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Management of lower urinary tract fibroepithelial polyps in children

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ABSTRACT

Introduction: Fibroepithelial polyps (FEP) of the lower urinary tract are relatively common in adults but rare in children, with fewer than 250 cases reported in the literature to date.
Objective: The aim of this study was to address the experience of FEP management in children.
Study design: A retrospective multicenter review was undertaken in children with defined FEP of the lower urinary tract managed between 2008 and 2018. The data at 18 pediatric surgery centers were collected. Their demographic, radiological, surgical, and pathological information were reviewed.
Results: A total of 33 children (26 boys; 7 girls) were treated for FEP of the lower urinary tract at 13 centers. The most common presentation was urinary outflow as hematuria (41%), acute urinary retention (25%), dysuria (19%), or urinary infections (28%). A prenatal diagnosis was made for three patients with hydronephrosis. Almost all of the children (94%) underwent ultrasound imaging of the urinary tract as the first diagnostic examination, 23 (70%) of them also either had an MRI (15%), cystourethrography (25%), computerized tomography (6%), or cystoscopy (45%). Two of these children (6%) had a biopsy prior to the surgery. The median preoperative delay was 7.52 (range: 1–48) months. Most of the patients were treated endoscopically, although four (12.1%) had open surgery and two (6.1%) had an additional incision for specimen extraction. The median hospital stay was 1.5 (range: 1–10) days. There were no recurrences and no complications after a median follow-up of 13 (range: 1–34) months.
Discussion: The main limitation of our study is the retrospective design, although it is the largest one for this pathology.
Conclusion: This series supports sonography as the most suitable diagnosis tool before endoscopy to confirm the diagnosis and to perform the resection for most FEP in children. This report confirms the recognized benign nature in the absence of recurrences.
Level of Evidence: Level V.

Urinary tract polyps occur rarely in children [1]. Since the 19th century, approximately 250 cases have been reported, mostly as case reports (Table 1) [1–11]. They are usually discovered in childhood or adolescence, although some authors have also described them in adults [12]. These fibroepithelial polyps (FEP) are congenital tumors of mesodermal origin and they most often occur in males [13]. They only rarely occur in females. They can affect the entire urinary tract, from the renal

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Table 1
Cases of fibroepithelial polyps reported in the past 25 years.

Series or case reports of FEP	Number of patients	Age at diagnosis	Gender	Symptoms	Diagnostic evaluation	Localization	Management	Follow-up
Current study	33	7.1	M (26) F (7)	AUR (8), UTI (9), hematuria (15), HN (3)	US, CT Scan, VCU, cystoscopy, MRI	bladder (14) urethra (19)	endoscopy (26) open surgery (7)	3–55 months
Ballard [2]	1	3	M	AUR	VCU, CT scan	verumontanum	endoscopy	
Akbarzadeh [3]	18	3.5	M (14) F (4)	AUR (7), UTI (6), dysuria (10), hematuria (14), HN(4), reflux (6)	VCU, cystoscopy	urethra and bladder	endoscopy (17) endoscopy + cystostomy (1)	3–17 years
Kaba [4]	1	14	M	hematuria	US, CT Scan, VCU, cystoscopy		open surgery	
Ala Natsheh [5]	2	3.5 [2–5]	M	hematuria (1), dysuria (1), AUR (1)			endoscopy (2)	1–5 years
Demircan [1]	2	1.75 [1.5–2]	M (1) F (1)	hematuria interlabial mass	VCU (2), US (2), cystoscopy (1)	urethra	endoscopy + cystostomy (1) direct surgery (1)	1 month
Isaac [6]	1	16	M	AUR	CT Scan, cystoscopy	urethra	endoscopy	
Beluffi [7]	1	0.08	M	hydronephrosis	US, VCU	verumontanum	endoscopy + cystostomy	
Barzilai [8]	1	0.8	M	AUR	US, VCU, cystoscopy	verumontanum	endoscopy	6 months
Rosenkilde [9]	1	2.5	M	AUR	VCU, cystoscopy, US	urethra	cystostomy	1.5 months
Gleason [10]	12	8.9 [1–14]	M (12)	hematuria (5), obstructive symptoms (4), AUR (2)	VCU (7), US	verumontanum (9), posterior urethra (3)	endoscopy (11), endoscopy + cystostomy (1)	12 months
De Castro [11]	17	<2 (6) 2–6 (5) >6 (6)	M	UTI (4) AUR (4) hematuria (7) dysuria (9)	US (3) VCU (17) Cystoscopy (3)	posterior urethra (17)	endoscopy (17)	12 months

Abbreviations; FEP: fibroepithelial tumor; AUR: acute urinary retention; UTI: urinary tract infection; HN: hydronephrosis; VCU: voiding cystourethrogram; US: ultrasonography.

pelvis to the urethra [14–16]. They are mostly located in the bladder [5] or the urethra. The posterior portion of the urethra is the predominant location [3,6,17–20], whereas anterior urethral polyps are only reported rarely [21–24]. They are usually described as a benign pedunculated polyp or bladder mass [4] at sonography (Fig. 1A). The main differential diagnosis is rhabdomyosarcoma, which is a heterogeneous mass with malignant characteristics. The pathology report typically confirms the presence of a fibroepithelial entity (Fig. 2) composed of vascular connective tissue [25].

The main features depend on the location of the FEP. As they have a stalk, these polyps are mobile in the bladder or the urethra. At the pathognomonic clinical level, they hence present as an intermittent or acute obstruction of the bladder. They can also cause bladder irritation that manifests as hematuria, dysuria, or urinary tract infections (UTIs). In case of an unusual presentation, the diagnosis can require supplementary preoperative imaging such as MRI (Fig. 1B) or endoscopic examination [2,8,26]. Surgical management is most commonly achieved endoscopically by transurethral resection. FEP of the lower urinary tract are benign lesions and no recurrences or malignant behavior have been reported to date. Although they are benign tumors, delayed diagnosis can lead to renal failure as a result of bladder obstruction [27]. Due to the rarity of this condition, no standard management and treatment have been published for this entity. The aim of our study was to report the current management of FEP in children.

1. Methods

A multicenter review was carried out to compile cases of FEP of the lower urinary tract in the past 10 years. Children operated on for FEP between 2008 and 2018 were considered for this study. This study was approved by the relevant ethics committee, with reference number 301–2019-67.

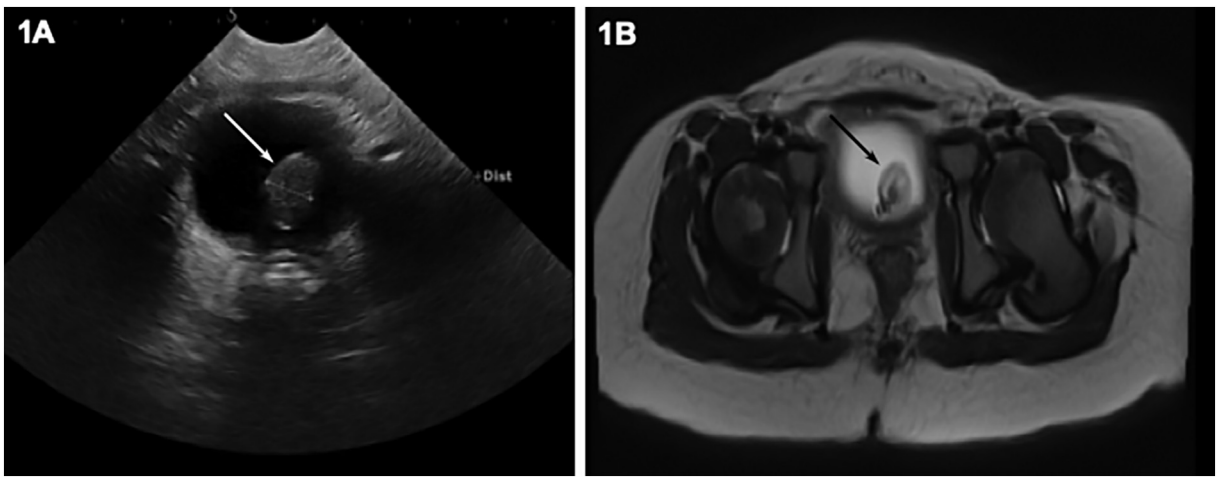

Fig. 1. Fibroepithelial polyp imaging. A: Ultrasonography typical presentation Image of a 20 × 12-mm-sized fibroepithelial polyp in a 21-month-old boy with intermittent bladder obstruction. B: MRI features of a fibroepithelial polyp in the bladder. T2 sequence showing a 16 × 12-mm-sized pedunculated lesion in a 21-month-old boy.

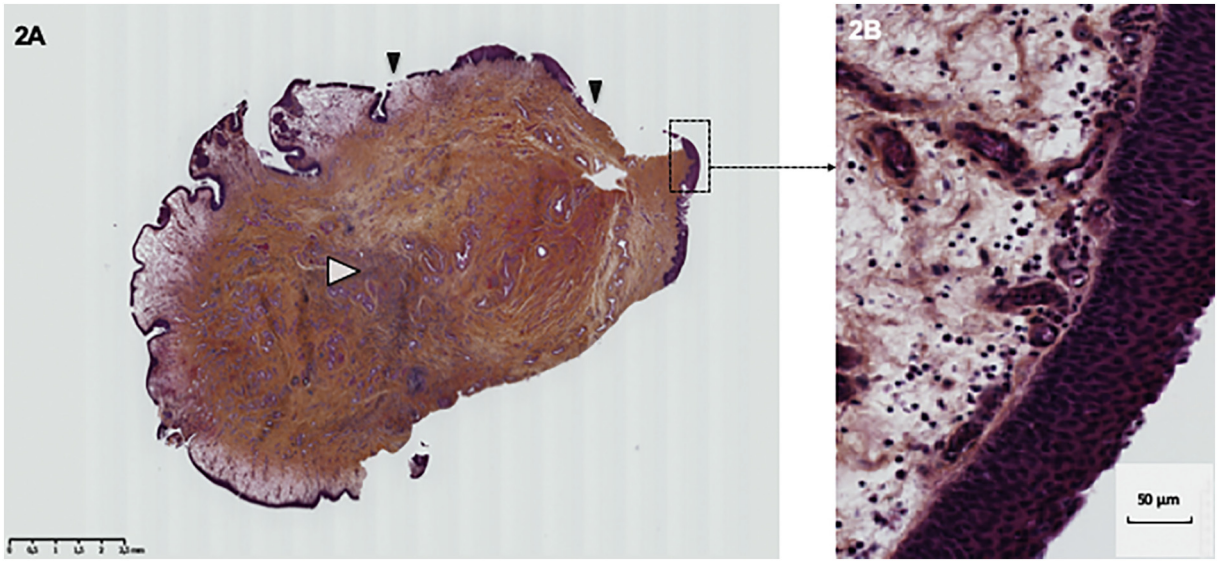

Fig. 2. Morphometry of a fibroepithelial polyp. A: Photograph of a 12-mm-sized fibroepithelial polyp (preparation with HES). The white arrow indicates the center of the lesion with fibrous connective tissue containing glands, smooth muscles, and nerves (10× magnification). The overlying epithelium is urothelium that contains areas of ulceration (black arrows). B: Simple hierarchical pattern of urothelium with a normal thickness and appearance (40× magnification).

A survey was sent to 34 centers in order to collect relevant clinical, radiological, and surgical data. These data included the age at presentation, the type of management (endo-surgery versus open surgery), prior medical history, associated anomalies, symptoms, the perioperative course, histopathology findings, and follow-up. Patients were included in case of FEP confirmed by histopathology and operated on between 2008 and 2018. Exclusion criteria were being over 18 years of age, a lack of pathology results, an upper urinary tract FEP, or an absence of follow-up. Descriptive statistics were performed using Fisher's exact test for the categorical variables, the Student's *t*-test for the parametric continuous data (means and the SD are presented), and the Mann–Whitney *U* test for the non-parametric continuous data (medians and the IQR were used). A p-value less than 0.05 was considered significant.

2. Results

Of the 34 centers, 18 centers replied to the survey. Three centers had unusable data and another one had not encountered cases of FEP. At the 14 remaining pediatric centers, a total of 36 medical files with FEP met the inclusion criteria. All of the patients were managed according to each center's protocols. Three of them were ultimately excluded due to a ureteral position of the FEP. The median age of the patients (26 boys and 7 girls) was 6.2 (range: 1 month–14 years) years of age, and none of them had a relevant prior medical history.

The clinical presentation (Table 2) was non-specific and most of the time comprised symptoms such as hematuria (39%), infections (27%), acute urinary retention (24%), dysuria (18%), hydronephrosis (9%), and pain (3%). For three infants, there was a prenatal diagnosis

Table 2
Characteristics of the patients.

Data	Total	Group 1 – bladder FEP	Group 2 – urethra FEP	Difference
Number	33	14	19	
Gender (M/F)	26/7	10/4	16/3	*p = 0.42*
Age at presentation (*years*)	7.11 (± 5.34)	7.4 (± 5.7)	6.9 (± 5.2)	*p = 0.82*
Symptoms (%)				
- AUR	- 8 (24%)	- 2 (14%)	- 6 (32%)	p = 0.42
- UTI	- 9 (27%)	- 1 (7%)	- 8 (42%)	p = 0.04
- Hematuria	- 14 (42%)	- 8 (57%)	- 6 (32%)	p = 0.17
- Pain	- 1 (3%)	- 1 (7%)	- 0	p = 0.40
- Dysuria	- 5 (15%)	- 2 (14%)	- 3 (16%)	p = 1
- Hydronephrosis	- 3 (9%)	- 2 (14%)	- 1 (5%)	p = 0.56
Diagnostic evaluation (%)				
- US	- 31 (94%)	- 14 (100%)	- 17 (89%)	*p = 0.49*
- MRI	- 5 (15%)	- 1 (8%)	- 4 (21%)	*p = 0.36*
- UC	- 8 (24%)	- 1 (8%)	- 7 (37%)	*p = 0.1*
- CT	- 2 (6%)	- 0	- 2 (10%)	*p = 0.5*
- Cystoscopy	- 15 (45%)	- 9 (64%)	- 7 (37%)	*p = 0.30*
Lesion size (*mm*)	11.6 (± 8.7)	8.6 (± 8.6)	13.9 (± 8.3)	*p = 0.13*
Surgery (%)				
- Laparotomy	- 4 (12%)	- 0	- 4 (21%)	p = 0.12
- Perineal approach	- 3 (9%)	- 0	- 3 (16%)	p = 0.24
- Endoscopic	- 26 (78%)	- 14 (100%)	- 12 (63%)	p = 0.01
• Trocar	• 2 (8%)	• 0	• 2 (12%)	• p = 0.5
• Cystostomy	•2 (8%)	• 0	• 2 (12%)	• p = 0.5
Urinary catheter (%)	15 (47%)	4 (28%)	11 (58%)	*p = 0.049*
Follow-up (*years*)	1.1	1.28	0.95	*p = 0.46*

Abbreviations: US: ultrasonography; UC: urethrocystography; CT: computed tomography; AUR: acute urinary retention; UTI: urinary tract infection.

according to the ultrasonography depiction of hydronephrosis during the third trimester of pregnancy.

Sonographic assessment was used extensively in the diagnostic process, followed by endoscopic evaluation and histological analysis. All but two of the children had ultrasonography as the first diagnostic examination. A total of 13 children (40%) had another complimentary exam, which was either an MRI (15%) (Fig. 1B), VCUG (Voiding Cystourethrography) (24%), or a CT scan (6%). Fifteen patients (45%) had a preoperative cystoscopy to sustain the diagnostic modality: five were performed extemporaneously during the same anesthesia to confirm the diagnosis by a brief consultation between two surgeons, and seven were performed systematically before laparotomy (12%) or before direct resection (9%) for FEP protruding through the external urethral meatus. The three remaining patients underwent two distinct cystoscopic procedures: during the first cystoscopy, a biopsy was performed for two of them (6%) because of an unusual endoscopic appearance, and a technical problem occurred in one case (3%). All of the other patients underwent direct surgical excision. The delay between the first symptoms and the surgical management of the polyp was between 1 week and 49 months, with a median of 7.52 months.

Endoscopic management with transurethral resection (79%) was performed for 26 patients. For nine cases (27%), Bugbee electrocautery was used to cut the polyp at its base, and polyp retrieval was performed transurethrally using forceps. A resectoscope was used in 17 cases (51%), and an additional trocar was necessary to stabilize large floating polyp in the bladder for two patients (6%). The specimen (median size 8.5 mm (range: 4–10.2)) was extracted transurethrally in 23 cases (70%) using forceps in 20 cases (60%) or a basket in three cases (18%). One polyp (3%) that was 13 mm in size was extracted by trocar incision, and two specimens (6%) were extracted by cystostomy (polyp sizes of 34 mm and 17 mm, respectively).

In case of large polyps of the bladder neck, an open approach was selected due to exposure difficulties at endoscopy. Seven (21%) patients were treated by open surgery: four (12%) boys by a Pfannenstiel incision after preoperative cystoscopy (the polyp sizes were 22 mm, 14 mm, 20 mm, and 10 mm) and three (9%) girls (Fig. 3) by direct perineal resection for FEP protruding through the external urethral meatus.

The polyp was located most frequently in the urethra (59%), which in 11 cases included a polyp of the urethral posterior wall (33%), and only one case of location at the urethral anterior wall was reported. The other main location was the bladder (41%). A statistically significant positive association between UTI and urethral localization was found ($p < 0.05$).

The median size of the polyps was 11.6 mm (range: 4.7–15). All of the specimens were histologically examined, which confirmed the diagnosis of fibroepithelial polyp (Fig. 2). Urethral polyps are cured statistically less frequently by exclusive endoscopic resection ($p = 0.02$).

Only 15 (45%) of the 33 patients had a postoperative urinary catheter, which was removed at a “median” time of 1.3 (range: 1–7) days postoperatively. In three cases (9%) involving patients who underwent an open approach, the catheter was a suprapubic catheter. No postoperative complications were reported. The mean duration of the hospital stay was 1.3 days ($\pm$2.1 days). Fifteen children (45%) were received treatment as outpatients.

There was no polyp recurrence after an average total follow-up of 13 months (1–34), and all of the patients became symptom-free. One child had reflux associated with the polyp, which was still noted during the follow-up. For all of the other patients, there was no reflux, no urinary retention, no hematuria, and no infection following the endoscopic resection.

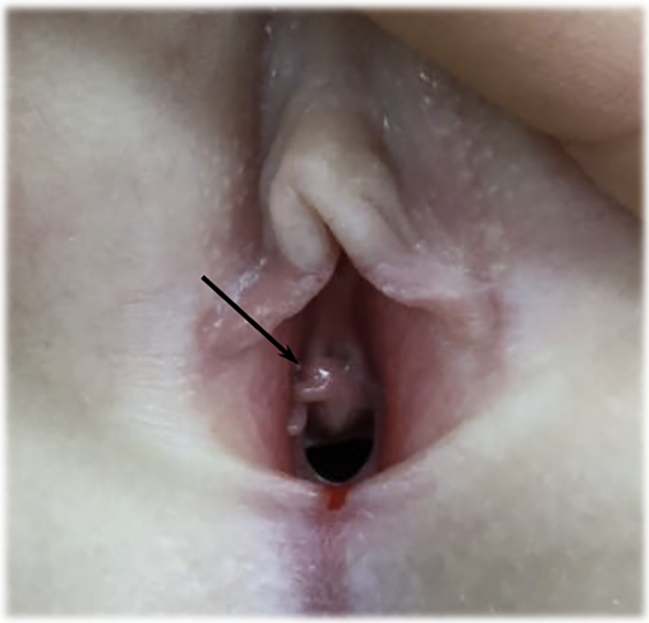

Fig. 3. Perineal aspect of a fibroepithelial polyp. Photograph of a 9-mm-sized fibroepithelial polyp protruding through the urethral meatus of a 14-month-old girl.

3. Discussion

Fibroepithelial polyps are a rare entity that can be encountered during childhood as a pedunculated lesion mostly in the urethral posterior wall (33% in this study). We here report the largest series of lower urinary tract FEP in children. The aim of polyp management is for the children to become symptom-free and to prevent any renal failure. Thus, it is important to identify these lesions and to reduce the preoperative delay. Given the rarity of this lesion, an algorithm for FEP management is proposed (Appendix 1).

The clinical triad of intermittent urinary retention, hematuria, and lower urinary tract symptoms has already been described by Akbarzadeh et al. in 2014 [3] as being clearly suggestive of urethral polyps in children. The clinical presentation of FEP depends on their location. Large posterior urethral polyps protrude and cause outlet obstruction, which can sometimes lead to acute symptoms. Bladder urethral stones have to be kept in mind as a differential diagnosis. In our series, this polyp location was revealed by acute urinary retention in 25% of cases and urethral localization of FEP was associated with larger-sized lesions and a higher incidence of UTI.

Ultrasonography is an excellent and non-invasive method to image and characterize bladder lesions. Urinary ultrasound can be considered to be the first-line and the only morphological examination, revealing a single, spherical, echoic, smooth lesion emanating from the bladder mucosa. A complex image with a grape-like appearance or cystic areas is suggestive of rhabdomyosarcoma. In case of suspected malignancy, contrast MRI provides higher resolution and it can reveal the origin and the local extension of the tumor [28]. In five cases (15%) in our series, an MRI was also performed, thereby confirming the ultrasonography results without providing supplementary information. In eight cases (24%), ultrasonography did not adequately reveal the polyp, and VCUG was hence performed. A diagnosis of a polyp lesion was made in light of a bladder defect. We, therefore, believe that ultrasonography is an adequate assessment tool when a diagnosis of FEP is likely. In case of doubt or no visible mass by ultrasonography, VCUG appears to be the second-line examination. It also has the advantage of excluding posterior urethral valves, which is the differential diagnosis in case of obstructive bladder symptoms in males.

Cystoscopy can be employed both for the diagnosis and for therapeutic purposes. We therefore recommend performing cystoscopy to confirm the diagnosis and the treatment at the same time. A typical radiological and endoscopic presentation allows FEP management with the administration of single anesthesia, as was the case for 30 patients (90%) in our study.

Prenatal diagnosis is extremely rare [7]. In three of our cases (9%), the hypothesis raised in light of hydronephrosis on prenatal ultrasonography. For two (6%) of them, no polyp could be discerned on the postnatal ultrasonography. VCUG was, therefore, performed to rule out vesicourethral reflux and it allowed for a successful diagnosis. Transurethral resection was performed in three of our cases (9%) of neonatal patients without encountering technical difficulties or postoperative complications.

Comparison with the adult population [12,14,29,30] indicates that the clinical presentation of FEP appears to be similar. The management, however, is not entirely the same. Indeed, when there is the possibility of a polyp in adult patients, cystoscopy is performed under local anesthesia to collect biopsies and to probe for the presence of a bladder tumor, without further imaging investigation. In our study, seven cases (21%) were found in girls, which is even rarer than in boys. Most of them exhibited a UTI or hematuria. Three polyps (9% of cases) were located on the urethra and were removed by urethral surgery under direct vision (Fig. 3), whereas the four other cases (12%) had a bladder location and were resected endoscopically.

The standard of care for the polyps is transurethral resection. Use of a resectoscope or forceps can achieve satisfactory fulguration of the base of the polyp. This series did not involve use of a laser fiber and there have been no publications of lower urinary tract location in children. Laser therapy is the treatment of choice for ureteral polyps and a number of polypectomies with Holmium have been reported in children [15]. In our opinion, it remains a good treatment option despite the limited resection depth.

In our series, urethral location is associated with less exclusive use of an endoscopic procedure, probably due to exposure difficulties and larger-sized lesions. When the polyp is too large or when it floats into the bladder, a bladder trocar is inserted for stabilization or exposure before endoscopic retrieval. Based on our series, the size of the incision for urethral retrieval appears to be 20 mm; above that size, a dedicated cystostomy appears to be required. Thus, in case of urethral lesion larger than 20 mm, a trocar or a cystostomy can be necessary to support the endoscopic procedure.

In cases of large FEP, fragmentation of the specimen was not considered in this series by the surgeons so as to favor the quality of the definitive pathology examination. This alternative can, however, be an option with an acceptable risk according to the long-term results in case of clear radiological and endoscopic FEP criteria. Such management must be decided at the beginning of the resection, before cutting the base. Indeed, endoscopic fragmentation of a floating lesion can be very difficult.

In our series, 45% of the patients had a postoperative urinary catheter, which was removed after a median of 1.3 days without hematuria. If the surgical procedure is accomplished without any complications, the procedure can be performed as an outpatient (as it was the case for 15 of our patients). No recurrences were reported after a follow-up of more than 12 months, which confirms the data in the literature: recurrence can appear if the stalk of the polyp is not completely excised [31].

The limitations of our study are that it was a retrospective study. Moreover, although if it is the largest study to date for this pathology, only a limited number of patients were included, thereby resulting in a lack of statistical power. However, it allowed the management of this rare disease to be refined.

4. Conclusion

This series supports the notion that the use of sonography is the most suitable diagnosis tool before endoscopic assessment and resection of FEP in children. In case of an unusual presentation, VCUG is the most informative morphological examination. In case of FEP larger than 20 mm, mini-invasive treatment may also require a bladder trocar for exposure, and sometimes a bladder incision for specimen retrieval. This report also confirms the widely recognized benign nature of FEP with the absence of recurrences.

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jpedsurg.2020.05.030.

Declaration of competing interests

The authors declare that they have no conflicts of interest.

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