

Successful Use of an Inflatable Penile Prosthesis for the Treatment of Distal Deficiency of the Tunica Albuginea and Cavernous Tissue



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ABSTRACT

Introduction: Congenital hypoplasia of the distal half of the tunica albuginea has not been previously described.

Aim: To review a patient presenting with erectile dysfunction secondary to congenital penile hypoplasia.

Methods: History, physical exam and penile Doppler ultrasound of the patient, followed by a discussed of treatment options and definitive management.

Results: Successful operative treatment of our patient with insertion of an inflatable penile prosthesis.

Conclusion: We present a case of congenital hypoplasia of the distal tunica albuginea and a successful treatment strategy. We highlight the need for further study of penile embryology. **Punjani N, McGarry P, Brock G. Successful Use of an Inflatable Penile Prosthesis for the Treatment of Distal Deficiency of the Tunica Albuginea and Cavernous Tissue. Sex Med 2018;6:356–359.**

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Key Words: Penile Hypoplasia; Erectile Dysfunction; Penile Embryology

INTRODUCTION

Congenital hypoplasia of the distal half of the tunica albuginea has not been described previously. Sex differentiation occurs early in gestation, followed by an androgen effect on external genitalia production, whereby the genital tubercle elongates to form the glans penis and corpora cavernosa. Limited data exist on penile embryology and development. We present a case of absent corporal bodies in the distal half of the penis, with normal erectile tissue proximal to this with abnormalities noted on both physical examination and imaging. We also discuss treatment strategies. Informed consent was obtained.

CASE REPORT

Clinical Scenario

The patient was a healthy 17-year-old male who had been seen initially 1 year earlier for erectile dysfunction. His urologic history included simple distal hypospadias repair at age 2 years at a tertiary academic hospital by a pediatric urologist. Regarding sexual function, he described rigidity of only the proximal third

of his penis, with the distal two-thirds remaining flaccid and floppy even with stimulation, which precluded penetrative sexual activity.

Physical examination revealed normal external male genitalia, a circumcised phallus, and normal-appearing meatus. Palpation revealed attenuation of the distal penile shaft at the approximate midpoint, with a marked loss of palpable cavernous tissue.

Duplex ultrasonography (Figure 1) was completed with the patient supine following injection of a vasoactive injectable (5 μ g of prostaglandin E1) into the right corpus cavernosum. At the time of vasoactive injection, there was obvious proximal dilation and rigidity of the penile shaft; however, at the approximate midpoint, there was a sharp cutoff in penile rigidity, and the remaining penile shaft was floppy. Cavernous arterial flow was 40 cm/second on the right side and 35 cm/second on the left side. Some septal scarring was noted, but, more interestingly, a pervasive lack of cavernous smooth muscle content distally and apparent loss of tunica albuginea with large sinusoids were noted on resolution scanning. These results were in alignment with our physical examination findings.

Initially, treatment options were reviewed, including conservative management, experimental therapy (ie, stem cell regeneration), and insertion of an inflatable penile prosthesis (IPP), with possible additional reconstruction depending on intra-operative findings. However, IPP insertion was considered a treatment of last resort, to be avoided if possible, to reduce the risk of infection. Unfortunately, while the treatment options

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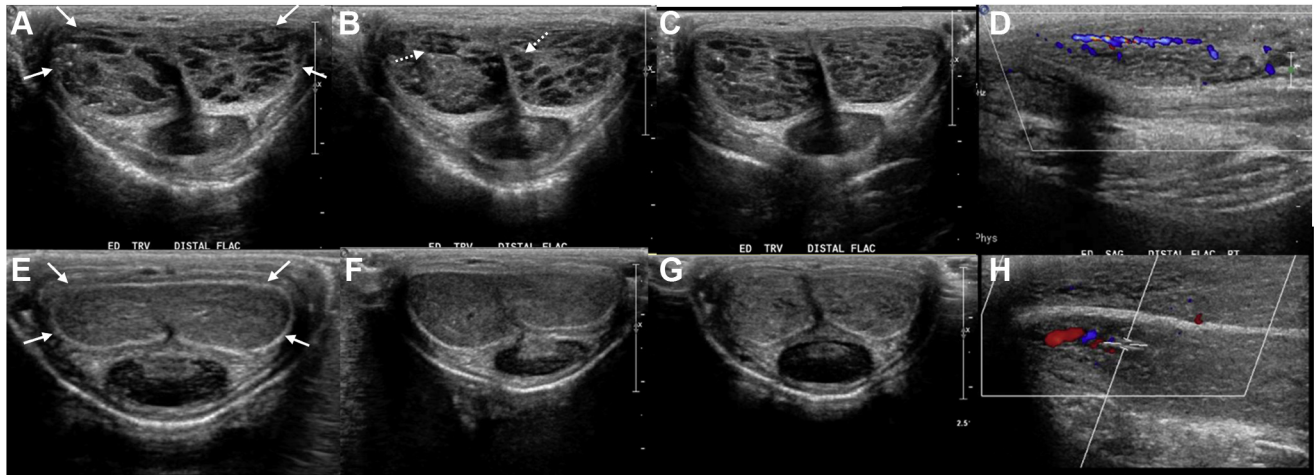


Figure 1. Duplex ultrasonographic images. Panels a to c demonstrate transverse imaging of the abnormal distal penis, and panel d illustrates sagittal imaging of the abnormal distal penis. Panels e to g demonstrate transverse imaging of the normal proximal penis, and panel h illustrates sagittal imaging of the normal proximal penis. The arrows indicate an absent tunical layer (a) in the distal abnormal penile shaft and a clear tunical layer (e) in the proximal penis. The dashed arrow in (b) further illustrate the large abnormal dilated sinusoids.

were being considered, the patient presented to the emergency department after a suicide attempt by a medication overdose, due to depression triggered by his sexual dysfunction.

The patient and family elected definitive management involving insertion of an IPP. Informed consent was obtained, and the risks of infection and device malfunction and the need for future surgeries were clearly conveyed. The device of choice was an AMS 3-piece inhibizone-coated IPP (Boston Scientific, Marlborough, MA, USA).

Operative Management

Using a penoscrotal approach, the urethra was identified with Foley catheter placement, and the bladder was drained. A transverse scrotal incision was made, and dissection was carried down to the tunica albuginea. Stay sutures were placed medially and laterally on each corporal body, and corporotomies were made. Dilamezins inserts measured the proximal corpora at 9 to 10 cm. Distally, however, resistance was met approximately halfway between the corporotomy and proximal glans, in the area thought to be devoid of cavernous smooth muscle. This area was very obviously blind ending and reflective of the anatomic location of the defect. The intraoperative decision was made to carefully perforate the distal corpora with Metzenbaum scissors. This created a neocorporal space at the level of the glans penis, and the decision was made to not use any further material to repair the defect, but instead to rely on the formation of a pseudocapsule around the device. We made this choice based on our experience with neophallus reconstructions, where we place the corporal cylinders into a skin-only neophallus sleeve and have not had any erosive issues. Total corporal length was 20 cm, and an 18-cm device with 2-cm rear-tip extenders was placed. Reservoir site dissection was challenging due to unexplained fibrosis but was successful

in the left retropubic space through the same incision. A 100-mL reservoir was placed and filled with 80 mL of injectable saline. The device was cycled, revealing a 30-degree ventral curvature. This was corrected to 15 degrees with remodeling (Figure 2). The valve pump was placed in a subdartos pouch. Closure of the dartos fascia was done in 3 layers, and the skin was closed with simple interrupted sutures. Copious irrigation with antibiotic-infused saline was provided at several points throughout the operation. A compressive scrotal dressing was placed, and the Foley catheter was removed.

Postoperative Course

On the first postoperative day, only minimal swelling and bruising was seen on dressing removal. Pain was minimal and voiding was satisfactory. The patient was discharged with the device partially inflated. At 1 week postoperatively, his pain was well controlled, and the device was deflated. The wound was clean, and there were no signs of infection.

Device cycling was taught at 1 month postoperatively. The device functioned well, and the patient was comfortable with both inflation and deflation. The patient was seen again several weeks later, at which point the device continued to cycle well, and he was able to perform this himself without difficulty. At this point, there seemed to be no further signs of mental illness, his mood had improved, and no further issues were reported by the patient or his family with regard to suicidal ideation.

DISCUSSION

Here we report the first case of absence of distal corporal tissue. The patient also presented with remote history of distal hypospadias, which raises suspicion of an embryologic or congenital phenomenon.



Figure 2. Postoperative insertion of penile prosthesis.

Sex differentiation begins between week 7 and week 17 of gestation, with the Y chromosome initiating differentiation through the SRY gene for testicular development.¹ Embryologic development of male external genitalia is then driven by an androgen-dependent component, an androgen-independent component, and endocrine and environmental influences.^{2,3} As an example, hypospadias is a common embryologic abnormality caused by a combination of hormonal and genetic factors. It is believed to occur from either an ectodermal or endodermal abnormality.³

More specific to male penile development, masculinization begins with a surge of luteinizing hormone in the presence of both testosterone and dihydrotestosterone.⁴ Following an increase in length of the perineum, masculinization involves phallic elongation, formation of the penile urethra, and prepuce development.⁵ Coronal sulcus separation occurs at around 12 weeks of gestation.⁴ From a cellular standpoint, the elongating phallus is composed of ectodermal tissue responsible for penile skin and mesodermal tissue forming the corporal tissue.⁶ The external penile skin was normal in our patient, indicating an abnormality of possible mesodermal origin. These mesodermal cells have been characterized as distinct dense mesenchymal condensations within the shaft of the developing penis and are thought to undergo epithelial–mesenchymal interactions for proper development.⁶ Given that our patient had hypospadias as well as absent corporal tissue, this may infer abnormalities in both

endodermal and mesodermal tissues during development. It also may suggest an alternative theory for corporal tissue development. Knowledge of this possibility is very limited, with no other clear reports in the literature.

We indicate in our case a treatment strategy and associated technical challenges. We illustrate a successful approach in the hands of a highly skilled surgeon using an IPP. The penile prosthesis was inserted in a usual fashion with a penoscrotal approach as described previously,⁷ because this is the method we are most comfortable with. Given the patient's young age, extra caution was taken to reduce the risk of infection and device herniation or injury. Evidence has shown that multilayer dartos closure may be associated with improved patient satisfaction and may help prevent future complications.^{7,8} Furthermore, the use of Metzenbaum scissors to aid in distal corporal dilation has been reported to be a suitable technique.⁹ Manual modeling was also used in this scenario, because the patient's curvature could not be assessed or predicted preoperatively, which was likely secondary to a lack of blood flow over time. Modeling was important to limit the need for further manipulation and reduce the risk of infection.¹⁰ Finally, after creation of the neocorporal space, the decision was made to not repair this area, because the abnormality was the absence of tunica and not simply a defect. We relied on the postoperative healing process and creation of a fibrotic pseudocapsule to provide further protection surrounding the IPP device.¹¹ We made this choice based on our experience with neophallus reconstruction where we place the corporal cylinders into a skin-only neophallus sleeve with no erosive issues. Fortunately, loss of elasticity in this area and its impact on penile length was not a concern for the patient. The device is functional in follow-up, with no palpable abnormalities or evidence of erosion of the device.

Other treatment options include experimental treatments, such as stem cell therapy, which is thought to relate to the potential ability of cells to differentiate into various cell types, including endothelial cells, smooth muscle cells, Schwann cells, and neurons.¹² Current clinical trials and studies are exploring these options for penile abnormalities. In our case, however, we were able to place the penile implant cylinders in a neocorporal location.

The implications of hypoplasia of the penis extend beyond the physical and functional to include a significant psychosocial impact. The patient in this report suffered a psychiatric event as a consequence. Studies have shown impacts on male mental health secondary to erectile dysfunction, with associated depression and anxiety.^{13,14} Our patient seemed to demonstrate improved mental status after the surgical treatment, highlighting the importance not only of treatment but also of setting realistic goals and expectations to prevent any future issues.

Our study is limited by the lack of actual tissue for pathological analysis to confirm the patient's abnormality. This

would have been unethical in this case, however, because we were able to place the IPP device safely through a single incision. Counterincisions or further manipulation to attain a biopsy of the distal corpora may have increased the risk of device infection.

CONCLUSION

Here we present the first reported case, to our knowledge, of hypoplasia of the distal aspect of the penile corporal tissue. Our report raises further questions and suggests the need for further study of the embryologic development of the corpora cavernosa in the development of male genitalia and whether there is a relationship with hypospadias.

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