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| RESEARCH ARTICLE

A Rare Cause of Post-circumcision Bleeding in a Male Infant: A Case Report

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| ABSTRACT
We present a male infant with postoperative bleeding following a circumcision procedure due to a rare hematological cause that was treated in our center. Circumcision is a commonly performed procedure. It is a relatively safe with a low overall complication rate. Patients with post circumcision bleeding should have a coagulation profile and those with prolonged or excessive bleeding should undergo a comprehensive hematologic evaluation to look for possible rare undiagnosed bleeding disorders.

| KEYWORDS
"Male Circumcision", "Coagulation", "Bleeding Disorders", "Factor X Deficiency", "FXD", and "Fresh Frozen Plasma".

| ARTICLE INFORMATION
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I. Introduction

Male circumcision is a common procedure done for religious and cultural reasons in this part of the world. Most of which is performed as a day case procedure with minimal risks and complications. Moreover, multiple health benefits have been cited with circumcision leading to increased popularity of the procedure worldwide. Systematic evaluation of English-language peer-reviewed literature from 1995 through 2010 indicates that preventive health benefits of elective circumcision for male newborns outweigh the risks of the procedure. Benefits include significant reductions in the risk of urinary tract infection in the first year of life [(1)]. Previous studies have reported rates of UTI to be 10-20 times higher in uncircumcised than in circumcised boys [(2)] and there is substantive evidence supports the premise that circumcision protects males from HIV infection, penile carcinoma, urinary tract infections and ulcerative sexually transmitted diseases [(3)] with overall complication rate of 0.2 to 5% [(4)]. We report hemorrhagic complication from circumcision in a patient with Factor X deficiency (FXD).

2. Case Presentation

An 11-day old Bahraini male infant, a product of full term normal vaginal delivery, with no previous documented history of bleeding disorder nor other health issues, was brought by his parents to our emergency department with a complaint of circumcision site bleeding. The circumcision was performed 5 days earlier at a private medical center followed by bleeding. Control of the bleeding was attempted by the physician at the private medical center 2 days following the procedure which failed to control the bleeding completely, so the patient was eventually brought to our hospital by his parents.
On examination upon arriving in our emergency department, the infant was hemodynamically stable, not pale, with good hydration status. Genital examination revealed a circumcised penis, with active bleeding from the ventral surface of the penis at the region of the frenulum. Surgical sutures were present in the same areas. The glans penis was pink and both testicles are

scrotal (Figure 1).

Figure (1): Pre-operative active bleeding from the ventral

Laboratory investigations showed a hemoglobin of 11.40 g\dL, a white blood cell count of 14.34 x10^9/L, platelets of 481 x10^9/L, and Reticulocyte% of 0.9%. The coagulation profile upon admission was deranged as follows: prothrombin time and INR showed no coagulation. Prolonged activated partial thromboplastin time (APTT) at 134.50s (N 25-43s) , prolonged APTT Ratio of 4.80s (N 0.9-1.4s), and thrombin Time of 20.20s (N 21.7-49.6s).

The patient was taken urgently by the pediatric surgery team to the operating room for control of the bleeding. The procedure was performed under local anesthesia – 1 ml of 0.5% plain marcaine (bupivacaine) - as penile ring block. The penile mucosal collar was seen to be separated from the penile skin with the presence of previous sutures over the mucosa with active bleeding. The area around the frenulum was also friable, and actively bleeding Fig (1)
Bipolar diathermy was applied to the bleeding points but failed to control the bleeding completely. Arrest of the bleeding was achieved with placement of hemostatic sutures over the frenulum. Skin to mucosa sutures were placed with a combination of 6-0 Monocryl (poliglecaprone 25) and undyed 5-0 vicryl (Polyglactin 910) sutures which helped to stop the bleeding Fig (2) Compressive dressing was also applied around the penis using Coban (Cohesive bandage 3M – self adhering bandage). Upon shifting the patient to the recovery room, the dressing was completely dry and there was no further bleeding during the post-operative stay.

Figure (2): Intra-operative hemostasis achievement with interrupted 6-0 monocryl and 5-0 undyed vicryl sutures.

During the admission, the patient was evaluated and investigated by the Pediatric Hematology team, investigations confirmed the presence of Factor X deficiency (FXD). Ultrasound skull was done to rule out intracranial bleeding and it was negative. The patient received 3 units of fresh frozen plasma during his hospitalization, compressive dressing was removed once his coagulation profile was improved and was discharged on the ninth post operative day in stable condition with no bleeding at the operative site. Patient underwent insertion of right sided internal jugular vein central line in view of the long-term need of recurrent administration of fresh frozen plasma and factor X replacement therapy.

3. Discussion

Circumcision is one of the oldest and most performed surgical procedures in practice today. In a large retrospective review of the Nationwide Inpatient Sample, estimated rates of newborn circumcision have risen from 48.3% nationwide in the years 1988–91 to 61.1% of male newborns from 1997–2001. This represents an increase in incidence of approximately 6.8% per year [(5)]. The procedure involves excision of the penile foreskin using various techniques. It is often used for religious, cultural, or medical purposes. Bleeding is the most common complication of circumcision, with an incidence of 1% in a large retrospective review [(6)]. Most of the post-circumcision bleeding can be controlled with the application of direct pressure or the careful application of silver nitrate. Rarely is wound exploration and suturing is necessary [(7)]. In a retrospective review of the Mayo Clinic Pediatric Hemophilia database, 48 patients with a range of coagulopathies were circumcised. 21 patients had known coagulation disorders, while the remaining 27 patients were diagnosed after prolonged bleeding from their circumcision. There were 11 bleeding complications, three of which were severe and required transfusion of RBCs for severe anemia despite preoperative factor replacement [(8)].

Inherited Factor X deficiency (FXD) is a rare recessive bleeding disorder (incidence 1:1000000) [(11)] .It causes bleeding from the nose, easy bruising, bleeding under the skin, bleeding of the gums, hematuria, and prolonged or excessive bleeding following trauma or surgery, as seen in our case. Factor X, originally named Stuart–Prower factor, is a vitamin K-dependent, liver-produced serine protease that serves a pivotal role in coagulation as the first enzyme in the common pathway to fibrin formation [(9)]. Once the damage to the vessel is done, the endothelial cells release tissue factor, which goes on to activate factor VII to factor VIIa. Factor VIIa goes on to activate factor X into factor Xa. This is the point where both extrinsic and intrinsic

pathways become one [10]. Phenotype diagnosis is based on the concomitant prolongation of the prothrombin time and activated partial thromboplastin time. Through the measurement of plasma level of FX antigen and its coagulant activity, two main types of deficiency can be distinguished: type I (concomitantly low levels of activity and antigen) and type II (low coagulant activity, but normal or borderline antigen levels) [12]. Treatment of FXD depends on the severity of bleeding. Guidelines for the management of FXD and other rare coagulation disorders based on literature and extensive clinical experience have been published by the United Kingdom Haemophilia Centre Doctors' Organization [13]. For minor bleeding symptoms, topical therapies and antifibrinolytic agents may be sufficient [9]. Replacement therapy can be accomplished with fresh frozen plasma (FFP) or plasma- derived FIX concentrates [prothrombin complex concentrates (PCC)] [9].

4. Conclusion

Circumcision is a commonly performed procedure. It is a relatively safe with a low overall complication rate. Patients with post circumcision bleeding should have a coagulation profile and those with prolonged or excessive bleeding should undergo a comprehensive hematologic evaluation to look for possible rare undiagnosed bleeding disorders.

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References

[1] Task force on circumcision of American Academy of Pediatrics. Circumcision policy statement. Pediatrics 1999; 103

[2] To T, Agha M, Dick PT, Feldman W. Cohort study on circumcision of newborn boys and subsequent risk of urinary-tract infection. Lancet. 1998 Dec 5;352(9143):1813-6. doi: 10.1016/S0140-6736(98)02392-7. PMID: 9851381.

[3] Moses, S., Bailey, R.C. and Ronald, A.R., 1998. Male circumcision: assessment of health benefits and risks. Sexually transmitted infections, 74(5), p.368

[4] Baskin LS, Canning DA, Snyder HM Duckett JW. Treating complication of circumcision Pediatric Emerg Care 1996; 12: 62 – 68

[5] Nelson C. P., Dunn R., Wan J., and Wei J. T., The increasing incidence of newborn circumcision: data from the nationwide inpatient sample, Journal of Urology. (2005) 173, no. 3, 978–981, 2-s2.0-13744253882, https://doi.org/10.1097/01.ju.0000145758.80937.7d.

[6] Gee W. F. and Ansell J. S., Neonatal circumcision: a ten year overview: with comparison of the Gomco clamp and the Plastibell device, Pediatrics. (1976) 58, no. 6, 824–827, 2-s2.0-0017030279.

[7] Krill, A.J., Palmer, L.S. and Palmer, J.S., 2011. Complications of circumcision. The scientific world journal, 11(1), pp.2458-2468.

[8] Rodriguez V., Titapiwatanakun R., Moir C., Schmidt K. A., and Pruthi R. K., To circumcise or not to circumcise? Circumcision in patients with bleeding disorders, Haemophilia. (2010) 16, no. 2, 272–276, 2-s2.0-77950252080, https://doi.org/10.1111/j.1365-2516.2009.02119.x.

[9] Brown, D.L. and Kouides, P.A., 2008. Diagnosis and treatment of inherited factor X deficiency. Haemophilia, 14(6), pp.1176-1182.

[10] Chaudhry, R., Usama, S.M. and Babiker, H.M., 2018. Physiology, coagulation pathways.

[11] Herrmann, F.H., Auerswald, G., Ruiz-Saez, A., Navarrete, M., Pollmann, H., Lopaciuk, S., Batorova, A., Wulff, K. and Greifswald Factor X Deficiency Study Group, 2006. Factor X deficiency: clinical manifestation of 102 subjects from Europe and Latin America with mutations in the factor 10 gene. Haemophilia, 12(5), pp.479-489.

[12] Menegatti, M. and Peyvandi, F., 2009, June. Factor X deficiency. In Seminars in thrombosis and hemostasis (Vol. 35, No. 04, pp. 407-415). © Thieme Medical Publishers.

[13] Bolton-Maggs PHB, Perry DJ, Chalmers EA et al. The rare coagulation disorders – review with guidelines for management from the United Kingdom Haemophilia Centre Doctors' Organisation. Haemophilia 2004; 10: 593–628.