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CASE REPORT

Glans ischemia after circumcision in children: Two case reports

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Abstract

BACKGROUND

Circumcision refers to the removal of the skin covering the tip of the penis and is one of the most common surgical procedures performed in childhood. Even though circumcision is a well-standardized operation, several minor and major complications may be experienced by paediatric surgeons. Glans ischemia (GI) has been widely reported in the paediatric literature as a complication following circumcision. Nonetheless, etiopathogenesis of GI is not well defined and management guidelines are lacking.

CASE SUMMARY

We describe our experience with this rare and scary complication using subcutaneous enoxaparin alone or in association with a topical vasodilator.

CONCLUSION

Hypothetical causes and different management strategies are discussed.

Key Words: Circumcision; Children; Complications; Glans penis; Ischemia; Case report

Core Tip: Glans ischemia (GI) after circumcision is a rare complication, which has been widely described by paediatric surgeons in the modern literature. To date, etiopathogenesis of GI is not well defined and management guidelines are lacking. In order to achieve a prompt diagnosis and to start appropriate treatment, an accurate postoperative medical assessment and parental education are crucial before hospital discharge for children undergoing circumcision.

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INTRODUCTION

Circumcision refers to the surgical removal of the foreskin covering the glans and is one of the most common paediatric procedures. The complication rate after circumcision in childhood varies between 0% and 16%[1]. Minor complications include penile shaft swelling, bleeding, meatal stenosis, recurrent preputial stenosis and unsatisfactory cosmetic appearance. Major complications reported in the literature are glans or penile amputation, septicaemia, and urethrocutaneous fistulas[1,2]. Glans ischemia (GI) after circumcision is an extremely rare but scary complication in children[3]. We describe our experience with two cases of GI after circumcision in males aged 8 and 10 years old. Hypothetical causes and different treatment strategies are debated.

CASE PRESENTATION

Chief complaints

Case 1: An 8-year-old boy underwent circumcision at our paediatric surgery department for a true phimosis. The child's medical history was uneventful. Surgery was performed under general anaesthesia with a dorsal nerve penile block using mepivacaine. During surgery, a monopolar electrocautery was used to excise the excessive foreskin and to execute the frenulotomy. The coronal suture was performed with 5-0 interrupted absorbable sutures. No excessive bleeding was noted neither during intervention nor in the immediate post-operative course. No compressive bandaging was used.

Case 2: A 10-year-old boy presented to our paediatric outpatient clinic for a true phimosis. Personal history was unremarkable, except for childhood vitiligo. Circumcision was performed under general sedation with spinal anaesthesia. Bipolar electrocautery was used and coronal suture was performed with 5-0 interrupted absorbable stitches. No compressive bandaging was applied. No excessive bleeding was noted neither during intervention nor in the immediate postoperative course. Minimum glans swelling was reported two hours after surgery.

History of present illness

Phimosis.

History of past illness

Case 1: Uneventful.

Case 2: Unremarkable, except for childhood vitiligo.

Personal and family history

Unremarkable.

FINAL DIAGNOSIS

Case 1

At the clinical examination 6 h after surgery, an ischemic appearance of the glans was documented, without pain or difficulty to urinate. A colour doppler imaging (CDI) showed normal flow in the dorsal penile artery.

Case 2

Four hours after surgery, an ischemic appearance of the glans was documented (Figure 1A). Whole blood count and blood clotting were checked and found to be within normal ranges.

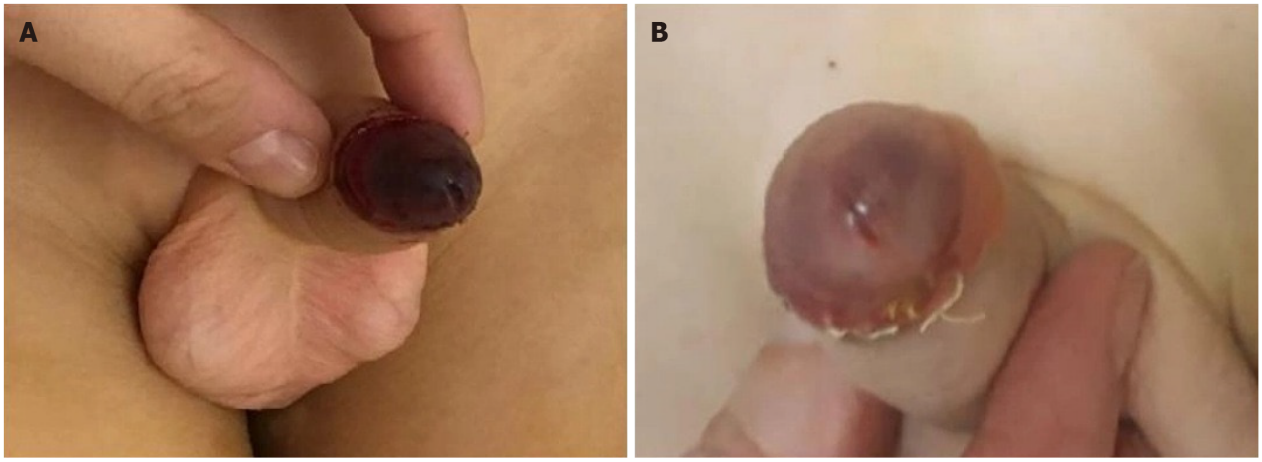

Figure 1 Close up view of circumcision procedure. A: Close up view of a glans ischemia four hours after the circumcision procedure; B: Glans appearance few days after starting therapy with subcutaneous enoxaparin injections.

Case 1

Subcutaneous enoxaparin 2000 UI injection was started and continued once a day for 5 d. Moreover, a galenic preparation of nitric oxide ointment was applied on the glans once a day for a week.

Case 2

Anticoagulant therapy was started with subcutaneous enoxaparin 3000 UI once a day for 5 d.

OUTCOME AND FOLLOW-UP

Case 1

The child was discharged home on postoperative day 6 when an improvement of the GI was noted. Complete restitution integrum was achieved one month after surgery.

Case 2

The colour of the glans rapidly improved to reddish (Figure 1B), and the patient was discharged home on postoperative day 4. At one-month follow-up, the penis and glans were found to be in a normal status.

DISCUSSION

Circumcision is a common paediatric surgical procedure; approximately 0.5% of patients require a repeat surgery. The most frequent complication reported in patients undergoing circumcision is haemorrhage (0.8%), with more than 60% of cases requiring surgical revision[2].

GI after circumcision has been widely reported in the paediatric literature. However, the etiopathogenesis of GI is not well known. The most commonly reported cause for GI is dorsal nerve block using local anaesthetics with or without vasoconstrictor agents[3]. Compression dressing, tight sutures, and excessive use of monopolar electrocautery are other potential reasons for GI after circumcision[3,4]. In our first case, anaesthesia was achieved by a dorsal penile nerve block; during surgery, a monopolar electrocautery device was used. In the second case, a spinal block and bipolar electrocautery were used. After surgery, we routinely use a combination of antibiotic and corticosteroid ointment on the coronal suture and the penis is gently covered with gauze but without any tight circumferential bandage. Notably, in a similar case, Efe *et al*[5] reported an elevated D-dimer level, with restoration to normal level after five days of enoxaparin treatment, suggesting a penile vascular thrombosis even though CDI showed normal penile and glandular blood flow. Conversely, both Karaguzel *et al*[4] and Gnatzy *et al*[6] reported their experiences, describing two cases of acute GI after circumcision with a normal level of D-dimer and good penile blood

flow at CDI. Regarding our cases, the first one showed normal blood flow at CDI but D-dimer value was not checked. In the second case, the D-dimer level was normal but CDI was not performed. Many authors have reported normal penile blood flow at CDI [5-8], and only one case in the paediatric literature described reduced penile blood flow[9]. Therefore, it is questionable whether a thrombosis may be responsible for GI after circumcision, as suggested by Efe *et al*[5], or whether a transient vasospasm of the dorsal artery may be to blame. Moreover, doubt persists regarding whether the use of monopolar electrocautery in our first case could have played a role in the development of GI.

To date, several treatment options for GI are reported in the literature, but a defined protocol or guidelines are still lacking. Some authors reported a successful outcome with endovenous or oral administration of pentoxifylline (PTX), alone or in association with other therapeutic stratagems. PTX is a hemorheological agent which improves the viscosity of blood and is used in peripheral vascular and cerebrovascular insufficiency [4,9,10]. Comparatively, caudal block reduces sympathetic tone, improves arterial supply and venous drainage, and has been proposed as the sole therapeutic strategy [7], or in association with intracavernous injection of glycerol trinitrate, to improve postarteriolar smooth muscle relaxation[11]. Furthermore, Aminsharifi *et al*[11] reported the use of topical testosterone, which has been shown to improve the vascular density of foreskin *in vitro*, in two cases of delayed GI after circumcision, which resulted in complete healing after one month. Selective angiography with intra-arterial injection of a vasodilator agent has been reported by Gnatzy *et al*[6] in association with oral sildenafil and infusion of L-arginine hydrochloride and unfractionated heparin. Lastly, as previously reported, anticoagulant therapy using enoxaparin has been effective in case of GI after circumcision[5]. In both our cases, we administered subcutaneous enoxaparin injection once a day for 5 d with complete resolution of GI. Notably, in the first case, a topical vasodilator was added and the complete resolution required additional days compared with the second case.

CONCLUSION

In conclusion, although a unique causative factor for GI after circumcision cannot be identified, a favourable outcome has been reported in nearly all cases. The unfavourable outcomes reported in literature are due to delayed discovery of the ischemic condition or late presentation of the patients back to the hospital. Consequently, we strongly recommend that discharge home should be preceded by an accurate medical assessment and should not be scheduled until at least 6 h postoperatively. Additionally, parents and patients should be well instructed in evaluating any possible signs of complication in the postoperative course. Lastly, we recommend rigorously following-up patients experiencing GI after circumcision for at least the first month after surgery.

REFERENCES

1 **Weiss HA**, Larke N, Halperin D, Schenker I. Complications of circumcision in male neonates, infants and children: a systematic review. *BMC Urol* 2010; **10**: 2 [PMID: 20158883 DOI: 10.1186/1471-2490-10-2]

2 **Cathcart P**, Nuttall M, van der Meulen J, Emberton M, Kenny SE. Trends in paediatric circumcision and its complications in England between 1997 and 2003. *Br J Surg* 2006; **93**: 885-890 [PMID: 16673355 DOI: 10.1002/bjs.5369]

3 **Ozzeybek D**, Koca U, Elar Z, Olguner M, Hakgüder G. Glycerol trinitrate plus epidural sympathetic block in the ischemia of glans penis. *Anesth Analg* 1999; **89**: 1066 [PMID: 10512297 DOI: 10.1097/00000539-199910000-00053]

4 **Karaguzel E**, Tok DS, Kazaz IO, Gur M, Colak F, Kutlu O, Ozgur GK. Postcircumcisional ischemia of the glans penis treated with pentoxifylline. *Case Rep Urol* 2013; **2013**: 278523 [PMID: 23431492 DOI: 10.1155/2013/278523]

5 **Efe E**, Resim S, Bulut BB, Eren M, Garipardic M, Ozkan F, Ozkan KU. Successful treatment with enoxaparin of glans ischemia due to local anesthesia after circumcision. *Pediatrics* 2013; **131**: e608-e611 [PMID: 23319528 DOI: 10.1542/peds.2012-1400]

6 **Gnatzy R**, Fuchs J, Siekmeyer M, Beeskow AB, Gosemann JH, Lacher M. Glans Ischemia after Circumcision in a 16-Year-Old Boy: Full Recovery after Angiography with Local Spasmolysis, Systemic Vasodilatation, and Anticoagulation. *European J Pediatr Surg Rep* 2018; **6**: e66-e69 [PMID: 30276065 DOI: 10.1055/s-0038-1667330]

7 **Kaplanian S**, Chambers NA, Forsyth I. Caudal anaesthesia as a treatment for penile ischaemia

following circumcision. *Anaesthesia* 2007; **62**: 741-743 [PMID: 17567354 DOI: 10.1111/j.1365-2044.2007.05060.x]

8 **Sterenberg N**, Golan J, Ben-Hur N. Necrosis of the glans penis following neonatal circumcision. *Plast Reconstr Surg* 1981; **68**: 237-239 [PMID: 7255584 DOI: 10.1097/00006534-198108000-00022]

9 **Cárdenas Elías MÁ**, Vázquez Rueda F, Jiménez Crespo V, Siu Uribe A, Murcia Pascual FJ, Betancourth Alvarenga JE, Paredes Esteban RM. [An unexpected complication: glans ischemia after circumcision. Review of the literature]. *Cir Pediatr* 2016; **29**: 127-130 [PMID: 28393509]

10 **Aslan A**, Karagüzel G, Melikoglu M. Severe ischemia of the glans penis following circumcision: a successful treatment *via* pentoxifylline. *Int J Urol* 2005; **12**: 705-707 [PMID: 16045570 DOI: 10.1111/j.1442-2042.2005.01129.x]

11 **Aminsharifi A**, Afsar F, Tourchi A. Delayed glans necrosis after circumcision: role of testosterone in salvaging glans. *Indian J Pediatr* 2013; **80**: 791-793 [PMID: 22752705 DOI: 10.1007/s12098-012-0820-y]

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