

Does prolonged semi-erection in prepubertal high flow priapism result in increased penile size?

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ABSTRACT

High flow priapism is a rare pathology resulting mainly from trauma to the perineum leading to arterial-lacunar fistula. Management includes arterial embolization using absorbable material, as well as conservative approach. In this case, the effect of prolonged semi-erection in prepubertal high flow priapism on increased penile size is discussed.

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Priapism is defined as a prolonged and persistent erection of the penis without sexual stimulation or desire. Two types of priapism have been described, the more common is the ischemic, low blood flow, veno-occlusive type in which inadequate venous out flow creates an acidotic hypoxic environment leading to a painful prolonged erection. Ischemic priapism is caused by corporeal smooth muscle relaxation precipitated by pharmacological or neurogenic factors. In addition, it could be caused by rare hematological conditions such as sickle cell anemia. The other type is less common and is a non-ischemic, a high flow priapism. It follows perineal or direct penile trauma resulting in an arterial-lacunar fistula from a lacerated cavernous artery, thus it bypasses the protective high resistance helicine arterioles, and results in uncontrolled high arterial inflow throughout the erectile tissue.¹

High flow priapism is characterized by constant, painless, incomplete erection after perineal or penile trauma.² The onset may be immediate but more often occurs after few days.

A case of prepubertal high flow priapism is presented with discussion of its management.

Case Report. A 13-year-old boy presented with 5 years history of incomplete and sustained painless erection not related to sexual excitement or abnormal behavior. The reason for presentation was mainly due to social embarrassment due to the continuous semi-erection. The semi-erect large penis was noticed also by his teacher at school. On detailed questioning there was a history of saddle type perineal injury 5 years earlier. Clinically the penis was enlarged, semi-rigid, with 14.5 cm length, 13 cm circumference at the base, and 15.5 cm mid shaft circumference. His workup investigations revealed that luteinizing hormone, follicle-stimulating hormone, prolactin, serum testosterone, and thyroid function tests, were all normal for his age. Pituitary and brain computed tomography (CT) scans were normal. Penile color Doppler U/S indicated increased flow in the left corpus cavernosum. Pelvic enhanced CT scan showed increased penile size with abnormally increased contrast enhancement along the left side of the penis suggesting arterial-venous fistula (**Figure 1**). We proceeded to carry out internal iliac angiogram, which revealed moderate

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Figure 1 - Enhanced computerized tomography scan of the pelvis, showing increased contrast enhancement in the left corpus cavernosum (arrow) and increased penile size.



Figure 2 - Selective arteriogram of the left pudendal artery, fistula present at the base of the left corpus cavernosum (arrow).

artery-cavernous fistula in the left corpus cavernosum (**Figure 2**). The patient reported increased penile size during stimulation, though during a pharmacological cavernosogram using 40 mg papaverine there was only slight increase in the penile size. It showed normal filling of corpus cavernosum on both sides, but there was an early proximal venous leak up to the internal iliac veins.

Discussion. Post-traumatic high flow priapism in prepubertal boys is a rare disease. Volkmer et al¹ reviewed the literature and described 33 cases including 3 reported by them. Traumatic arterial laceration resulting from trauma to the perineum or the penis was found to be the most common cause for high flow priapism in children, in addition rarely it could be due to inherited metabolic disorders such as Fabry's disease, or hematological disease, such as sickle cell anemia. Diagnostic evaluation includes history, physical examination, corporeal aspirate, color flow Doppler sonography, and angiography. The most important aspect is the history of recent perineal or penile trauma as well as the minimal or absent pain or tenderness in the turgid cavernosal bodies in the presence of soft glans penis. Hatzichristou et al,³ introduced the concept of the priesis sign and found it useful as part of the physical examination in children to differentiate arterial from corporeal veno-occlusive priapism. It includes perineal compression with the thumb, resulting in immediate detumescence, with relapse after withdrawal of the finger imposed force.

In adults with high flow priapism, this maneuver resulted in partial resolution only. In this patient perineal compression produced only slight detumescence. The corporeal aspirate of bright red arterial blood provides corroborating information.⁴ In our patient, the clinical presentation of continuous semi-erection preceded by blunt perineal trauma 5 years earlier, suggested the diagnosis. The

incomplete rigidity during high flow priapism suggests that there is an incomplete corporeal smooth muscle relaxation, most likely secondary to the absence of neurogenic mediated relaxation.⁴ The delay in seeking medical help may be explained by the absence of pain, and maintenance of erectile function. Several reports in the literature describe patients presenting with long standing history of several years.^{2,5}

The typical diagnosis and standard follow up in patients with arterial priapism are based on color Doppler ultrasonography as the method is non-invasive and highly sensitive and specific for detecting arterial-lacunar communication.² This was confirmed in this case by detecting increase flow in the left corpus cavernosum. Another non-invasive method using enhanced CT was reported once in the literature to be useful in the diagnosis.⁶ Again in this case, CT scan showed abnormally increased contrast enhancement along the left side of the penis suggesting arterial-venous fistula. The diagnosis of arterio-cavernous fistula is accomplished using selective internal pudendal artery angiography.⁷ Regarding treatment, the goal of active intervention is to restore normal physiological erections. Unlike low flow priapism, pharmacological treatments with intra-cavernous injections of alpha-adrenergic drugs,⁴ or methylene blue,⁸ have been used with temporary results. The rationale for use of Alpha-Agonist agents is the vaso-constrictive effect on smooth muscle, while the methylene blue prevents the formation of cyclic guanine monophosphate, an important second messenger in corporeal smooth muscle relaxation. However, due to the high flow state blood is not trapped in the penis and local administration of these agents may lead to systemic complications despite tourniquet application.⁴ Selective internal pudendal arteriography with transcatheter embolization of absorbable material such as

autologous blood clot, or gelatin sponge has been advocated as the main stay of active therapy, with long term follow up revealing excellent rate of resolution (100%) and restoration of erectile potency (86%).⁴ The advantage of the absorbable material is the transient occlusion of the cavernous artery, which lasts for 1-2 days. Permanent embolizing agents, such as coils should not be considered due to the potential for irreversible occlusion of the arterial inflow, causing irreversible changes in erectile function.²

Although embolization of the cavernous artery seems to be the treatment of choice, significant complications has been reported including a 66% rate of recurrence following initial embolization, formation of cavernous abscess, and permanent cavernous artery occlusion and persistent impotence.² In addition, complications of the angiography technique should not be underestimated. Reports of long-term preservation of erectile function of up to 31 years in patients with high flow priapism,² led to proposing a new approach of expectant management, starting with an observational period which may be helpful for patients who need time to estimate practically the impact of a constant semi-erection state on their social and personal lives, and at the same time physicians may avoid unnecessary intervention in cases of spontaneous resolution and/or unmask possible hemodynamic consequences associated with blunt perineal trauma. Hemodynamic consequences rarely coexists with other pathophysiological changes associated with blunt perineal trauma, such as the presence of arteriogenic impotence due to unilateral artery occlusion or, like in this case corporeal veno-occlusive dysfunction due to site specific fibrosis of the corpus cavernosum.² Such patients may risk permanent impotence from active priapism treatment and the persistence of the arterial-lacunar fistula may promote potency that could be enhanced by tension rings.²

Regular follow up examinations are suggested to asses erectile function changes at baseline and following sexual stimulation, as well as serial duplex Doppler ultrasound studies to measure the stability of the arterial-lacunar fistula.² Hatzichristou et al,¹ reported the complete resolution of priapism in 2 children during the observation period, one spontaneously within 10 days, and the other after placing a compressive dressing in the perineum for 24 hours, and suggested that only immediate intervention considered in children should be perineal compression. Observational treatment is not associated with ultra-structural changes to the erectile tissues due to the continuous

flow of completely oxygenated blood into the corpora;² however no mention has been reported in the literature regarding the effect of observational treatment on the size of the penis due to the continuous stretching of the tunica albuginea.

In this 13-year-old patient, and after 5 years of high flow priapism, penile measurements of the semi-erect penis were 14.5 cm length, 13 cm base circumference and 15.5 cm mid-shaft circumference. This is compared with fully erect penile mean length reported in normal 18-19 years olds of 14.48 cm.⁹ The mean erect base shaft circumference, in adult Caucasians reported by Da Ros et al,¹⁰ is 11.92 cm. Whether this increase in penile size especially the circumference measurements will have any deleterious effects on the penile function is to be seen. Due to the presence of concomitant venous leak, in addition to the family wishes, it was opted for conservative management, meanwhile, monitoring the progress of erectile function.

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