

case report

Symptomatic imperforate Cowper's syringocele in a 5-year-old boy

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Background. Cowper's syringocele is a rare anomaly in childhood. It is caused by the obstruction of the duct of Cowper's gland. Depending of the type and size of the syringocele, and the age of the patient, the treatment for symptomatic lesions could be endoscopic deroofting or open perineal surgery.

Case report. We report a case of symptomatic imperforate syringocele in a 5-year-old boy. Although the syringocele are usually best shown on voiding cysto-urethrography, there was not any detectable extrinsic impression or filling defect in the bulbar urethra. Ultrasonography guided perineal puncture with contrast filling of the cystic lesion was used to detect the connection of the Cowper's duct to the ventral surface of bulbar urethra.

Conclusions. In imperforate syringocele, ultrasonography could be useful imaging technique especially in young patients, to evaluate urethra and perineal lesions and for percutaneous guided procedures.

Key words: bulbourethral glands-abnormalities; child; preschool

Introduction

Cowper gland is an accessory sexual organ that contributes to semen coagulation and urethral lubrication.¹ The two main Cowper's glands are situated within the urogenital diaphragm, with a second pair of accessory glands situated in the bulbous spongiosal tis-

sue. The main Cowper's ducts enter the ventral surface of the bulbar urethra near the midline by piercing the spongiosum.

The accessory ducts can enter the urethra directly or drain into the main duct.

A retention cyst of a Cowper gland duct (syringocele) is caused by the obstruction of the duct of the gland; it causes a smooth rounded filling defect in the bulbous urethra just proximal to the expected site of the Cowper duct insertion. The true aetiology of Cowper's duct cysts remains uncertain; most Cowper's gland duct lesions are congenital; they are present in boys with reports of cystograms and have an incidence of 1.5%.^{2,3} Although usually asymptomatic, large reten-

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tion cysts may cause urethral obstruction, urinary infection and hematuria.^{4,5} The differential diagnosis of a Cowper's duct retention cyst includes urethral diverticulum, urethral duplication, fistula and Mullerian duct remnant.⁶ Depending of the type and size of the syringocele, and the age of the patient, the treatment for symptomatic lesions could be endoscopic deroofting or open perineal surgery.^{3,4}

Case report

A 5-year old boy presented with a one-month history of scrotal and perineal pain and discomfort, without dysuria or haematuria. On physical examination, fullness and medially protruding mass in the perineum were observed. The patient's urinary flow rate, blood and urinary profiles were normal. Scrotal and perineal ultrasonography revealed a well delineated thin-walled cystic lesion located in the lower scrotum and perineum; no clear communication with urethra could be detected (Figures 1a, 1b).

There was not any detectable extrinsic impression or filling defect in the bulbar urethra

during the voiding cysto-urethrography (Figure 2). Because of these findings, we decided to perform ultrasonography guided perineal puncture with contrast filling of the cystic lesion instead of cystourethroscopy; there was a small duct emerging from the proximal end of the cyst towards the bulbous urethra (Figure 3).

Due to the age of the patient and type of the syringocele, an open perineal surgery was carried out with complete resection of the syringocele. During the dissection of the cyst, there was a small duct emerging from the proximal end of the cyst towards the pelvic diaphragm, which was mostly resected after cannulation. During the follow-up period there was complete relief of perineal pain with normal urine profile and flow rate.

Discussion

According to urethrographic and endoscopic findings, the lesions of the Cowper's gland have been classified by Maizels *et al*⁷ into four groups: (1) simple syringocele - minimally dilated duct; (2) perforate syringocele - a



Figure 1a. Syringocele. Ultrasonography demonstrated well defined cystic lesion proximal to the urethra; longitudinal view.

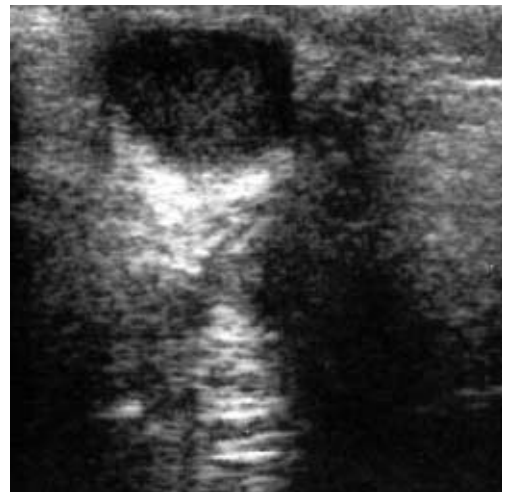


Figure 1b. Syringocele. Ultrasonography demonstrated well defined cystic lesion proximal to the urethra; transversal view.



Figure 2. Voiding cysto-urethrography. The normal urethra without filling defects or impression in the floor of the bulbous urethra.



Figure 3. US-guided puncture of the cystic lesion; contrast material injected into the cystic dilatation of the Cowper's gland with small duct emerging from the proximal end of the cyst towards the bulbous urethra.

bulbous duct that drain into the urethra via the patulous ostium and appears as a diverticulum; (3) imperforate syringocele - a bulbous duct that resembles a submucosal cyst and appears as a radiolucent mass; and (4) ruptured syringocele - the fragile membrane that remains in the urethra after dilated duct ruptures. Furthermore, syringoceles can be either within the corpus spongiosum (bulbar) or outside, lying posteriorly to it (perineal).⁸

The diagnosis of Cowper's syringocele is based on the voiding cysto-urethrography, cystourethroscopy and ultrasonography. Schultheiss *et al*⁸ reported MRI appearance of imperforate syringocele. Although the syringocele are usually best shown on voiding cysto-urethrography, if imperforate, there shouldn't be any filling defect in the bulbous urethra or reflux into the Cowper's gland ducts. In such cases, ultrasonography could be a useful imaging technique especially in young patients, to evaluate urethra and perineal lesions and for percutaneous guided procedures.⁹

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