

Transperineal ultrasonography in perianal Crohn's disease and recurrent cryptogenic fistula-in-ano

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Abstract

Aim Successful anal fistula care is aided by specialized imaging accurately defining the site of the internal opening and fistula type. Imaging techniques are complementary, designed to answer specific anatomical questions. There are limited data concerning the clinical value of transperineal ultrasound (TP-US) in both cryptogenic fistula-in-ano and perianal Crohn's disease (PACD). The aim of the study was to assess the accuracy of TP-US compared with operative findings in patients with perirectal sepsis.

Method Patients with recurrent cryptogenic anal fistula and PACD referred for sonography were examined using TP-US by a single examiner blinded to the operative results. Fistulae were categorized by the Parks classification predicting the site of the internal fistula opening. Ancillary horseshoe collections, abscesses and secondary tracks were defined.

Results Fourteen patients with PACD and 27 patients with recurrent cryptogenic fistula-in-ano were analysed with comparative images and operative data. Correlation of fistula type for cryptogenic and PACD patients respectively was 23/27 (85.2%) and 12/14 (85.7%),

with a correlative internal opening site (when found at surgery) of 16/22 (72.3%) and 12/14 (85.7%). Misclassification of fistula type in cryptogenic cases occurred in the presence of ancillary abscesses with associated acoustic shadowing. In PACD patients, TP-US was used when anal stenosis precluded endoanal ultrasonography, assisting in the diagnosis of recto-vaginal fistulae.

Conclusion TP-US is a useful complementary technique to assess fistula-in-ano and has special advantage when there is anal canal distortion, complex fistula type or suspicion of a recto-vaginal fistula.

Keywords Transperineal ultrasound, fistula-in-ano, perianal Crohn's disease

What is new in this paper?

Transperineal ultrasonography, although little used, provides useful information in complex and recurrent perirectal sepsis. It assists in the definition of the primary fistula track, translevator and supralelevator extensions and in the diagnosis of recto-vaginal fistula. It is a complementary diagnostic tool for complex fistula-in-ano.

Introduction

The bulk of cryptogenic anal fistulae are relatively simple and do not require specialized imaging for successful management [1,2]. Although the indications for imaging and the type to be used in complicated and recurrent cases are controversial, MRI has the highest sensitivity for the definition of the anatomy of primary and secondary tracks and collections [3]. In this regard, the principal

reason cited for fistula recurrence after surgery is the failure to locate the internal opening of the fistula track [4]. Perianal sepsis is also a common manifestation of Crohn's disease, occurring in 20–40% of cases during the course of the disease [5] and frequently preceding disease diagnosis, particularly in children [6]. The array of perianal lesions in Crohn's disease (PACD) is complex, resulting in significant morbidity from destructive perirectal sepsis and incontinence, where many patients undergo multiple anal procedures accompanied in some cases by proximal diversion or proctectomy. In PACD, the relative success of both medical and surgical approaches is also dependent upon accurate preoperative and intra-operative

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disease classification [7] which in turn is contingent upon reliable imaging methodologies, in particular MRI [8].

For both cryptogenic disease and PACD, two- and three-dimensional endoanal ultrasonography (EAUS) which is more widely available than MRI has shown high accuracy for the delineation of complicated fistula-in-ano as well as the capacity precisely to define its relationship to the main sphincter complex [9,10]. However, given the complexity of some cases, many patients with severe PACD may have recto- or ano-vaginal fistula or anal stenosis precluding the insertion of an endoanal ultrasound probe, where disease may extend beyond the focal distance of an endoluminal transducer and which is difficult to delineate accurately even with EAUS. Such cases frequently necessitate the complementary use of other techniques to diagnose the extent and type of perianal pathology [11]. Previously, our group has shown moderate accuracy of transperineal ultrasound (TP-US) in the delineation of the preoperative anatomy of *de novo* and recurrent cryptogenic fistula-in-ano [12], with similar benefit being described by others using this technique in selected patients with complicated PACD [13,14]. In this study, we have blindly assessed TP-US in patients with recurrent cryptogenic fistula-in-ano and with PACD to determine its accuracy in the definition of fistular and abscess anatomy compared with operative findings under anaesthesia and to establish a potential complementary clinical role for TP-US in complex perirectal sepsis.

Method

Permission for retrospective analysis of patients undergoing TP-US for assessment of PACD and recurrent cryptogenic fistula-in-ano was provided by the local institutional review board. Patients were recruited from the inflammatory bowel disease clinic in the Department of Gastroenterology at the Chaim Sheba Medical Center, Israel, referred for ultrasound assessment and from the endoanal ultrasound database of patients with cryptogenic sepsis referred before surgery. All cases of PACD were confirmed on local histology or from histology of previously resected specimens or endoscopy.

Transperineal ultrasound technique

Transperineal ultrasound (TP-US) was performed by one of the authors (MB-G) who was blinded to the operative findings. It was conducted initially by identifying the axial anal canal which provided images similar to those obtained with EAUS as previously described

[12] using a curvilinear 7.5 MHz probe (B-K Copenhagen, Denmark). During axial imaging pressure was applied to the perineal body with the probe angled and rocked back and forth to permit full visualization of the complete length of the anal canal as previously described [15]. The site of entry of a primary fistula track is recorded by its position on the clock face. The transducer was placed on any visible external opening with oblique images being obtained so that the course of a visible external fistulous track could be followed free-hand and traced back to determine its relationship to the sphincter complex. In patients with a recto- or ano-vaginal fistula, TP-US was supplemented in some

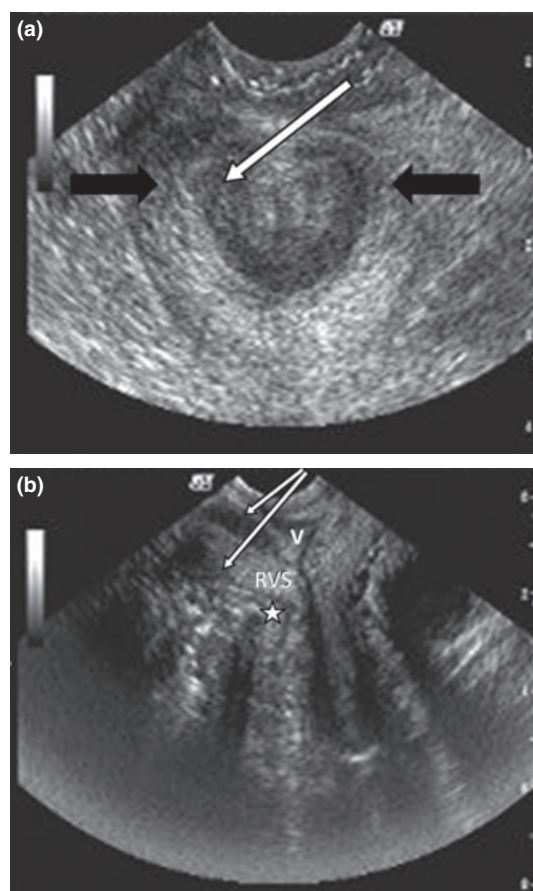


Figure 1 (a) Axial transperineal ultrasonography (TP-US) image showing landmarks that are similar to those obtained with two-dimensional endoanal ultrasound. The internal anal sphincter appears as an hypoechoic ring (white arrow) with the external anal sphincter appearing as an hyperchoic complete ring flattened anteriorly (black arrows). (b) Sagittal TP-US image. The hypoechoic internal anal sphincter (arrows) is identified above and below the anal canal in profile. The area of the puborectalis is seen end-on (star). The vagina (V) may be instilled with contrast if required to delineate the recto-vaginal septum (RVS).

Table 1 Demographics of 14 patients with perianal Crohn's disease (CD) undergoing transperineal ultrasonography.

Patient	Age (years)	Gender	Duration CD	Prior CD	Biological use	Prior anorectal surgeries
1	26	F	5 years	Ileal	–	2
2	25	F	4 years	PACD only	–	1
3	27	F	20 years	Colonic	–	3
4	27	F	6 months	PACD only	–	2
5	14	M	2 years	Colonic	+	1
6	50	F	6 months	PACD only	–	2
7	27	F	6 months	PACD only	–	3
8	32	M	12 years	Ileocaecal	–	4
9	36	F	3 months	PACD only	–	1
10	66	F	3 years	Mixed SB/ colonic	+	3
11	44	F	25 years	Ileocaecal	–	3
12	29	F	10 years	Colonic	+	3
13	40	M	19 years	Colonic	–	1
14	19	F	8 years	Mixed SB/ colonic	+	5

PACD, perianal Crohn's disease; SB, small bowel

cases by trans-introital sonography. In sagittal mode (after counterclockwise rotation of the transducer head by 90°), the fistula was followed for its relationship to the hypoechoic ring of the internal anal sphincter (seen as two parallel hypoechoic lines lying above and below the anal canal) and for assessment of the level in relation to the puborectalis seen *en face*. Axial and sagittal images obtained with the TP-US technique and the relevant anatomical landmarks for a diagnosis of fistula-in-ano are shown in Fig. 1.

Fistulae were divided by the Parks classification [16] into intersphincteric, trans-sphincteric, suprasphincteric and extrasphincteric with the definition of any abscess cavity as perianal or ischio-rectal with circumferential (horseshoe) infralevator or supralevator extension. The opening of a fistulous track seen on TP-US was described in accordance with the endoluminal ultrasonographic criteria previously established for diagnosis of an internal opening by Cho as a hypoechoic break in the internal anal sphincter and the subepithelial space [17]. This was recorded for operative comparison in relation to the clock face (anterior, posterior, right- and left-hand sides) that would be detected at surgery in the lithotomy position. Definition of the internal opening was where appropriate supplemented by hydrogen peroxide installation as previously described for ultrasonographic fistula enhancement with EAUS [18]. Additional data obtained included the presence of significant horseshoeing of the primary track, the presence of any secondary tracks or collections (and their

relation to the puborectalis) and any other ancillary information such as the presence of an ano- or recto-vaginal fistula and external or internal anal sphincter damage.

Patient subgroups

Transperineal ultrasound (TP-US) and examination under anaesthesia were performed on 14 patients with proven PACD during the period from 30 June 2007 until 4 April 2012, with surgery being carried out within a mean of 5 weeks (range 7 days–4 months) after ultrasound examination. All patients had pre-diagnosed Crohn's disease confirmed on biopsy based on prior resection specimens, endoscopy, associated CT or MR enterography and perianal biopsy.

TP-US was performed on 27 patients with a recurrent cryptogenic fistula-in-ano during the period from 30 June 2010 to 4 April 2012 where operative data for comparison were available (median number of operations one, range 1–4). The mean period of time between ultrasound examination and surgery was 40.6 (range 1–90) days.

Results

Crohn's disease patients

During the study period 14 patients (11 females) were analysed with a median age of 28 (14–66) years

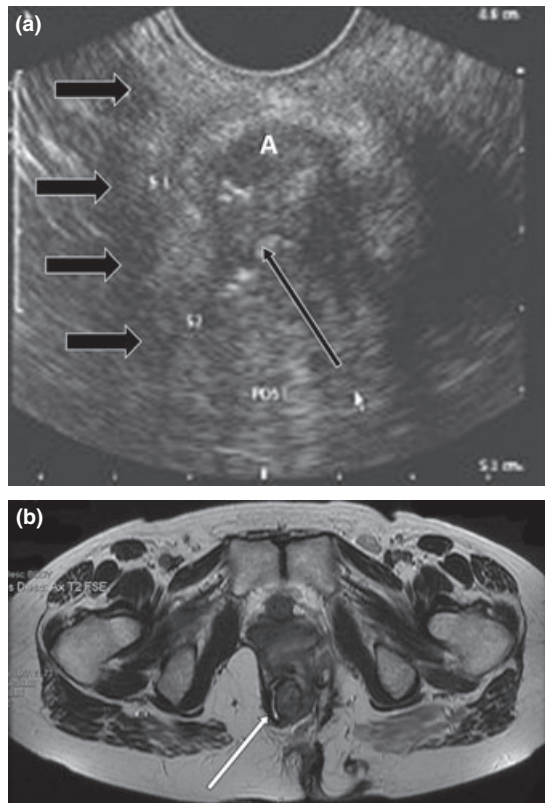


Figure 2 (a) Transperineal axial ultrasound in a female with severe anal stenosis preventing insertion of an endoanal probe. A circuitous fistula beginning in the midline posteriorly (arrow) extends anteriorly (arrowheads) and was traced to the right labium majorum, although a recto-vaginal fistula could not clearly be demonstrated on sagittal transperineal ultrasonography views. A, anal canal. (b) The corresponding MRI image (T2 FSE image) showing a circuitous gadolinium-enhancing track extending alongside the anal canal to the perineum (arrow).

(Table 1). The mean duration of Crohn’s disease was 7.8 years with a range of 0.25–25 years, with five of the patients having isolated perianal disease. In two patients a significant anal stenosis and pain precluded deployment of an endoanal probe, and only TP-US could be performed. Both cases were supplemented by MRI which confirmed the TP-US and operative findings. Overall, four patients in this group underwent comparative MRI. In one case, there was a high trans-sphincteric fistula with an opening confirmed at the 6 o’clock position posteriorly, coursing anteriorly towards the right labium majorum with evidence of a recto-vaginal fistula in a patient with anal stenosis preventing insertion of an endoanal probe (Fig. 2).

Four patients were treated with biological therapy with a median number of anorectal procedures amongst the cases of 2.5 (1–5). Table 2 shows the comparison of TP-US imaging with surgery for fistula type, determination of the internal opening, the presence of secondary abscess formation and horseshoe extension (Fig. 3). Ultrasound correlated with the surgical identification of the internal opening in 12/14 (85.7%) cases (Fig. 4) and with fistula type in 12/14 (85.7%). In three patients ultrasound indicated the presence of a recto-vaginal fistula, each confirmed at surgery. In one patient a trans-sphincteric fistula was over-assessed by ultrasound as suprasphincteric (Fig. 5), and in one extrasphincteric fistula TP-US suggested that the fistula was suprasphincteric. Five abscesses found at the time of surgery were all confirmed with TP-US, three of which were translevator. All horseshoe abscesses were detected by TP-US although two secondary extensions were missed, one extending suprasphincterically and one into the gluteal muscles.

Table 2 Transperineal ultrasonography (TP-US) findings with surgery in 14 patients with perianal Crohn’s disease.

TP-US	Surgery								
	ISF	TSF	SSF	ESF	RVF	IO	Abscess	2nd track	Horseshoe
ISF	–								
TSF		10							
SSF		1	1						
ESF				2					
RVF					3				
IO						12			
Abscess							5*		
2nd track								4†	
Horseshoe									4

ISF, intersphincteric fistula; TSF, trans-sphincteric fistula; SSF, suprasphincteric fistula; ESF, extrasphincteric fistula; RVF, recto-vaginal fistula; IO, internal opening identified and correlated; Abscess, abscess identified; 2nd track, secondary track identified; Horseshoe, horseshoe abscess formation.

*Supralelevator abscess.

†Four secondary tracks detected at surgery but only two identified with combined TP-US plus endoanal ultrasonography.

Cryptogenic fistula cases

Of the 27 patients (two females) with recurrent cryptogenic fistula-in-ano who underwent TP-US and examination under anaesthesia for comparison, the median overall age was 40 years (range 4–75 years). The median number of operations in this group was one with a range of 1–4, and the mean interval between ultrasound and surgery was 40.6 days with a range of 1–90 days. Overall three patients in this group underwent additional MRI. Table 3 shows the comparison of TP-US imaging *vs* surgery for fistula type, determination of the internal opening, the presence of secondary abscesses and extensions and for horseshoe abscesses. In five patients a fistula was predicted by TP-US examination (four trans-sphincteric and one extrasphincteric), whereas at surgery only an abscess (all ischiorectal and infralevator) was found without an internal opening detected. Where an internal opening was detected at surgery TP-US correctly predicted its presence and location in 16/22 (72.3%) patients. The fistula type was correctly identified in 23/27 (85.2%) patients. In two patients, a horseshoe abscess was detected at operation where the fistula was trans-sphincteric and where such an abscess extension was not identified by TP-US. In four patients ancillary abscesses around the main fistula track were identified by TP-US and at operation. Two secondary intersphincteric infralevator tracks leading away from the primary fistula were detected by TP-US and confirmed at surgery.

Discussion

This small select study showed that hand-held TP-US is an accurate imaging adjunct to surgery in the diagnosis of fistula type in both recurrent cryptogenic fistula-in-ano and perianal Crohn's disease. It is capable of diagnosing the site of the internal opening of the main fistula track in 70–85% of cases. Prospective blinded studies comparing different ultrasonographic modalities are currently under way to determine the specific advantage of TP-US in particular clinical situations. Despite its limitations, the study confirms that TP-US is particularly accurate in determining the relationship of the fistula track to the sphincter although there may be difficulty in the precise identification of horseshoe tracks and secondary abscess formation, as previously reported by our group for never operated and recurrent cryptogenic fistula [12]. In the case of trans-sphincteric fistula in particular, it may be that gas-containing collections are less well demonstrated by TP-US possibly because of excessive acoustic shadowing in the anal canal. The relative inability of TP-US to delineate some larger sec-

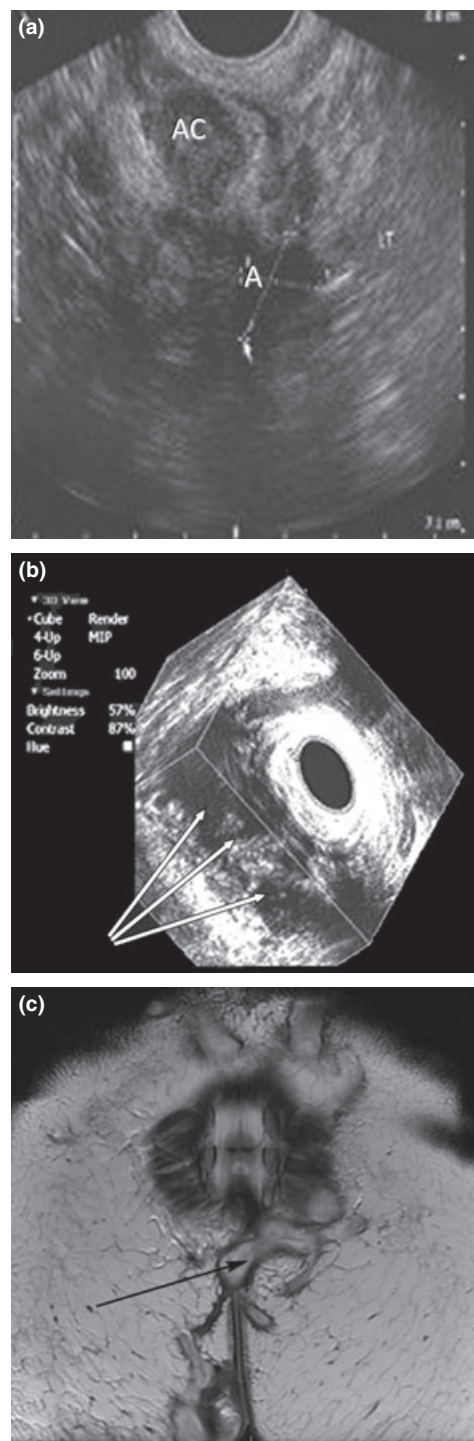


Figure 3 (a) Horseshoe abscess (axial transperineal ultrasound image). The anal canal (AC) with the hypoechoic internal anal sphincter is shown with the abscess posteriorly located (A). (b) The corresponding three-dimensional endoanal ultrasound view showing an extensive horseshoe abscess (arrows) in a patient with perianal Crohn's disease. (c) The corresponding MRI (T2 FSE) view of the extensive horseshoe collection (arrow).

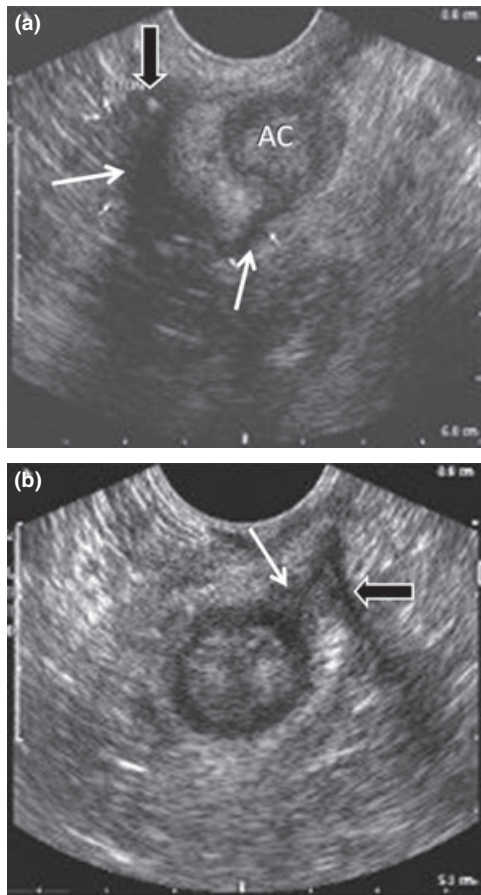


Figure 4 (a) Posterior internal opening site on transperineal ultrasonography axial image in a patient with anal canal stenosis where an endoanal ultrasound probe could not be deployed. The fistula (arrows) is shown clearly entering the anal canal (AC) posteriorly in the midline. In this case a hyperechoic seton is also identified (block arrow). (b) Anterior internal opening site seen on transperineal ultrasonography axial image (arrow) coursing laterally into the ischiorectal space (arrow-head).

ondary abscesses has also been previously reported by Wedemeyer *et al.* [13]. Here it may be that a hypoechoic para-anal collection will prevent an adequate transperineal examination particularly if it is large or contains gas which reduces the contrast with the air-filled anal canal. A smaller very low lying abscess may also not be readily detected by endoluminal ultrasound [19] so that in the circumstance where the EAUS images are non-contributory it is wise to supplement the ultrasound examination by a transperineal approach.

Transperineal ultrasound (TP-US) may also allow simultaneous digital anal examination which may be helpful to assess subcutaneous tracks and collections by giving a better three-dimensional understanding than that provided by conventional EAUS. Furthermore

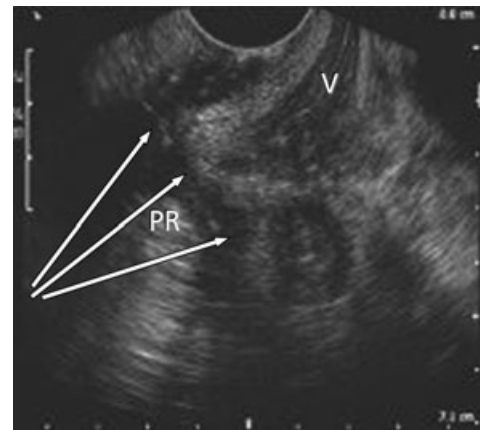


Figure 5 Example of an erroneously diagnosed suprasphincteric fistula on transperineal ultrasound in a patient with anal canal stenosis. The sagittal image shows the vagina (V) and the puborectalis (PR) with a circuitous fistula (arrows) tracking around the puborectalis muscle. At operation the fistula was found to be of infralevator trans-sphincteric type with a translevator extension and an internal opening at the dentate line rather than a true suprasphincteric fistula.

TP-US distinguishes perianal from perineal disease and anal fistula from hidradenitis suppurativa. The present study suggests no benefit for the instillation of hydrogen peroxide in TP-US to show secondary tracks, a finding confirmed using a similar technique to ours by Kleinübing *et al.* [20] although this was not specifically examined in our patient cohort.

One particular advantage of TP-US appears to be in selected patients with PACD, particularly where there is anal stenosis (about 20%) or discomfort which prevents insertion of the endoanal probe [21]. In both cases TP-US showed the presence of complex disease with supralevator extension and recto-vaginal involvement where the probe could be used to trace the relationship of the fistula track to the pelvic floor, as confirmed by MRI and at surgery.

Although there are limitations of the present study in which TP-US and EAUS were not directly compared, TP-US appears relatively accurate in the definition of fistula type and in the identification of the internal opening in patients with recurrent cryptogenic fistula and in those with PACD. Transperineal sonography is helpful in patients with anal pain and stenosis where the use of an endoanal probe is impractical and can define secondary tracks running towards the vagina and labia which are beyond the focal length of the endoluminal probe.

Transperineal sonography may demonstrate some secondary abscesses and horseshoe extensions, but large gas-containing abscesses may be missed, particularly in patients with a trans-sphincteric fistula because of acoustic shadowing in the anal canal. Tracks above the levator

Table 3 Transperineal ultrasonography (TP-US) findings with surgery in 27 patients with cryptogenic fistula-in-ano.

TP-US	Surgery								
	ISF	TSF	SSF	ESF	RVF	IO	Abscess*	2nd track	Horseshoe
ISF	–	1							
TSF		21					1		2
SSF			1	2					
ESF			–	2					
RVF					–				
IO						15			
Abscess							4		
2nd track								2	
Horseshoe									2

ISF, intersphincteric fistula; TSF, trans-sphincteric fistula; SSF, suprasphincteric fistula; ESF, extrasphincteric fistula; IO, internal opening identified and correlated; Abscess, abscess identified; 2nd track, secondary track identified; Horseshoe, horseshoe abscess formation.

*In five cases a fistula was predicted by combined ultrasound but no internal opening was detected at surgery.

are well demonstrated where an endoanal probe may fail to see them or where MRI would normally be required to demonstrate supralelevator extension or a primary pelvirectal origin. In some patients with anal stenosis and an anterior fistula TP-US may be supplemented by transvaginal ultrasound [22].

It is recommended that the colorectal trainee and consultant become conversant with TP-US so that certain surgical questions can be answered by its use. This suggests that a formal accreditation process of hands-on instruction may be valuable during colorectal training. Based on these preliminary data TP-US should be complementary to other methods of assessment of complicated cryptogenic anal fistula, particularly those associated with PACD.

Author contributions

MB-G, APZ: study concept and design. AN, MB-G, AS, AL, DC: data acquisition. AN, DC: analysis. MG, APZ: writing of the manuscript.

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