Suprapubic Percutaneous Assisted Cystoscopic Excision of Posterior Urethral Fibroepithelial Polyps in Pediatric Patients

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Purpose: The aim of this study was to evaluate pediatric posterior urethral fibroepithelial polyps, their diagnosis and endoscopic treatments with suprapubic assisted transurethral polyp excision which is described by us.

Materials and Methods: We reviewed the charts of patients (n=6) who underwent suprapubic percutaneous assisted cystoscopic excision for posterior urethral fibroepithelial polyp from 2014 to 2019. Their data were retrospectively reviewed in terms of clinical features, diagnostic methods, endoscopic approaches, and postoperative results.

Results: The 6 patients, the mean age of 3 years (4 months-6 years), with a solitary polyp of posterior urethra diagnosed and removed by suprapubic percutaneous assisted cystoscopic excision in five years. The most common complaint was urinary tract infection (n:3). The urethral polyps were diagnosed by ultrasound and cystoscopy. There was no intraoperative or postoperative complication except for one patient with bleeding from the trocar site. All of the specimens after histopathology examinations showed fibroepithelial polyps and no recurrence was seen.

Conclusion: Posterior urethral polyps may cause obstructive effect in the urinary tract. The treatment should be performed with the least possible invasive method without injuring urethral wall. We believe that suprapubic percutaneous assisted cystoscopic resection, described by us is an easy, reliable and effective method for treatment procedure of posterior fibroepithelial urethral polyps.

Keywords: solitary urethral polyp; children; endoscopic resection; cystoscopy; voiding cystourethrography

INTRODUCTION

Isolated polyps can emerge and develop in any area of the urinary tract. The sources of their origination are mostly the ureter and renal pelvis, as well as the posterior urethra in rare cases[(1)]. The posterior urethral fibroepithelial polyp (PUFP) is known as a rare and usually benign lesion emerging from the verumontanum or posterior urethral mesodermal tissue[(1-4)]. Polyps usually provoke a variety of explicit symptoms such as recurrent urinary tract infection (UTI), urinary retention, and lower urinary tract symptoms among pediatric patients[(1)]. To diagnose PUFP, ultrasound (US), voiding cystourethrogram (VCUG), and cystourethroscopy are used[(3)]. The treatment approaches of PUFP include open cystostomy and transurethral resection or fulguration of the polyp by cystoscopy[(3)]. The current study involves the six pediatric patients who underwent PUFP excision with suprapubic percutaneous assisted transurethral excision (SPATE) endoscopically. In the present study, we aim to reflect on the experience of clinical appearance for PUFPs and treatment method by SPATE.

MATERIALS AND METHODS

This retrospective study is based on the evaluation of the data of patients diagnosed with PUFP who were treated in the pediatric surgery department of University of Health Sciences, Bursa Medical Faculty between January 2014 and June 2019. The required data for the analyses were obtained from the electronic database of the institution. The clinical characteristics, diagnostic methods, endoscopic approaches, surgical techniques, as well as postoperative results, were recorded for the cases with urethral polyp (**Table 1**). The data revealed that the endoscopically polyp excision by SPATE method was applied to all patients by the same surgeon over the previous five years. Written informed consent was obtained from each parent before the procedure. This method is visually illustrated in **Figure 1**. Since the cystoscopic approach is now a routinely practiced surgery for evaluation of various urinary diseases, we did not seek approval from the ethics committee because our method did not pose an additional risk for patients and is a modification of previous methods. All tissue samples were examined histopathologically.

Operative procedure

SPATE: A smooth, mobile, pedunculated polyp developing within posterior urethra was identified via cystoscopy (**Figure 2**). During a cystoscopic operation, a 2 mm laparoscopic grasper was placed into the patient's filled bladder transvesically through a sharped-edged suprapubic bladder catheter's trocar. Mannitol solution was used for intermittent or continuous irrigation to get a brighter cystoscopic view. A laparoscopic instrument was utilized to hold polyp as tightly as possible. Stabilization of polyp secured total resection with ensuring a safe distance to the base of the bladder mucosa. In

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Table 1: Patient's demographic data (AD: Accompanied disease, AUR: Acute urinary retention, BH: Bilateral hydronephrosis, UNC: Ureteroneocystostomy, UTI: Urinary tract infection, R: Right side, SPATE: Suprapubic percutaneous assisted transurethral excision, VCUG:Voiding cystouretrography, VUR: Vesicoureteral reflux).

Patient No	Age	Gender	Symptoms	Diagnose	Size	AD	Treatment
1	4 month	M	UTI	Cystoscopy	10x10mm	R Grade III VUR	SPATE
2	6 years	M	-	US	20x9 mm	-	SPATE
3	4 years	M	Intermittent micturition AUR	US	10x10 mm	R Grade III VUR	SPATE
4	5 years	F	UTI	Cystoscopy	15x10 mm	R Grade III VUR R Duplex System	SPATE UNC
5	10 month	M	UTI	VCUG	15x10 mm	BH	SPATE
6	2 years	M	Intermittent	Cystoscopy	12x10 mm	Bilateral Grade III VUR	SPATE

the meantime, a transurethral excision was performed easily by using the electrocautery resectoscope for the lesion's complete removal (**Figure 3**). The removed polyp was retrieved by transurethral basket catheter. Samples taken during the operation were examined histopathologically.

RESULTS

The pediatric population included five boys and one girl aged between four months and six years (the mean age of 3). Of these, three patients (50%) had recurrent UTI with fever, vomiting, hematuria, and abdominal pain. Two patients had voiding issue with acute urinary retention and intermittent micturition resulting from polyp obstacle. The last boy presented with extra-urinary symptoms and had an incidentally detected urethral polyp by US. In addition, patients had concomitant urinary diseases such as vesicoureteral reflux, urinary collecting system duplication, and hydronephrosis. The patients' characteristics and the diseases accompanied by PUFP were presented in **Table 1**. Diagnostic modalities for the PUFP were as followed: in two cases the polypoid mass image was clearly identified by US (**Figure 4**), in three cases the polyp was detected by cystoscopy during vesico ureteral reflux (VUR) treatment, and for the last one, the VCUG revealed a polyp as a filling defect in the posterior urethra (**Figure 5**). All polyps, solitary and sizes were ranging from 5×8 mm to 9×20 mm emerged in the posterior urethra, and all of the polyps were removed successfully by SPATE. The urethral catheters were removed within 36 hours, following the surgical intervention. The extra surgical intervention included sub ureteric injection for three patients with VUR and ureteroneocystostomy in one patient who also has right-sided duplicated collecting system with VUR. Further, the clinical course of the patient with bilateral hydronephrosis was improved remarkably after SPATE procedure without extra intervention. Except for bleeding that stops spontaneously in the trocar line, no intraoperative or postoperative complications were observed. The mean duration of hospitalization was 3,4 days. Histopathologic examinations revealed fibroepithelial polyps in all patients (**Figures 6-7**). The patients were continued to be monitored for a period of one year. They were followed up with the urinary US and urine analyses every month. Afterward, all patients were evaluated by VCUG at the first 6th months and control cystoscopies at the first year. There were good cosmetic outcomes for all and no complications or recurrence was observed throughout this follow up period.

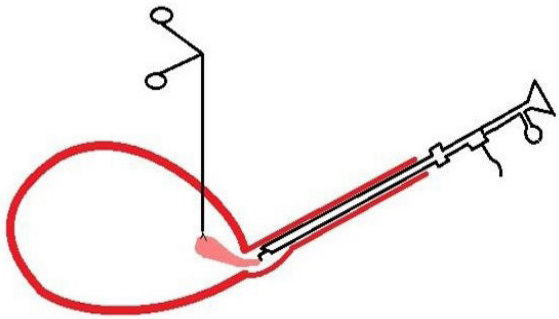

Figure 1. SPATE is illustrated

DISCUSSION

In childhood, PUFPs are rare lesions originating in the lower urinary tract, yet they are considered to be a benign fibrous tumour[3,4]. Regarding the current literature, the reported epidemiologic age of patients diagnosed with PUFP ranges from first months to 80 years, whereas 82% of all cases usually occur in patients younger than 20 years and manifest themselves in the first decade[5-7]. As claimed by Kimchhe and Downs, the average age of manifestation is between 8.25 and 9.7 years, yet our series of clinical studies represented the mean age of 3 years[6,7]. Additionally, boys are known to have a higher risk of developing PUFP than girls; for all that, the literature and clinical data related to female patients are quite limited[3,8-10]. Alike, in our series, male patients were the majority.

The etiology of PUFP still remains unclear, and it is suggested that the pathology has congenital origins. In the literature, experts on PUFP propose certain concepts on the etiology of PUFPS, including the developmental failure in the invagination process of submucous glandular material of the prostate gland's inner zone, an abnormal protrusion of the urethral wall or epithelial changes secondary to the maternal estrogen[7,11,12]. Besides, infectious, obstructive and traumatic causes have been theoretically considered as the physiological triggers of this disease[7,13].

From the clinical viewpoint, PUFPs are accompanied with explicit symptoms due to intermittent or acute obstruction of the bladder outlet, such as hesitancy, dimin-

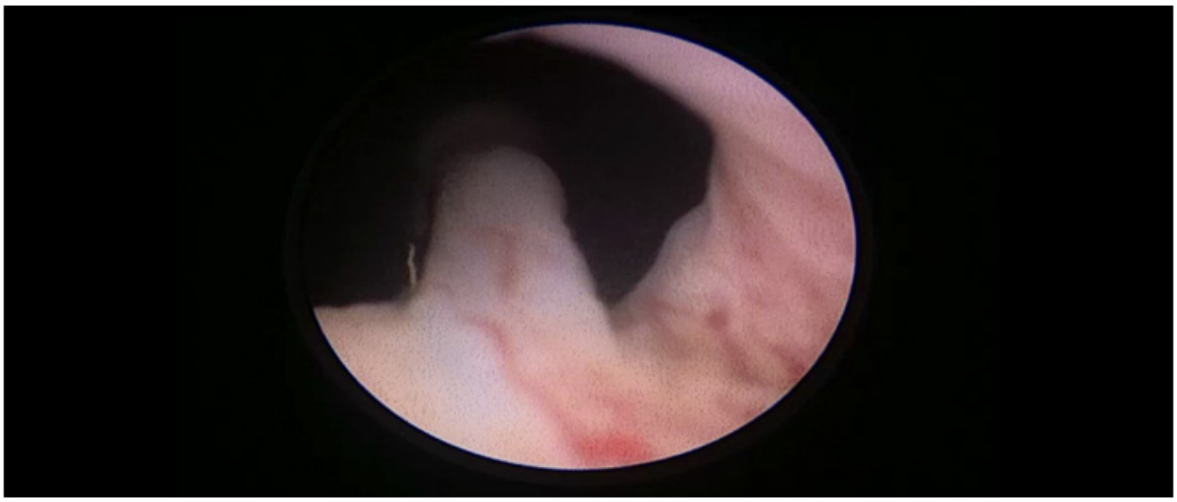

Figure 2. Intraoperative cystoscopic view

ished urinary stream, incomplete emptying, and urinary retention. Hematuria and dysuria are other common symptoms associated with urinary tract infections(1,8). The clinical triad, including intermittent urinary retention, hematuria, and lower urinary tract symptoms, indicates highly probable and noticeable signs of urethral polyps in children[1,8]. By reviewing the outcomes of 48 cases, Kearney et al. found obstruction (48%) to be the most common presenting symptom followed by hematuria (27%) and retention (25%)[14]. However, PUFPs could also be asymptomatic depending on sizes and location of polyp. De Castro et al. reported on a series of 17 cases where patients had been treated throughout a 16-year period. In fact, 14 out of 17 cases were symptomatic, and three were identified and diagnosed incidentally[8]. In our series, the mostly observed presentation was reccurrent UTI (n:3), urinary retention with intermittent voiding (n:2), and the last one was asymptomatic.

The diagnosis of PUFP is typically based on clinical findings, imaging modalities (US, VCUG) and cystoscopic evaluation[3,4,7]. While US tends to show polyps as an echogenic foci projecting into the lumen, VCUG reveals them as a polypoid filling defect, prolapsing through an external sphincter into the bulbous urethra. Intravenous urography is rarely applied because of radiation levels. Although US and VCUG are efficient diagnostic imaging, cystourethroscopy is utilised as a confirmatory strategy[3]. Cystoscopic evaluation is the most specific examination for identifying polyps. Additionally, because of the overlap in clinical presentation and radiological findings, diagnosis of polyp becomes a diagnostic challenge to distinguish a fibroepithelial polyp from a blood clot, radiolucent calculi or neoplasm. Cystoscopic evaluation; reveals the existence of polyps precisely and contributes to the excision of them simultaneously. In addition, to overcome the overlap, CT and urine cytology could be helpful for the diagnosis of calculi and malignancy[11,13]. Cystoscopy remains to be the most effective examination method, while the macroscopic appearance of the polyp being diagnosed in the majority of cases represents solid, mobile, smooth and pedunculated fibrous tumours that originate from the vicinity of the verumontanum and lie along the surface

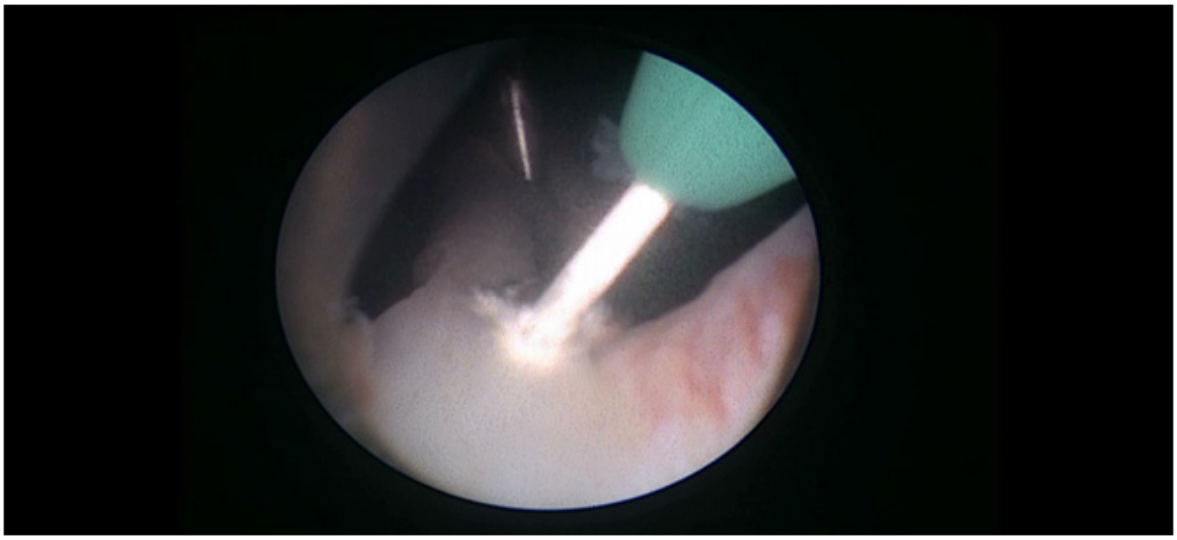

Figure 3. The cystoscopic view presented that the using of electrocautery resectoscope for the complete removal of the lesion.

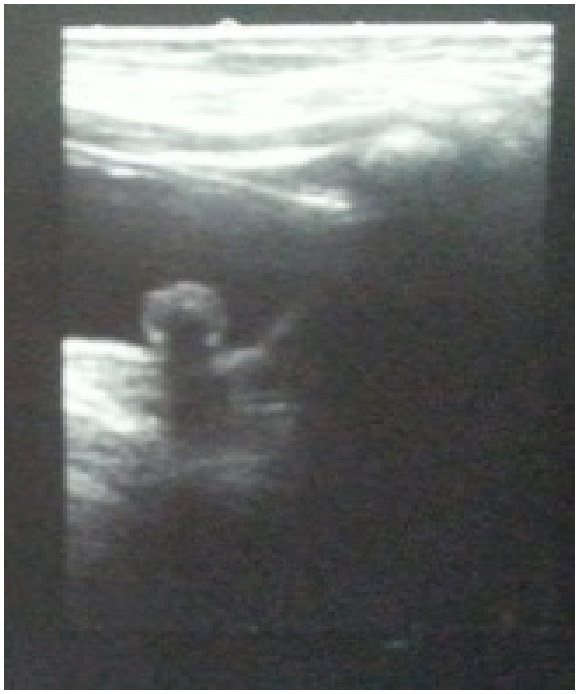

Figure 4. US examination demonstrates that an irregular mass, arises from the bladder neck and extended into the bladder.

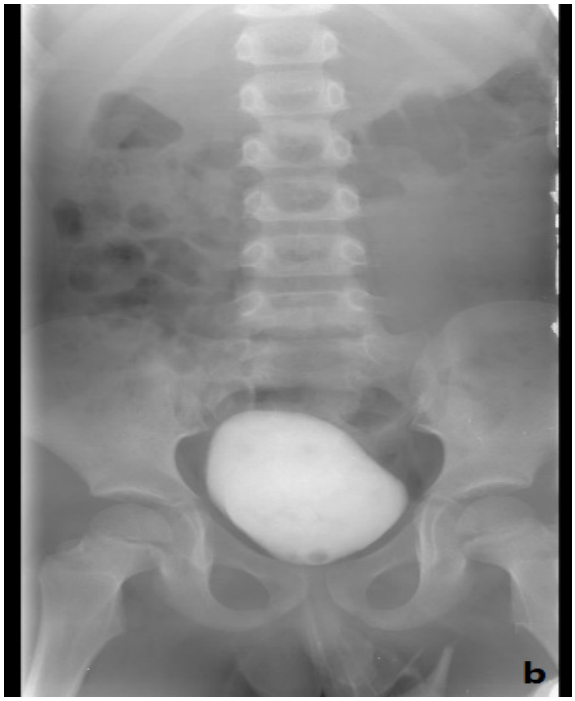

Figure 5. VCUG image demonstrate that polypoid filling defect on bladder neck.

of the posterior urethra[1,3]. In our series, all polyps were observed on the posterior urethral wall by cystoscopy and confirmed by the histopathological examination. With reference to the current literature, a polyp can be accessed and removed by transurethral excision or open cystostomy[3,15-17]. In 1985, Bruijnes et al. emphasized the importance of the general suprapubic approach through open cystostomy for the purpose of resecting polyps; in the meantime, it was mentioned that smaller lesions could be resected transurethrally[17]. In addition, Schafer reported on three cases with intravesical obstruction in 1989: two polyps were excised by open cystotomy, and the last one was treated transurethrally[18]. De Castro managed 17 cases of PUFP's endoscopically without complications or relapses[8]. Nowadays, open cystostomy is rarely required and can be applied mostly for removing large lesions. Common urethral polyps are usually removed transurethrally by the endoscopic resection electrocautery, cold knife or laser[1,2,6,7,11]. In the present series, patients were successfully treated by SPATE procedure with the electrocautery for PUFP's endoscopic resection. In addition, we relied on the transvesically suprapubic approach without conducting open surgery.

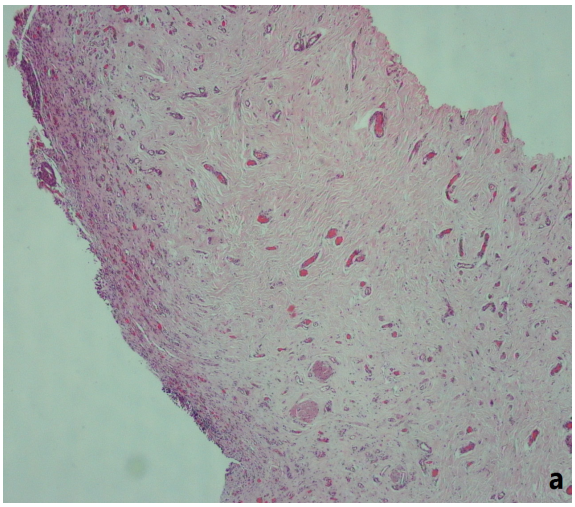

Figure 6. Histopathologic examination shows polypoid lesion with papillary fronds lined by transitional urothelium and associated submucosal fibrous stroma exhibiting mild chronic inflammation (Hematoxylin and Eosin x 40).

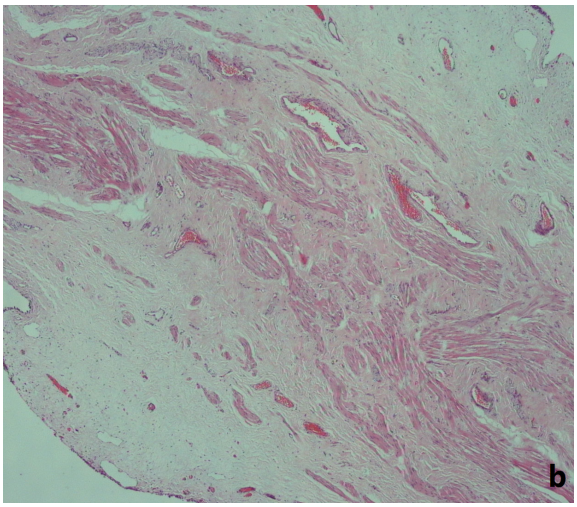

Figure 7. Histopathologic appearance with large magnification of polyp presents enriched stroma with various component, such as vascular formations with angiomatous features, smooth muscular fibers, pseudoglandular structures and inflammatory infiltrates (Hematoxylin and Eosin x 200).

It is important to note that to detect the original base of the polyp for the successful endoscopic treatment is significant. Although it is stated that the urethral approach can be easily performed in adults, it may not be able to reach the root of polyp cause of large peduncle that leads to the obstacle in childhood. Reaching the root of polyp might be complicated in some cases due to the obstacles that arise from the folding of the lesion's pedunculated body. Moreover, during cystoscopy, hypermobile pedunculated polyp might glide in and out of the bladder neck due to the endoscopic fluid flow. The use of a percutaneous dissector for the proper holding simplifies the identification of the polyp's root and helps to differentiate the urethral wall from the polyp itself during the resectoscopic excision. Thus, a full excision is secured under a direct vision without residual tissue. To the best of our knowledge, this is the first clinical series of children undergoing successful suprapubic assisted, transurethral polyp excision in terms of percutaneous treatment of PUFPs.

A diagnosis of urethral polyps must be distinguished from inflammatory and neoplastic lesions located within the urethra. This usually includes non-opaque stones, a foreign body, posterior urethral valves, Cowper's duct cyst, ectopic urethral insertion, urethral diverticulum and/or hypertrophy of the verumontanum[14]. These lesions provoke outlet obstructions of different degrees, and thus they require the differential diagnosis to identify a posterior urethral polyp[19]. It is crucial to keep in mind that without accurate tissue diagnosis, the invasive open surgery management may cause a negative impact on the clinical follow-up, such as subsequent fibrosis or urethral strictures, thus requiring and leading to a second surgical operation[14]. As the cystoscopy becomes the gold standard for diagnosis, treatment has to be performed with the least possible invasive method to avoid precedents for injuring the urethral wall. In terms of SPATE, the proper holding of polyp guarantees a stretched appearance on the pedicle that allows the resectoscope to commit a complete excision without damaging the surrounding tissue. The SPATE procedure ensures reliable outcomes from conducting an operation with direct observation of the process. In this sense, it is easier to perform as compared to other invasive strategies. In addition, this methodology requires no new or extra tools for SPATE, since all shaped-edged suprapubic bladder catheter's trocars are suitable and sufficient for use.

In the case of children, polyps emerging from the verumontanum are congenital in origins[1,3,5,7,11,12]. The verumontanum is viewed as a landmark for the exit of the mesonephric duct. Kuppusami and Moors conducted a histological analysis of the verumontanum to find that it incorporates smooth muscle and small glands; in all cases, it was lined by transitional cell epithelium[12]. Histopathologically, PUFPs are characterised by a fibrous core covered by transitional epithelium. The edematous stroma becomes the most variable element which is often filled with various components, such as vascular formations with angiomatous properties, smooth muscular fibres, nerves, glands, pseudoglandular structures, and inflammatory infiltrates[5,8,12,13]. Also, in our experience, the histopathological examination confirmed the clinical diagnosis of fibroepithelial urethral polyps in all cases.

There have not been reports on the malignant transformation of PUFP to date. Nevertheless, the possibility of recurrence because of the incompletely removed polypoid lesion has been mentioned[20]. Thus, we recommend that close follow-up, even if the risk of recurrence is low.

Several urethral polyps have been associated with other congenital urinary tract anomalies[7,14,19]. For example, De Castro reported that 50% of the patients with urethral polyps had a urinary pathology of different nature, especially VUR[8]. Kimche and Lask reported about 50 cases with a similar clinical picture; dilatation of the upper collecting system was identified in 10 patients (20%), while VUR was diagnosed in four (8%) patients[6]. Additionally, bladder diverticulum was present in four patients (8%). In the series of 18 patients reported by Akbarzadeh et al., six patients had VUR (33%), and five patients had dysfunctional voiding findings (27%)[19]. In our series, the associated diseases involved duplicated urinary system in one patient and VUR in four patients (66,6%).

Drawbacks in our study concerning SPATE include the limited number of cases, a low degree of control over cystoscopic evaluation and the shortage of long-term uroflowmetry results after the procedure. However, our initial observations presume that the SPATE procedure is easy to perform, which makes it also efficient for complete excision without residue.

CONCLUSIONS

PUFP is a rare pathology in children, whereas cystoscopy remains the gold standard for diagnosing the disease. Despite the status of a benign lesion, urethral polyps may cause obstructive effect in the urinary tract. PUFP resection by endoscopic methods should be considered a complete treatment in the majority of patients.

The current research review has revealed that transurethral resection is the best treatment choice, while SPATE is the most reliable strategy for ensuring complete excision.

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CONFLICT OF INTEREST

The authors declare that they have no competing interests.

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