

Cycling Trauma as a Cause of Arterial Priapism in Children and Teenagers

Aldo Franco De Rose, MD,¹ Irene Paraboschi, MD,² Guglielmo Mantica, MD,¹⁻³ Alexander Szpytko, MD,³ Hilgard Ackermann, MD,³ Giovanni De Caro,⁴ Carlo Terrone,¹ Girolamo Mattioli²

¹Department of Urology, Ospedale Policlinico San Martino – Istituto di Ricerca e Cura a Carattere Scientifico per l'Oncologia, University of Genova, Genova, Italy; ²University of Genoa, Pediatric Surgery Unit, DINOEMI Istituto Giannina Gaslini, Genova, Italy; ³Department of Urology, Stellenbosch University and Tygerberg Hospital, Cape Town, South Africa; ⁴Department of Vascular and Interventional Radiology, Galliera Hospital, University of Genova, Genova, Italy

Bicycle riding has multiple beneficial cardiovascular effects; however, it is a well-documented source of significant urologic injuries. Priapism is a rare condition in children, and occurs primarily because of congenital hematologic diseases or adverse drug reactions. A pediatric clinical case and literature review of a high-flow priapism secondary to cycling trauma is described here to highlight their etiopathologic correlation. Bicycle riding trauma is a rare but possible cause of high-flow priapism in children, and a high index of suspicion should ensure appropriate management.

[Rev Urol. 2017;19(4):273–277 doi: 10.3909/riu0768]

© 2018 MedReviews®, LLC

KEY WORDS

Priapism • Cycling • Trauma • Pediatric

Priapism is a prolonged penile erection unrelated to sexual stimuli, primarily resulting from lacunar venous outflow obstruction or, less commonly, from arteriolacunar fistula formation. It represents an infrequent clinical condition in the pediatric population, the underlying causes of which may be difficult to identify. A case of childhood arterial priapism that occurred after cycling injury is reported, followed by a literature review to ensure appropriate management.

Case Report

A 12-year-old white boy presented to our outpatient clinic with a 6-month history of painless penile erection. The erection was partial, although persistent.

The patient's past medical history showed no features of congenital hematologic disease, drug intake, or prior surgery. A detailed anamnesis disclosed a low-energy traumatic cycling injury that occurred 2 days prior to the initiation of the persistent penile

erection. The patient described having injured his genitalia with low energy against the bicycle wheel at an approximate velocity of 20 to 30 km/h. The trauma resulted in minimal pain, of the type that often occurs among cyclists. He continued cycling for the next 30 minutes, and did not use any medications or ice packs. The patient reported a mild dull pain since the day of injury; however, no signs of significant perineal or penile hematoma were evident.

The prolonged delay between the traumatic incident and the medical consult was due to the patient feeling shy and embarrassed, in particular with regard to disclosure of the problem to his parents. Lack of understanding regarding the gravity of the condition and its requirement of time-sensitive management was also an issue. On clinical evaluation, the patient had a rigid, non-tender corpora cavernosa, with a soft glans and corpus spongiosum.

Routine laboratory test results (full blood count; urea, creatinine, and electrolyte levels; and sickle cell disease screening) were within normal limits, excluding the onset of any acquired hematologic disorders. Diagnostic intracorporeal blood gas analysis revealed bright red blood with a high oxygen saturation ($PO_2 = 87$ mm Hg; $PCO_2 = 41$ mm Hg; $Ph = 7.36$).

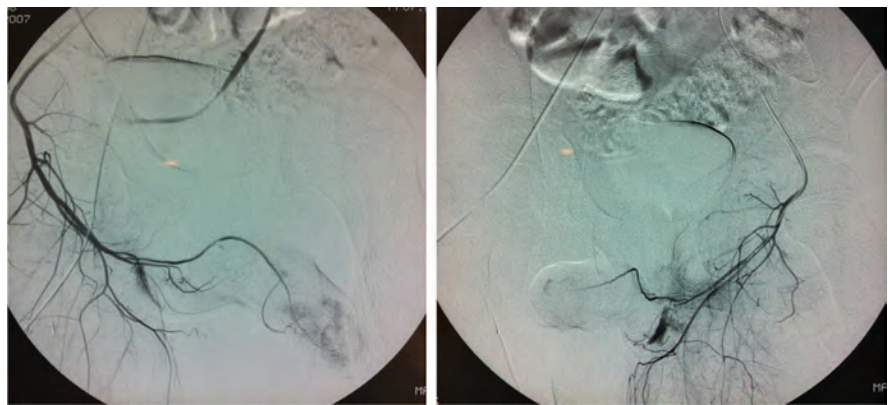


Figure 1. Selective arteriography of the right and left pudendal artery. A post-traumatic arteriolacunar fistula is easily visible in a contrastographic blush area to the left.

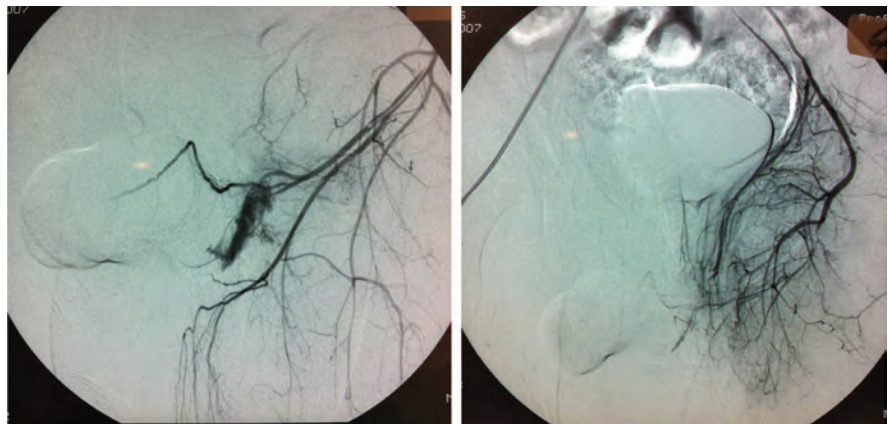


Figure 2. Panoramic arteriography after embolization with an absorbable hemostatic gelatin sponge proximal to the arteriolacunar fistula.

an absorbable hemostatic gelatin sponge. The immediate result was complete detumescence with return to a completely flaccid penis. A follow-up pelvic arteriogram was performed 2 weeks after embolization, and showed the absence of any

pathophysiologic types. The most common ischemic (or low-flow) subtype is characterized by a painful erection of the corpora cavernosa due to venous occlusion and vascular stasis, primarily associated with hematologic disorders (eg, sickle cell disease, hematopoietic malignancies, hypercoagulable states) or adverse drug reactions.¹ Sickle cell anemia accounts for up to 65% of all priapism episodes occurring in children,² whereas 29% to 42% of patients with sickle cell anemia develop priapism.³ In these cases, prompt intervention is required to avoid development of serious complications (eg, irreversible sexual dysfunction and disfigurement with subsequent emotional sequelae).

Stuttering priapism is relatively rare and mostly associated with

The patient underwent pelvic arteriography, which clearly depicted a post-traumatic arteriolacunar fistula coming from the left bulbourethral artery.

On penile Doppler ultrasonography, high arterial and venous flow in the corpora were shown, although pathologic fistula was not detected. The patient underwent pelvic arteriography, which clearly depicted a post-traumatic arteriolacunar fistula coming from the left bulbourethral artery (Figures 1 and 2).

The fistulous communication was selectively embolized with

fistula and patency of the left dorsal artery. At his 1-year follow-up visit, the boy reported normal spontaneous and reflexogenic erections with no evidence of priapism recurrence.

Discussion

Priapism is a prolonged penile erection unrelated to sexual arousal. There are three different

various hematological disorders, including sickle cell disease.

The nonischemic (or high-flow) subtype is the result of arteriolacunar fistula formation with unregulated arterial filling of the corpora

smooth-muscle damage and fibrosis are possible.^{6,7}

The diagnostic work-up of priapism is based on a detailed history and physical examination, followed by intracorporeal blood gas

The diagnostic work-up of priapism is based on a detailed history and physical examination, followed by intracorporeal blood gas analysis and color Doppler ultrasonography.

cavernosa, primarily occurring 24 to 48 hours after severe perineal blunt trauma and initiated by sexual stimuli.⁴ Because tissue oxygenation is usually preserved, the prognosis is typically favorable, even in the case of delayed treatment.⁵ However, cavernosal

analysis and color Doppler ultrasonography. For arterial priapism, the definitive diagnosis is by standard or magnetic resonance imaging pelvic angiography,^{8,9} which can detect arteriolacunar fistula.

Although it has established cardiovascular beneficial effects,

bicycle riding is also a well-known cause of urologic disorders in adulthood (including nerve entrapment syndromes, erectile dysfunction, urethritis, prostatitis, hematuria, and spermatic cord torsion),¹⁰⁻¹² among which high-flow priapism is included.¹⁰⁻¹⁴ Priapism is a rare but possible clinical condition in children; clinical work-up can be extremely arduous when the most frequent causes are excluded in this age group. A case report and literature review of cycling-induced priapism is reported here (Table 1) to draw attention to this rare cause for pediatric consultation.

Clinical onset of cycling-induced priapism depends on the entity of

TABLE 1

Literature Review of Cycle-induced High-flow Priapism

Study	Age (y)	Site or Cause of Injury	Onset After Trauma	Treatment	Detumescence Time
Shankar KR et al ¹⁷	8	Handlebar	24 h	Selective embolization of the internal pudendal artery	Immediate
Moscovici J et al ¹⁶	8	Bicycle frame	72 h	Spontaneous resolution	21 d
	7	Bicycle frame	6 d	Twice failed gelatin sponge embolizations followed by spontaneous resolution	30 d
Golash A et al ¹²	35	Handlebar	10 d	Gelatin sponge embolization	36 h
De Rose AF et al ¹³	27	Top tube	Immediate	Previous failed gelatin sponge embolization followed by successful microcoil embolization	Immediate
	36	Top tube	Several hours	Gelatin sponge embolization	60 d
Emir L et al ¹⁵	9	Handlebar	7 d	Conservative (ice packs, imipramine hydrochloride)	60 d
Corbetta JP et al ²⁹	11	Fall	Immediate	Spontaneous resolution	27 d
Burns J et al ²⁷	7	Crossbar	Immediate	Microcoil embolization	immediate
Ul Islam J et al ¹⁴	22	Crossbar	Not reported	Previous failed conservative treatment (pressure dressing) followed by gelatin sponge embolization	Immediate
Present case	12	Crossbar	24 h	Gelatin sponge embolization	Immediate

the vascular lesion: severe arterial damage usually leads to immediate penile tumescence, whereas small injuries cause priapism only when penile blood flow is increased during spontaneous erection, removing the clot that was previously masking the arteriolacunar fistula.¹¹

Both conservative¹⁵ (mechanical or pharmacologic) and invasive¹⁶⁻²⁴ (interventional radiologic and surgical) therapeutic options are described in children, even in cases reporting spontaneous resolution.^{8,16} However, the current

Although most pediatric cases are due to venous outflow obstruction associated with hematologic disorders, the post-traumatic arterial subtype is also possible. Recognition

Most pediatric patients treated with both the conservative approach and embolization report complete detumescence and preserved functional results after a short-term follow-up period.

of the latter form, particularly in the pediatric population, may be challenging due to its delayed onset, its intermittence, and the absence of pain. Furthermore, the embarrassment a teenager may feel

Most pediatric patients treated with both the conservative approach and embolization report complete detumescence and preserved functional results after a short-term

Nonabsorbable ... materials should be used in case of failure of embolization with absorbable materials, due to the increased risk of erectile dysfunction.

accepted standard is angiographic superselective embolization (with absorbable autologous clot or gelatin sponge), which was safely demonstrated in our pediatric case. Nonabsorbable (microcoils or N-butyl-cianoacrilato) materials²⁵ should be used in case of failure of embolization with absorbable materials, due to the increased risk of erectile dysfunction.²⁶

Because priapism is an uncommon problem in childhood, its management could be a clinical challenge for pediatric urologists.

may prevent him from disclosing the problem to his parents, further delaying diagnosis and management. This delay can often be as long as days or weeks^{27,28} and is usually related to low-intensity trauma; it is not necessarily associated with other symptoms (eg, hematoma, hematuria)^{27,29} and therefore can be inadequately evaluated by children and parents. For all these reasons, a delayed diagnosis is possible, which can lead to irreversible penile changes with future sexual problems.

follow-up period.^{15-17,27,29} When the onset is immediate, it is possible to delay the invasive approach and wait for possible spontaneous resolution of the disease. Alternatively, when conservative management fails or there is delay in presentation to the pediatric/urologic department beyond 6 weeks,²⁷ as in our case, an arteriography with embolization becomes mandatory. An imaging control study 1 to 2 weeks after the embolization is advisable. Doppler sonography should be the first choice, but a repeat angiogram may be indicated in specific cases. The absence of fistula detected at the first sonography, and the huge delay before treatment, coupled with an increased risk of recurrence, led us to choose the execution of control arteriography to ensure the complete resolution of the fistula.

Every day, millions of children practice cycling. Although it has

MAIN POINTS

- The bicycle is a widely used tool among children, both for sport and play. Cycle riding is a well-documented source of significant urologic injuries, although it has significant health benefits. One of the potential urologic disorders due to cycle riding is nonischemic priapism.
- Nonischemic priapism is a rare condition, especially in children and teenagers. Typically it is the result of an arteriolacunar fistula formation with unregulated arterial filling of the corpora cavernosa. Even if high-flow priapism is not a urologic emergency, it can lead to irreversible damages to the corpora cavernosa if it is not properly treated.
- High-flow priapism, both for the peculiarity of its intermittent presentation and for the embarrassment that children may have in speaking about it to their parents, can be misdiagnosed and underestimated. For this reason, and because of the wide use of bicycles among children, urologists and pediatricians should always investigate the presence of the disorder.

cardiovascular benefits, the perineal position on a bicycle exposes boys and men to the risk of recurrent blunt trauma with resultant urogenital injuries. To ensure prompt diagnosis and management, pediatricians should be aware that, though uncommon, childhood high-flow priapism could be related to cycling-related trauma. Early recognition and appropriate treatment of this distinct entity is important as it is associated with excellent functional prognosis. ■

References

- Lue TF, Hellstrom WJ, McAninch JW, Tanagho EA. Priapism: a refined approach to diagnosis and treatment. *J Urol*. 1986;136:104-108.
- Bivalacqua TJ, Musicki B, Kutlu O, Burnett AL. New insights into the pathophysiology of sickle cell disease-associated priapism. *J Sex Med*. 2012;9:79-87.
- Burnett AL. Therapy insight: priapism associated with hematologic dyscrasias. *Nat Clin Pract Urol*. 2005;2:449-456.
- Witt MA, Goldstein I, Saenz de Tejada I, et al. Traumatic laceration of intracavernosal arteries: the pathophysiology of nonischemic, high flow, arterial priapism. *J Urol*. 1990;143:129-132.
- Kim KR, Shin JH, Song HY, et al. Treatment of high-flow priapism with superselective transcatheter embolization in 27 patients: a multicenter study. *J Vasc Interv Radiol*. 2007;18:1222-1226.
- Savoca G, Pietropaolo F, Scieri F, et al. Sexual function after highly selective embolization of cavernous artery in patients with high flow priapism: long-term followup. *J Urol*. 2004;172:644-647.
- Broderick GA, Kadioglu A, Bivalacqua TJ, et al. Priapism: pathogenesis, epidemiology, and management. *J Sex Med*. 2010;7:476-500.
- Corbetta JP, Durán V, Burek C, et al. High flow priapism: diagnosis and treatment in pediatric population. *Pediatr Surg Int*. 2011;27:1217-1221.
- Eracleous E, Kondou M, Aristidou K, et al. Use of Doppler ultrasound and 3-dimensional contrast-enhanced MR angiography in the diagnosis and follow-up of post-traumatic high-flow priapism in a child. *Pediatr Radiol*. 2000;30:265-267.
- Aitken SA, Biant LC, Court-Brown CM. Recreational mountain biking injuries. *Emerg Med J*. 2011;28:274-279.
- Leibovitch I, Mor Y. The vicious cycling: bicycling related urogenital disorders. *Eur Urol*. 2005;47:277-286.
- Golash A, Gray R, Ruttley MS, Jenkins BJ. Traumatic priapism: an unusual cycling injury. *Br J Sports Med*. 2000;34:310-311.
- De Rose AF, Giglio M, De Caro G, et al. Arterial priapism and cycling: a new worrisome reality? *Urology*. 2001;58:462.
- Ul Islam J, Browne R, Thornhill J. Mountain bikers priapism: a rare phenomenon. *Ir Med J*. 2014;107:21-22.
- Emir L, Tekgöl S, Karabulut A, et al. Management of post-traumatic arterial priapism in children: presentation of a case and review of the literature. *Int Urol Nephrol*. 2002;34:237-240.
- Moscovici J, Barret E, Galinier P, et al. Post-traumatic arterial priapism in the child: a study of four cases. *Eur J Pediatr Surg*. 2000;10:72-76.
- Shankar KR, Babar S, Rowlands P, Jones MO. Post-traumatic high-flow priapism: treatment with selective embolisation. *Pediatr Surg Int*. 2000;16:454-456.
- Chung E, McKnight J, Hosken B. Post traumatic prepubertal high-flow priapism: a rare occurrence. *Pediatr Surg Int*. 2008;24:379-381.
- Miller SF, Chait PG, Burrows PE, et al. Post-traumatic arterial priapism in children: management with embolization. *Radiology*. 1995;196:59-62.
- Mizuno I, Fuse H, Junicho A, Kageyama M. An experience of percutaneous embolization to post-traumatic arterial priapism in a child. *Int Urol Nephrol*. 2001;32:695-697.
- Bastuba MD, Saenz de Tejada I, Dinlenc CZ, et al. Arterial priapism: diagnosis, treatment and long-term followup. *J Urol*. 1994;151:1231-1237.
- Visvanathan K, Burrows PE, Schillinger JF, Khoury AE. Posttraumatic arterial priapism in a 7-year-old boy: successful management by percutaneous transcatheter embolization. *J Urol*. 1992;148(2 Pt 1):382-383.
- Stock KW, Jacob AL, Kummer M, et al. High-flow priapism in a child: treatment with superselective embolization. *AJR Am J Roentgenol*. 1996;166:290-292.
- Jameson JS, Terry TR, Bolia A, Johnstone JM. An unusual case of priapism in a child: diagnosis and treatment. *Br J Urol*. 1996;77:462-463.
- Oztürk MH, Gümüş M, Dönmez H, et al. Materials in embolotherapy of high-flow priapism: results and long-term follow-up. *Diagn Interv Radiol*. 2009;15:215-220.
- Salonia A, Eardley I, Giuliano F, et al; European Association of Urology. European Association of Urology guidelines on priapism. *Eur Urol*. 2014;65:480-489.
- Burns J, Rajendran S, Calder A, Roebuck D. High-flow priapism following perineal trauma in a child [published online May 12, 2015]. *BMJ Case Rep*. doi: 10.1136/bcr-2014-208694.
- Molina Escudero R, Hernández Atance J, Ballesteros García Mdel M, Páez Borda Á. [Posttraumatic high-flow priapism in pediatric patient treated with selective arterial embolization]. *Arch Argent Pediatr*. 2015;113:e199-e202.
- Corbetta JP, Durán V, Burek C, et al. High flow priapism: diagnosis and treatment in pediatric population. *Pediatr Surg Int*. 2011;27:1217-1221.