CASE REPORT

High-flow priapism following perineal trauma in a child

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SUMMARY

Priapism is a rare condition in children and can be classified as high or low flow. We present a case of traumatic high-flow priapism that was successfully managed by selective embolisation of a branch of the internal pudendal artery. The pertinent clinical features are antecedent trauma and prolonged painless partial erection. The investigation of choice is Doppler ultrasound. Management can either be conservative, radiological or surgical. While conservative management can be safely attempted for 6 weeks, given the nature of the condition, early intervention is often required. Radiological embolisation is the first line and is successful in 97% of cases. Surgery carries a higher morbidity and is only reserved in cases where repeated embolisation has failed. This case highlights the importance of a thorough history and careful interpretation of imaging with protocolised follow-up of patients by a paediatric urologist for early and accurate diagnosis of erectile dysfunction and prompt intervention to prevent future complications.

BACKGROUND

This case is rare and required multidisciplinary input at a tertiary referral centre.

CASE PRESENTATION

A 7-year-old child presented with a 10-day history of a painless erection. The mechanism of injury was a fall onto a bike crossbar sustaining direct perineal and penile trauma. Initial perineal pain subsided after several hours and, subsequently, the patient developed tumescence, which persisted over 10 days. He denied urinary symptoms or haematuria. Bloods investigations were unremarkable.

Ultrasound at 10 days showed features suggestive of a pseudoaneurysm at the base of the left corpus cavernosum. At this point, he was referred to a tertiary paediatric centre with both urological and interventional radiology expertise. Repeat ultrasound 2 days later demonstrated similar characteristics of pseudoaneurysm (yin-yang sign) with arterialising flow extending into the cavernosa ([figure 1](#)). Given the duration of symptoms, and lack of clinical and radiological improvement, intervention was undertaken. Pelvic angiography demonstrated an arteriocavernosal fistula at the base of the left corpus cavernosum, arising from a branch of the right internal pudendal artery. No arteriocavernosal fistula was demonstrated from the left side. Microcoils were used to successfully embolise the fistula ([figures 2 and 3](#)). Following embolisation, the patient was partially detumesced with a soft glans. Repeat imaging using Doppler ultrasound the following day showed no evidence of the fistula, and normal flow at the base of the penis. The child made a gradual and complete recovery over the next few days.

INVESTIGATIONS

- Doppler ultrasound
- Angiography

DIFFERENTIAL DIAGNOSIS

- Idiopathic priapism
- Blood dyscrasias
- Medication
 - PDE-5 inhibitors
 - Hormones (eg, testosterone)
 - Antipsychotics, antidepressants

Figure 1 (A) Transverse high-resolution sonogram of base of penis, probe placed on ventral aspect. There is engorgement of the left corpus cavernosum (arrow). (B) Transverse sonogram obtained more caudally shows a hypoechoic space medially within the left crus of the penis (arrow). Swirling echoes were seen within. (C) Longitudinal oblique sonogram with colour Doppler imaging through left crus of penis. There is pulsatile colour flow within the abnormal space in the medial aspect of the crus in a 'yin-yang' pattern (arrow), as seen in a pseudoaneurysm. Pulsatile colour flow extends from this into the engorged spaces of the corpus cavernosum (arrowheads).

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Figure 2 Digital subtraction arteriogram from selective right internal iliac artery injection: (A) Unsubtracted image (B) subtracted image (C) late subtracted image. A branch of the right internal pudendal artery supplies the arteriocavernosal fistula (arrows). On the late image, contrast 'blushes' into the corpus cavernosum.

- Antihypertensives (including α -blockers)
- Erythropoietin
- Anaesthetic (including spinal)
- Recreational (alcohol, cocaine and marijuana)
- ▶ Metabolic conditions
 - Henoch-Schonlein purpura
 - Haemodialysis
 - Parenteral nutrition
 - Fabry's disease
- ▶ Tumours
- ▶ Neurological conditions

Figure 3 Selective arteriography of right internal pudendal artery following deployment of two microcoils within the fistula (arrow). Flow into the fistula has been arrested.

OUTCOME AND FOLLOW-UP

The patient was discharged and will be followed by urology service locally.

DISCUSSION

Priapism is defined as prolonged full or partial erection lasting over 4 h. Priapism in children is reported to have an incidence of 1.5 per 100 000 per annum, of which traumatic priapism accounts for 10%.¹ It is caused by an alteration of penile blood flow, and is commonly classified into high and low flow. High-flow priapism is typically caused by trauma, leading to formation of an arteriocavernosal fistula and increased arterial flow. The veno-occlusive mechanism is predominantly intact and ischaemia is not a feature. Patients are pain-free and erection is typically incomplete. In contrast, in low-flow priapism, the veno-occlusive mechanism is abnormal, resulting in stasis of deoxygenated blood in the corpus cavernosum, leading to ischaemia. This classically presents acutely with severe pain and carries a higher risk of future complications such as erectile dysfunction. Doppler flow ultrasound, the most widely used imaging modality, has high sensitivity, detecting nearly 100% of arteriovenous fistulae in children.^{1 2} History and ultrasound features are central in the investigation of priapism, in order to identify those at risk of developing ischaemia. The aim of management of all patients with priapism is to achieve detumescence and preserve erectile function. Management options in high-flow priapism include conservative, radiological and surgical. Conservative strategies using ice packs and perineal compression are believed to reduce blood flow and aid thrombus formation across the fistula, and can be used safely for up to 6 weeks. Radiological embolisation of the internal pudendal artery via selective angiography is successful in 80% of children, the first time, with an overall success rate of 97% after a second embolisation.^{1 2} Surgical management with transcorporeal fistula ligation is only reserved in cases where repeated embolisation has failed, as it carries a higher risk of future erectile dysfunction. In adult practice, patients are followed up using questionnaires about sexual activity. In a prepubescent child, such questionnaires are not applicable, and therefore the current literature is lacking in long-term outcome data. Although there are no clear guidelines, we recommend that in non-adult patients, erectile function after the procedure can be evaluated with the control of morning erections, and are followed up by a paediatric urologist to identify erectile dysfunction early.

Learning points

- ▶ All patients presenting with priapism should have a Doppler ultrasound.
- ▶ High-flow priapism is classically painless, with longer duration and partial erection.
- ▶ Conservative management may be appropriate for up to 6 weeks.
- ▶ Highly selective internal pudendal artery embolisation is successful in the majority of patients.

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REFERENCES

- 1 Donaldson JF, Rees RW, Steinbrecher HA. Priapism in children: a comprehensive review and clinical guideline. *J Pediatr Urol* 2014;10:11–24.
- 2 Cherian J, Rao AR, Thwaini A, *et al.* Medical and surgical management of priapism. *Postgrad Med J* 2006;82:89–94.

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