J Ped Surg Case Reports 1 (2013) 94–95

Contents lists available at SciVerse ScienceDirect

Journal of Pediatric Surgery CASE REPORTS

journal homepage: www.jpscasereports.com

Epididymocutaneous fistula in a patient with neurogenic bladder

Danesh Bansal, Paul H. Noh*

Division of Pediatric Urology, Cincinnati Children's Hospital Medical Center, 3333 Burnet Avenue, ML 5037, Cincinnati, OH 45229, USA

ARTICLE INFO

Article history:
Received 27 March 2013
Accepted 27 March 2013

Key words:
Pediatrics
Epididymocutaneous fistula
Neurogenic bladder

ABSTRACT

We describe the development of an epididymocutaneous fistula in a patient with a complicated genitourinary history, including recurrent epididymitis and neurogenic bladder.

Retrograde flow of urine in the vas deferens has long been proposed as the cause of epididymitis and epididymo-orchitis [1]. This phenomenon may become evident clinically by the occurrence of urethrovasocutaneous and vasocutaneous fistulas [2]. The development of an epididymocutaneous fistula due to this reflux phenomenon is a rare occurrence. To our knowledge, only one other case of an epididymocutaneous fistula has been reported, in an immunocompromised patient with acquired immunodeficiency syndrome and Marfan's syndrome [3]. We report the first pediatric case of an epididymocutaneous fistula in a patient with neurogenic bladder. The patient's medical history necessitated special considerations in the diagnosis and management of the fistula.

1. Case report

A 4-year old male, with VACTERL syndrome, including an anorectal malformation, hypospadias, solitary left kidney, and neurogenic bladder was monitored with urodynamic studies that indicated severe detrusor instability. Management recommendations included intermittent catheterization, anticholinergic therapy, antibiotic prophylaxis, and bowel management. Noncompliance led to recurrent urinary tract infections, with culture results revealing pathogenic bacteria sensitive to prescribed antibiotic prophylaxis, and persistent urinary incontinence.

At 12 years of age, during a routine follow-up for neurogenic bladder, a nodule was found attached firmly to the inferior left hemiscrotum. The nodule had the consistency of a cyst without any pustule or erythema. Genitourinary exam was also notable for a right inguinal testis and left intrascrotal testis, with the nodule caudal to the testis. On return follow-up 6 months later, the nodule persisted with occasional drainage and was tender to palpation. Operative intervention was scheduled.

Left scrotal exploration revealed fibrosis around the epididymis. It became evident that the suspected scrotal cyst was an epididymocutaneous fistula. The fistula was amputated at the level of the epididymis. Bilateral orchidopexy was performed as well, via scrotal approach. A multi-layer closure of the left scrotal surgical site was utilized. No drains were used. There were no intraoperative complications or blood loss. The patient was discharged on the same day as surgery. The patient resumed normal activities upon discharge. No outpatient narcotic analgesia was used.

At a 1-month postoperative visit, incisions were healing well and both testes were noted to be in the scrotum without atrophy. Recommendations for management of neurogenic bladder were reinforced, due to continued suspicion of family non-compliance with treatment. Subsequent evaluations revealed more bacterial urinary tract infections during urodynamic studies. Increases in anticholinergic dosing were recommended, with no significant improvement of urodynamic findings. Upper urinary tract imaging has been stable without hydronephrosis on ultrasounds. At a 15-month follow-up after the procedure, urodynamic study was still consistent a non-compliant detrusor. Physical examination was notable for a recurrent left epididymocutaneous fistula. The family continues to be asked to comply with medical management.

2. Discussion

Epididymocutaneous fistulas are an uncommon occurrence. More commonly seen, but still rare, are urethrovasocutaneous and

* Corresponding author. Tel.: +1 513 636 4975; fax: +1 513 636 6753.
E-mail address: paul.noh@cchmc.org (P.H. Noh).

2213-5766
http://dx.doi.org/10.1016/j.epsc.2013.03.017

vasocutaneous fistulas following scrotal or testicular surgery [2]. Since the first case report in 1926, about 22 other cases have been reported. To the best of our knowledge we report the first pediatric case of an epididymocutaneous fistula occurring in a patient with neurogenic bladder. One case has been reported describing an epididymocutaneous fistula in an immunocompromised patient with acquired immunodeficiency syndrome and Marfan's syndrome [3].

The patient's medical history necessitated special considerations in the diagnosis and management of the fistula. Our patient's fistula formed due to non-compliance with the recommended medical management to optimize the patient's bladder function. Initially, non-compliance led to the development of recurrent bacterial urinary tract infections and epididymitis. Eventually, persistent non-compliance led to the development of an epididymocutaneous fistula.

Neurogenic bladder allows for urine to reflux into the vas deferens at high intravesical and intraurethral pressures [4], potentially causing vasitis to form if the problem is not corrected. Most often, before vasitis or other issues arise, the problem is corrected. However, uncontrolled neurogenic bladder can lead to increased pressure in the epididymis causing epididymitis. As seen in our patient, prolonged untreated neurogenic bladder can lead to pressure increases along the epididymis causing an epididymocutaneous fistula to form.

Appropriate management includes proper identification of the etiology: infectious, postsurgical, or non-compliance. Demonstration of the fistula may include a voiding cystourethrogram or a fistulogram [2]. Medical treatment can relieve the high intravesical and intraurethral pressure and prevent urine reflux into the epididymis [3]. However, surgical excision still remains part of the management in an effort the decrease the probability of reoccurrence [1]. Treatment goals include preventing epididymitis and preserving renal function. This patient's clinical course demonstrates the spectrum of disease and clinical sequelae of neurogenic bladder.

3. Conclusion

Neurogenic bladder can lead to an epididymocutaneous fistula. Appropriate treatment should be directed at management of the neurogenic bladder, including prevention of infections.

Conflict of interest/sources of funding

We have no direct or indirect commercial financial incentive associated with publishing the article. There are no sources of extra-institutional funding.

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