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ORIGINAL ARTICLE

Preliminary report on endoscopic pilonidal sinus treatment in children: results of a multicentric series

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Abstract

Background Pilonidal disease is a troublesome acquired condition for whom various surgical treatments have been proposed with relatively high recurrence and complication rates. Since EPSiT technique has been described in 2013, it became an alternative treatment in adult practice. Our study reports the results of a multicentre series of pediatric patients who underwent EPSiT procedure over a 21-month period.

Methods Between July 2015 and March 2017, all consecutive patients undergoing EPSiT in four different pediatric surgical units have been enrolled. Details regarding demographic data, detailed surgical procedure, outcome and complications have been recorded.

Results A total of 43 patients underwent EPSiT procedure. Mean age was 15 years. There was a slight female preponderance. Mean weight and height at surgery were 67 kg and 168 cm, respectively. In 14% of cases a previous ineffective procedure was performed. Mean length of procedure was 34 min and median hospital stay was 24 h (12–72 h). Median length of follow-up was 4 months (range 3–18 months). Complications leading to reoperation were reported in 9% of cases with an overall resolution rate of 88%.

Discussion EPSiT proved to be feasible and safe even in the pediatric population. The effectiveness and safety of the procedure suggest that this technique can represent a valid alternative for the treatment of pilonidal disease in children.

Keywords Abscess · Children · EPSiT · Pilonidal disease · VAAFT

Introduction

Pilonidal disease is an acquired condition characterized by the presence of median or paramedian openings at the intergluteal groove deriving from ingrowing of hair into the deep sacrococcygeal subcutaneous tissue [1]. Sondenaa et al. reported in 1995 the most salient clinical features of patients. The calculated incidence of the disease proved to be of 26 into 100,000 with a strong male preponderance. Reported male-to-female ratio was 2.2:1 [2]. The onset in adolescence seems to be due to the increased sebum and hirsutism compared to younger children [1]. The management of this annoying issue, which can have an onset up 2 years before medial referral, is debated and many different approaches have been proposed so far [1]. Some authors suggested to treat the disease by epilation only, others suggest to simply go for sinus incision and drainage, others suggest marsupialization, radical excision with open wound or direct closures. Recently full-thickness flap closures have been suggested to improve results. Regardless of the adopted treatment, the recurrence rate of pilonidal disease proved to remain stable and ranges between 20 and 30% of cases [3–10]. Recently, Meinero et al. published a new minimally invasive technique for the treatment of pilonidal disease in adults [11]. The technique was named endoscopic pilonidal sinus treatment

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(EPSiT) and was demonstrated to have a surprisingly low recurrence rate, lower than 10% [11, 12]. This multicentric study is aimed at reporting the results of EPSiT in patients younger than 18 years of age and at providing details regarding this series of patients treated over a 21-month period.

Patients and methods

Study period was between 1st of July 2015 and 30th of March 2017 (21 month period).

All consecutive patients admitted during the study period for the treatment of pilonidal disease by the participating pediatric surgery units in Genoa, Alessandria, Naples and Legnano were enrolled. A specific informed consent, describing available alternatives, details of the technique and expected complications, was signed by the parents.

Inclusion criteria

1. Patients younger than 18 years of age (17 years, 11 months, 30 days),
2. Pilonidal disease with chronic discharge, inflammation or abscess,
3. Surgeon trained in EPSiT technique.

Exclusion criteria

1. Patients older than 18 years of age,
2. Follow-up shorter than 3 months (based on the reported incidence of delayed healing occurring within the first 2 months postoperatively [11], patients with a shorted follow-up were excluded to avoid miscategorising),
3. Immunocompromized patients with a high likelihood of persistence or recurrence,
4. Refusal of parents to sign informed consent.

All authors in the four participating units prospectively recorded demographic data, diagnostic details, presence of co-morbidities, preoperative imaging, previous surgical treatments, weight and height at surgery, detailed surgical information, duration of EPSiT procedure, type of anaesthesia, postoperative pain assessed 6 h postoperatively with visual numeric pain distress analogue scale (VAS), length of hospitalization, type and duration of antibiotic treatment, length of follow-up and complications observed in the medium to long term. Data were stored in a digital database according to Data Protection Act. Of note, most of the patients belonging to the series from Naples have been already published and described under the term PEPSiT procedure, which merely represents the pediatric application of the EPSiT technique [16].

Technique

All pediatric surgeons performing EPSiT (APP, CM, CE, and AD) had attended a coloproctology course to train on the technique and had been supervised by expert surgeons during their learning curve. In all centers EPSiT was performed according to what reported by Meinero and coworkers [11, 12]. The technique consists of:

1. Patients are prepped using either povidone solutions or chlorexidine.
2. A 2% solution of either saline or mannitol + glycine is used to perform fistuloscopy and to irrigate the pilonidal sinus.
3. Either a 3.3 mm standard co-axial pediatric Cystoscope or a 10 Ch Fistuloscope (Karl Stortz, 78532 Tuttlingen, Germany) are used to perform the procedure.
4. High-pressure irrigation of the fistula and/or of the pilonidal cavity is performed to achieve enlargement of their internal lumen.
5. Fistuloscopy and direct visualization of the internal lining of the pilonidal cavity permit the identification of all side openings and tracts and the removal of all hair and follicles.
6. High power electrocoagulation with monopolar diathermy under direct endoscopic view of the inner aspect of the fistula and of all cavitation and side tracts.
7. Removal of debris and of all residual hair and follicles under direct visualization.
8. Electrocoagulation of bleeding vessels and complete removal of the external opening of the fistula tracts.

Follow-up and definitions

Patients were followed up on a regular basis in an outpatient pediatric surgical setting to check for the correct dressing and wound irrigation, as previously reported by Meinero in his original description [11]. All patients underwent a single long-term clinical re-evaluation by a qualified pediatric surgeon that was undertaken after a median of 4 months (3–16 months) to collect results for this study. Outcome measures were recorded according to the following definitions:

- *Recurrence* was defined as "persistence of discharge after more than 3 months with either abscess formation, infection or local pain and discomfort".
- *Delayed healing* was defined as "wound closure occurring after more that 6 weeks postoperatively": this issue was mostly due to granuloma formation requiring topic silver nitrate administration.

- *Infection* was defined as "persistent smelly discharge with pain and redness possibly associated to systemic signs (i.e., fever) requiring antibiotic administration".
- *Success* was defined as "complete wound closure, no discharge, no pain in the area of surgery both spontaneously or during palpation, no signs of infection/inflammation".

Statistical analysis

Descriptive statistics were reported as absolute frequencies and percentages for qualitative data. Median with range was used for categorical variables. Mean and standard deviation was used for continuous variables. Chi square test, Fisher Exact and *t* test were used to compare outcome measures in different subgroups of patients based on possible risk factors, namely BMI, previous recurrences and number of fistulas. A *p* lower than 0.05 was considered as statistically significant.

Results

During the study period, 43 patients aged less than 18 years were included. Mean age at surgery was 15 ± 1.4 years. Male-to-female ratio was 0.87:1. Two patients had co-morbidities: brainstem tumor and congenital heart malformation. None reported clotting abnormalities or immunodeficiency. Mean weight at surgery was 67 ± 12.5 kg, mean height was 168 ± 10.6 cm. Mean BMI was 23 ± 3.5. Six out of 43 patients previously underwent 11 ineffective sinus removal with a "lay open" healing technique to deal with pilonidal disease. One of these patients experienced 6 recurrences. Preoperative imaging was performed in 14 patients (32%) before referral for EPSiT. Imaging included eight ultrasound scan (USS) and six magnetic resonance imaging (MRI) that provided confirmation of the abscesses and fistula without adding any further information. Pilonidal sinus presented with chronic discharge in 38 (88%) and abscess with or without fistulisation in 5 (12%). Up to 3 fistula were recorded preoperatively. Fistula were detected in the midline in 33 patients (77%) and laterally in the remaining 10.

Anaesthesia was general with endotracheal intubation in 11 patients, spinal in 17, local in 12 and blended in 3. Mean length of EPSiT procedure was 34 ± 7.2 min. In case of a previous ineffective procedure length of EPSiT was 35 ± 3.7 min ($p = 0.7414$). Antibiotic prophylaxis was administered in 29 patients (cefazoline 40 mg/kg up to a maximum of 2 g). Fourteen patients did not receive any antibiotic prophylaxis. Mean VAS score was 2 ± 1.6 six hours postoperatively. Local anaesthesia provided the worst pain management with a mean VAS score of 3.2 ± 1.1, which proved to be significantly higher when compared to general and spinal anaesthesia that, respectively, scored 0.2 ± 0.4 and 2.06 ± 0.6 ($p < 0.0001$ and $p = 0.0004$, respectively) (Table 1). Median length of hospital stay was 24 h (range 12–72 h).

Median length of follow-up was 4 months (range 3–16 months). Complete healing occurred in 38 patients (88%) after a median of 3 weeks (range 2–6 weeks). Overall, six patients (16%) experienced a total of nine complications. Five patients experienced recurrences (12%), two patients reported granuloma in the site of endoscopy that was treated with silver nitrate applications (5%), and two patients reported delayed healing (5%). One patient (2%) experienced surgical site infection. Four patients required reoperation (9%) and two of them demonstrated persistence of subcentrimetic subcutaneous cavitation at surgery but no true recurrence at all.

Comparing the incidence of complications based on different risk factors (antibiotic prophylaxis, number and type of fistulas, BMI) none proved to significantly interfere with the outcome. Of note, BMI in patients who experienced complications was higher (24.97 ± 6.33 versus 23.31 ± 2.84) but the difference proved not to be be statistically significant ($p = 0.2955$, see Table 1 for details).

Discussion

In the last decades various published papers and reports regarding treatment of pilonidal disease in children addressed many available options, including primary midline repair [8, 9] open healing [1, 7], lateralized full-thickness flaps [13, 14], fibrin sealant [5], phenol application [15] and even hair epilation alone [4].

Endoscopic pilonidal sinus treatment (EPSiT) has been described in 2013 by Meinero et al. [11] and quickly gained popularity in adult practice [12]. The first reported series in pediatrics is that by Esposito et al. (Authors CE and ME in this study) who reported a series of 15 patients who underwent EPSiT procedure for noninfected pilonidal sinuses [16]. The authors demonstrated feasibility and safety of the technique and excluded complications or recurrences [16]. Our multicentre study included most of the patients belonging to the paper published by Esposito et al. (15 out of 17 patients) [16] and basically confirmed their results in a larger series and with higher consistency. Given the size of our series, we could provide more realistic outcome data and complication rates that proved to overlap those already reported in adults clinical practice [11, 12].

Although this aspect did not represent a focus of our study, we could observe that preoperative imaging for classic pilonidal disease is unnecessary. In fact, both USS and MRI proved to be of no use (both diagnosis and treatmentwise) in our series of patients.

Table 1 Details regarding our series of patients who underwent EPSiT

Demographics	*N* (%)			
Total patients	43 (100)			
Males	20 (53)			
Females	23 (47)			
M:F ratio	0.87:1			
Patients' features	Mean ± SD			
Age at surgery (years)	15 ± 1.4			
Weight at surgery (Kg)	67 ± 12.5			
Height at surgery (cm)	168 ± 10.6			
BMI at surgery	23 ± 3.5			
Preoperative history	*N* (%)			
Preoperative MRI	6 (14)			
Preoperative USS	8 (19)			
Previous surgery	6 (14)			
Clinical features	Median (range)	Complications	*p*	
Number of fistulas	1 (1–3)			
More than 1 fistula	17 (39%)	2/17	*1.0000*	
Fistula tract	*N* (%)			
Midline fistulas	33 (77)			
Lateral fistulas	10 (23)			
Clinical details	Mean ± SD	Issue	*p*	*p*
Antibiotics	*N* (%)	Complications		
Antibiotic prophylaxis	29 (67)	4/29	*1.0000*	
Anaesthesiologic plan	*N* (%)	Pain (VAS)		
General anaesthesia	11 (26)	0.2 ± 0.4	*0.0001*	*0.0001*
Spinal anaesthesia	17 (39)	2.06 ± 0.6	*0.0004*	
Local anaesthesia	15 (35)	3.2 ± 1.1		
Length of surgery, hospital stay and F-Up				
Length of surgery (min)	34 ± 7.2			
Hospital stay (h)	24 (12–72)			
Length of follow-up (months)	4 (3–16)			
Results	*N* (%)	BMI		
Complete healing	38 (88)	23.31 ± 2.85	*0.2801*	
Overall patients with complications	6 (14)	24.97 ± 6.33		
Recurrence	5 (12)			
Granuloma	2 (5)			
Delayed healing	2 (5)			
Reoperation	4 (9)			

Both general and spinal anaesthesia yielded better pain control compared to local anaesthesia alone. BMI effect was not statistically significant but showed an interesting trend ($p = 0.2801$)

M:F male to female, *BMI* body mass index, *SD* standard deviation, *MRI* magnetic resonance imaging, *USS* ultrasound scan

When coming to the most salient aspects of the study, we could demonstrate that EPSiT procedure is feasible both under general and regional or local anaesthesia in children as it is in adults. EPSiT is not painless but postoperative pain control proved to be satisfactory with the use of routine pain-relief medications. In fact, postoperative pain remained well within acceptable ranges (mean VAS was 2 ± 1.4). Although local anaesthesia provided the worst pain control, mean postoperative VAS score was reasonable and confirmed the applicability of all concepts of fast track care with quick return to normal daily activities. In fact, EPSiT has been performed either as an outpatient or as a day case procedure with a hospital stay that was short as 12 h.

Complete wound healing proved to occur earlier in case of EPSiT when compared to other available procedures [11, 12, 16]. Complete healing in EPSiT was reported after an average of 1 month postoperatively and this proved to be shorter compared to what reported for both open and closed

repair thus confirming the effectiveness of the procedure [1, 3, 6–10, 13]. Furthermore, EPSiT showed a relatively low incidence of complications, being recurrences, infections and reoperation rates basically halved (overall 14% versus over 30% for other techniques) [1, 4, 5, 7–9, 13, 15].

In particular, infections following EPSiT proved to be extremely rare. Of note, a meta-analysis by McCallum et al. in 2008, comparing open versus closed repair of pilonidal sinuses, reported an infection rate of up to 22% [13]. Although the authors themselves underlined that most of papers reported relatively low infection rates, the results of EPSiT in our series showed infections to be nearly absent. Most of the patients in our series were treated with antibiotic prophylaxis (30 out of 43) and its use did not seem to interfere with the incidence of complications ($p = 0.3023$). Although, the only patient who developed infection did not receive antibiotic prophylaxis perioperatively, the relatively small series does not allow to draw definitive conclusions on this regard. On the ground of these considerations we cannot provide evidence-based recommendations but suggest to administer a short term prophylaxis in a single-shot administration of Cefazoline 40 mg/kg (max dose 2 g).

Recurrence and reoperation rates have been reported to be around 25% in previous paediatric published series, regardless of the adopted procedure [1, 4, 5, 7–9, 13, 15].

EPSiT proved to be more effective as we observed a halved incidence of recurrence. As a consequence, reoperations were required by only less than 10% of our patients.

Focusing on possible risk factors for complications, nothing turned out to be statistically significant. Although complications occurred in patients with the highest BMI, even this issue turned out not to be significantly correlated. Nonetheless, we cannot exclude that larger series could disclose the potential role of BMI as a risk factor predictive for complications in children, as previously reported [10] (Table 1).

EPSiT proved to be feasible and safe even for redo. In fact, the similar results of EPSiT (length of procedure and results) in redo compared to overall series confirmed its versatility and its possible role even as a second-line treatment for patients whose first therapeutic choice was different.

Recently, some authors advocated the use of hair epilation alone for the treatment of pilonidal disease [4]. Nonetheless, results resemble those of other available options as subcutaneous retained hair or follicles are not removed, thus facilitating recurrences and infections. Anyway, based on these reports, we are can speculate that postoperative hair epilation could be proposed to our patients, to maximize the results of EPSiT and to reduce the likelihood of recurrences.

A limitation of our study is represented by the relatively short median follow-up that could limit the possibility to identify late recurrences, as those reported by Meinero et al. in a recent prospective multicentre trial [12]. Another limitation is represented by the absence of comparison of the results of our series with those of other commonly performed techniques, either retrospective or prospective. In fact, we could only compare our results with available literature data and reports. Nonetheless, given the excellent results provided by EPSiT, at least in our series of patients, we can conclude that endoscopic pilonidal sinus treatment (EPSiT) in paediatric population represents a safe, effective and versatile option for the treatment of pilonidal diseases that could become the gold standard for its treatment in the next future.

Author contributions APP designed the study and drafted the manuscript, CM, GM, ME, CE, AD, LCA, LL, CC and FR enrolled patients and collected notes and details from each participating surgical unit, PCM revised the drafts of the final version of the paper.

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Compliance with ethical standards

Ethical approval Not requested nor needed (procedure already adopted either in adults and pediatrics).

Conflict of interest The author Meinero PC is a consultant for Karl Strotz (Karl Stortz, 78532 Tuttlingen, Germany) and patented and invented the fistuloscope used in most centers to perform EPSiT and VAAFT procedures. All other authors declare that they have no competing interests.

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