dorsum of tongue

LITERATURE REVIEW

Clotting factor VIII (Haemophilia A) or factor IX (Haemophilia B) or factor XI (Haemophilia C) deficiency/defect is the hallmark of haemophilia, an inherited bleeding disorder that is inherited in an x-linked recessive manner for haemophilia A and B and autosomal recessive in haemophilia C. Haemophilia A and B being X-linked recessive disorders primarily affect males.[8] Haemophilia A accounts for 80-85% of all cases of haemophilia, making it the most common of the three. Due to the possibility of persistent bleeding after even minor injuries or dental procedures, the oral and dental consequences of haemophilia are as relevant, even though the majority of the research focuses on the musculoskeletal system.[9]

Regarding residual clotting factor levels, haemophilia is classified as mild, moderate, or severe. Severe patients are defined as those with less than 1% residual factor activity, and they are particularly vulnerable to spontaneous bleeding events, including oral cavity episodes.[10] Lack of clotting factors hinders the production of thrombin, which is necessary for stabilizing fibrin clots and prolongs bleeding even in cases of minor injuries.[11] Often, bleeding occurs once a week, and the condition is typically diagnosed within the first year of life. In moderate cases, there is 1%-5% residual factor level activity. Affected individuals present majorly following trauma but can also bleed spontaneously. In mild haemophilia, there is 6%-40% residual factor level activity. Affected individuals predominantly bleed after trauma, but the condition is often not identified till later in life.[1]

Sometimes the earliest sign of an undetected bleeding disorder occurs in the oral cavity. Typical symptoms include, but are not limited to, submucosal haemorrhages, petechiae, ecchymoses, haematomas of the tongue and lips, spontaneous bleeding of the gingivae and persistent bleeding following tooth eruption/extraction. The likelihood of spontaneous gingival bleeding is worsened by the presence of periodontal disease, which might result from poor oral hygiene brought on by a fear of bleeding.[12]

In addition, during teething or trauma from biting on teething rings, children with haemophilia might experience oral bleeding.[13] Haemophilic pseudo-tumor could occur in the mandible due to recurring sub-periosteal hemorrhage. In spite of the greater likelihood of bleeding, available published data have not typically demonstrated a greater frequency of periodontal disease in individuals with haemophilia in comparison to healthy counterparts. This might be related to better knowledge of preventative care among haemophiliacs.[14] Periodontal disease, if left untreated, can cause persistent bleeding, perpetuating the pattern of exacerbated inflammation as a result of inadequate dental care.[15] Haemarthrosis is frequently observed in the weight-bearing joints of individuals with haemophilia, however, it is uncommon in the temporomandibular joint (TMJ).[16]

There are very few reported cases of TMJ haemarthrosis.[17] Chronic haemophilic TMJ arthropathy may also occur, which requires arthrotomy, arthroscopic adhesion lysis, factor replacement, splint therapy and physical therapy.[18] Haemophilic individuals are at high risk of bleeding during dental operations, particularly invasive ones like tooth extractions and periodontal surgery. To avoid these treatments, it is recommended to prioritize preventive dental care and adherence to daily oral hygiene practices.[19] To lessen the risk of bleeding, factor replacement therapy is frequently used prior to surgery. To reduce postoperative bleeding, antifibrinolytics, notably tranexamic acid or epsilon-aminocaproic acid, may be administered as an extra course of therapy.[20]

To prevent mucosal damage, the use of soft-bristled toothbrushes, chlorhexidine rinses, and atraumatic techniques is advised.[21] Coordination between Paediatric dentists and haematologists is essential for safe and successful therapy[7]. Management of haemophilia A among patients undergoing dental surgery consists of increasing factor VIII levels (by the use of Desmopressin), systemic haemostatic therapy (replacing factor VIII), use of local haemostatic agents and inhibiting fibrinolysis (oral antifibrinolytic agents). Haemophilia B is managed by replacement therapy with highly purified, virally inactivated factor IX concentrates.[20]

More recently, Gene therapy, non-factor replacement therapies and extended half-life clotting factors have improved disease management and minimized the risk of spontaneous bleeding, particularly oral bleeding.[22] Emicizumab, a bispecific monoclonal antibody, has shown effectiveness in lowering the risk of bleeding, providing new options for dental care in patients with haemophilia A.[23] Notwithstanding, in underdeveloped nations or low-resource settings, lack of access to replacement treatment remains a serious barrier for oral healthcare in haemophilic populations.[24]

DISCUSSION

This case series emphasizes the importance of individualized treatment planning and multidisciplinary collaboration in managing dental procedures in patients with haemophilia. Ensuring haemostatic control pre-, intra-, and post-operatively is critical to avoid life-threatening complications. Haemophilia increases the risk of excessive bleeding during dental procedures; therefore, early diagnosis and haematologic support are crucial. Local and systemic haemostatic strategies must be integrated in management plans, which should involve multidisciplinary care to improve patient outcomes.[25] The initial suspicion of haemophilia in a child may be attributed to paediatric dentists, who play a significant role in the management of the condition. Uncontrolled bleeding during or after oral surgery may serve as a pointer to haemophilia, corroborated by a positive history of post-circumcision bleeding,[26] as observed in case two. History taking is especially crucial in our setting, where laboratory tests are either not readily available and/or affordable.

At initial presentation to the clinic, history taking revealed no obstetric or postpartum complications in the first patient. In contrast, in the second reported case, there was a history of prolonged post-circumcision bleeding. Although bloodless circumcision using a diathermic knife – such as the Turkish method – has been documented as a reliable and practical surgical alternative for boys with haemophilia.[27,28] Details regarding the technique used in this case could not be verified, as the accompanying caregiver was unable to provide this information.

Children with haemophilia are prone to recurring incidents of oral bleeding due to the extremely vascular architecture of the oral cavity.[29–31] Common sites of bleeding include the labial frenum and the tongue, often following trauma or induced by dental procedures. Spontaneous gingival bleeding is also frequently observed, triggered by minor stimuli such as tooth brushing, food abrasion, or infection.[3,5] These oral regions contain a dense network of superficial, dilated capillaries, contributing to their susceptibility.[32] Similarly, case one reported bleeding from the gingivae as a result of chronic inflammation, while case two reported bleeding from the tongue secondary to trauma.

While patients with mild to moderate haemophilia are typically managed on an outpatient basis within dental clinics,[10,33,34] these two cases presented in this series necessitated inpatient admission due to haemodynamic instability. This can be attributed to both patients exhibiting significantly reduced haematocrit and haemoglobin (Hb) concentrations, following substantial blood loss before they arrived at the dental facility. As a result, multidisciplinary management involving both a haematologist and a paediatrician was required to stabilize their haematological parameters before proceeding with dental care. In addition, to mitigate post-operative haemorrhage after invasive dental operations, supplementary therapy with tranexamic acid has been advised, as demonstrated in case two.[35] Preventive dental care is a critical component of oral health management in patients with haemophilia, as it significantly reduces the need for invasive treatment and lowers the incidence of emergency visits due to oral bleeding episodes.[5,29] Anticipatory guidance should be provided to caregivers, emphasizing the importance of maintaining oral hygiene and the role of regular, routine dental assessments in preventing complications.[36] Despite these recommendations, financial constraints often act as a barrier to follow-up visits, as was the case with the patients in this report.

CONCLUSION

Haemophilia has a profound impact on oral health, with symptoms ranging from slight mucosal bleeding to life-threatening haemorrhages following dental procedures. Early detection, prevention, and interdisciplinary teamwork are critical for reducing complications. Ongoing research and increasing availability of innovative medicines offer hope for better oral and systemic health outcomes in haemophilic individuals.

Conflicts Of Interest

Nil

Sources of support

None declared.

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