Early Experience in the Surgical Management of Perianal Fistulizing Crohn's Disease: Cross-sectional Study

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ABSTRACT

Introduction: This is our initial experience in the surgical management of perianal fistulizing Crohn's disease (PFCD). This is a cross-sectional review of consecutive Crohn's disease patients presenting with perianal fistula who were surgically managed between November 2020 to September 2024.

Objectives. 1) To present profiles of patients with PFCD; 2) To classify patients with PFCD using the American Gastroenterological Association (AGA) classification; and 3) To describe our approach to management.

Methods: Only patients diagnosed with Crohn's disease presenting with anal fistulas were included. Patient demographics, classification of anal fistula using the AGA classification were described. Pelvic MRIs were done in all patients to define the fistula tracts and detect the presence or absence of concomitant abscess. For each patient, the number of external and internal openings, and the type of surgical management were described.

Results: Twenty-nine patients with PFCD were surgically managed. There were 15 males and 14 females, ages ranging from 16 to 47 years (median age of 26.5 years). Six patients (21%) had prior history of anal operation for anal abscess and/or fistula. Using the AGA classification, all patients had complex fistula. On physical examination, 14 patients (48%) had two or more external openings. One-third of female patients had ano-vaginal fistula. At the time of surgery, two or more internal openings were demonstrated in more than half of patients (55%). Draining setons were performed in all patients.

Conclusion: Awareness of the possible association of inflammatory bowel disease to complex anal fistula offers an additional differential diagnosis to this challenging problem, and may alter approaches to disease management.

Key words: perianal fistulizing Crohn's disease, surgical management

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In the Philippines, Inflammatory Bowel Disease (IBD) is not a frequently considered differential diagnosis for gastrointestinal disorders. This can be attributed to the unfamiliarity with the various aspects of diagnosis and management of this complicated disease. Having no specific defining clinical or pathologic feature[1], IBD can be confused for extra-pulmonary or intestinal tuberculosis. Further, pathologic reports of "non-specific colitis" added to the diagnostic and management dilemma.

The IBD Club of the Philippines was established in 1990 to promote awareness and recognition of IBD as an emerging health concern in the country. Bringing together specialists from Gastroenterology, Pathology and Radiology, the regular conferences served as a platform for multi-disciplinary meetings. Discussions on the role of endoscopy with proper specimen collection, quality pathology reporting and the correct use of the different imaging modalities such as Computed Tomography (CT) scan and Magnetic Resonance Imaging (MRI), contributed to the advancement in knowledge and improved ability to diagnose inflammatory bowel diseases. Just recently, colorectal surgeons have also become part of this growing multi-specialty group.

There are two main types of inflammatory bowel disease: ulcerative colitis (UC) and Crohn's disease (CD). While UC mainly involves the rectum and colon, involvement of CD can be anywhere in the gastrointestinal tract, from the mouth to the anus.

The Montreal classification (2003) for IBD is used to subclassify UC and CD for clinical and research purposes[2]. Among the spectrum of manifestations of Crohn's disease is fistulizing disease affecting the perianal area (Table 1). Referred to as perianal fistulizing Crohn's disease (PFCD), there is no current consensus for classification. The most widely used is the classification system developed by the American Gastroenterological Association (AGA)[3] (Table 2).

The authors are presenting their initial experience in the surgical management of PFCD. The objectives of this

Table 1. Montreal classification of Crohn's disease (2005)

Age at diagnosis (A)	A1- below 16 y A2- between 17-40 y A3- above 40 y
Location (L)	L1- ileal L2- colonic L3- ileocolonic L4- isolated upper disease
Behavior (B)	B1- non-stricturing, non-penetrating B2- stricturing B3- penetrating

"p" is added to B1-B3 when concomitant perianal disease is present

Table 2. American Gastroenterological Association (AGA) classification of PFCD (2003)

SIMPLE fistula	**COMPLEX** fistula
LOW - Superficial or low intersphincteric or low transsphincteric origin of the fistula tract	HIGH - High intersphincteric or high transsphincteric, or - Extrasphincteric or suprasphincteric origin of the fistula tract
Single external opening	Multiple external openings
No pain or fluctuation to suggest perianal abscess	Presence of pain or fluctuation to suggest a perianal abscess
No evidence of a rectovaginal fistula	May be associated with the presence of a rectovaginal fistula
No evidence of anorectal stricture	May be associated with the presence of an anorectal stricture
	May be associated with the presence of active rectal disease at endoscopy

*Additional diagnostic evaluation with examination under anesthesia (EUA), anorectal EUS, and pelvic MRI may be required to accurately classify some patients

paper are: 1) to present profiles of patients with PFCD, 2) to classify patients with PFCD using the American Gastroenterological Association (AGA) classification, and 3) to describe our approach to patient management.

METHODS

Study Design and Setting

This is a cross-sectional review of consecutive patients with perianal fistula associated with Crohn's disease surgically managed by the authors (RL, MAF) in four hospitals in Metro Manila, Philippines (Delos Santos Medical Center, Cardinal Santos Medical Center, East Avenue Medical Center, Gen. Miguel Malvar Medical Foundation Hospital) between November 2020 and September 2024.

Patients

The diagnosis of Crohn's disease was established during colonoscopy by demonstrating one or more inflammatory changes (mucosal edema, ulcerations, strictures, pseudo-polyp formation, cobble-stoning), supported by biopsy and imaging studies. Though there are many perianal manifestations of Crohn's disease, only patients with anal fistulas were included. Patient demographics, classification of anal fistula using the AGA classification were described. Pelvic MRI using anal fistula protocol were requested in all patients to define the fistula tracts and detect the presence or absence of concomitant abscess. At the time of surgery, the number of external and internal openings, and the type of surgical management were recorded. Obtaining additional tissues for histopathology was not routine, performed when the surgeon felt there was need to confirm or rule out proctitis.

Operative Technique

All operations were performed under regional anesthesia (except for one patient presenting with septic encephalitis for severe perineal sepsis who had to be operated under general endotracheal anesthesia). Positioning patient supine or on dorsal lithotomy depended on patient habitus, ease of exposure and surgeon's preference. After positioning, an anal speculum retractor was inserted to examine the anal canal and lower rectum. It is essential at this point to have the MRI images available for review and correlation. An 18-gauge IV catheter was introduced into the external opening and injection of hydrogen peroxide was our preferred method to identify the internal opening. After injection of hydrogen peroxide, bubbling inside the anal canal indicated the site of the internal opening. Once the internal opening was identified, gentle probing was performed from the external opening towards the site of the internal opening to establish the course of the fistula tract. For multiple external openings, the same maneuver was performed on the other external openings. This was done to determine if other external openings drained into one common internal opening, or drained independently to separate internal openings. Bubbling in the anal canal at a site distinctly separate from the initially identified internal opening established the presence of more than one internal opening. With the fistulous tract identified, a silicone vessel loop was used as seton and passed from the internal opening to the external opening, creating a loop encompassing the internal opening, fistulous tract, anal sphincters, overlying perianal skin, and the external opening. Making sure the loop was not constricting the underlying structures, the ends were tied. The anal sphincter muscles and the perianal skin overlying the fistula tract were not transected. For fistulas with multiple internal openings, the same technique of seton placement was repeated for each tract.

RESULTS

From November 2020 to September 2024, there were thirty-one consecutive patients with perianal Crohn's disease. Two patients were excluded: one patient underwent incision biopsy at the perianal area for suspicious malignant transformation, and another patient underwent anal dilatation for stenosis. The remaining 29 patients all had anal fistulas. Two patients (2/29 or 6.8%) had no evidence of luminal inflammation. After excluding other causes (iatrogenic injury, obstetric, trauma, radiation, and cancer) from history and physical examination, both patients were given presumptive diagnoses of PFCD due to the complex nature of the fistula, and included in this review. There were 15 males and 14 females, ages ranging from 16 to 47 years (median age of 26.5 years). Six patients (21%) had prior history of anal operation for abscess and/or fistula. Using the AGA classification, all patients fulfilled the definition of complex fistula (see Table 2 definition). Fourteen patients had two or more external openings (48%). One-third of female patients presented with ano-vaginal fistula. Peri-operative antibiotics were not routine, and was given only when there was documented abscess on pelvic MRI, cellulitis of the perianal skin, or presented with systemic manifestations, such as fever.

On examination under anesthesia, the locations of the internal openings were not confined to the area around the dentate line. With the use of hydrogen peroxide injected through the external opening, sites of bubbling (internal opening) were demonstrated anywhere from the mucocutaneous junction to the lower rectum. In 16 out of 29 patients (55%), two or more internal openings were identified. Some patients had single external opening draining into two separate internal openings. After proper identification of the fistula tracts, and abscesses evacuated whenever present, draining setons were placed in all patients (Table 3). There were five patients with draining secondary tracts, defined as one or more branching tracts extending from an external opening. In these cases, the depth of the skin and muscle involvement of the secondary tracts were determined: a) if the branching tract involved the superficial skin and muscle layer, the tract was laid open, b) if with deeper involvement of the muscle, a draining seton was placed. The number of setons placed ranged from one to eight per patient.

Table 3. Summary of patient profiles and treatment of patients with PFCD (n=29)

	Male (n= 15)	Female (n= 14)
Age	16- 47 y (median 28.5 y)	16- 33 y (median 24.15 y)
Age (Montreal classification) A1 (<16 y) A2 (17-40 y) A3 (>40 y)	 0 13 2	 0 14 0
AGA classification of anal fistula	SIMPLE: 0 COMPLEX: 29	
Number of external openings 1 2 or more	 15/29 (52 %) 14/29 (48 %, from 2-5)	
Ano-vaginal fistula	N/A	5 (35.7 %)
Prior operations	6/29 (21 %, range 1-6 operations)	
Number of internal openings 1 2 or more	 13/29 (45%) 16/29 (55 %, from 2-4)	
Number of setons placed	1 – 8 (average 2.2) per patient	

Two patients (6.8%) had no endoscopic and radiologic signs of inflammation. The first male patient had previous drainage of the perianal abscess, and presented with five external openings. During surgery, four internal openings were demonstrated. The other female patient had one internal opening with two external draining sites: one at the lower vagina and the second at the vulva. Both patients were managed with draining setons.

One female patient presented with septic encephalopathy at the emergency room from perineal sepsis from the ano-vaginal fistula with multiple internal and external openings. After resuscitation and antimicrobial therapy, she was operated on the third hospital day. Multiple pockets of abscesses were drained, and three setons were placed for separate internal openings identified between the anal canal and vagina. Another five setons were needed for secondary tracts that encompassed much of the external anal sphincters. The patient stayed in the hospital for more than two weeks, but was subsequently discharged improved. The rest of the patients had uneventful post-operative courses. Need for analgesics were minimal, and all were discharged the following day.

Some atypical features observed among patients with PFCD in this review are listed in Table 4.

DISCUSSION

In the natural history of Crohn's disease, perianal fistula (PFCD) develops in approximately 20 percent of patients[4]. An ulcer develops in the anal canal or lower rectum resulting from chronic inflammatory changes in CD. Another process soon develops where epithelial cells in the ulcerated area are transformed into mesenchymal cells, a process known as epithelial-mesenchymal transition (EMT). In EMT, newly transformed mesenchymal cells gain migratory and invasive properties[5]. The ulcer penetrates the full thickness of the anal canal (or rectum), resulting to abscess formation, and finally exiting through the perianal skin as a fistula. The initiating event (ulcer formation) can occur anywhere in the anal canal and rectum, and may or may not involve the anal glands. Therefore, the location of the internal opening may not be confined or limited to the level of the dentate line (the common site of cryptoglandular infection). Some have described this migratory and invasive behavior as showing little respect for anatomic barriers[6]. The unpredictability of the location and number of the internal openings, and tracts that do not conform to anatomic planes, makes surgery for PFCD technically challenging. Operative findings were so variable that it was difficult for us to predict a disease pattern. Many of our observations during the physical examination and at the time of surgery, demonstrated many gross differences between Crohn's and cryptoglandular fistulas (Table 4).

While cryptoglandular fistula are mainly treated with surgery, CD-associated fistula follows a different course of management. Our management practices for PFCD's are based on current standard literature and expert consensus statements. In the absence of rectal disease, simple low-lying fistula can be treated with fistulotomy[7]. Complex fistulas are best managed by a combined medical-surgical approach. Pharmacologic therapy is an important backbone in management. Anti-TNF medications (Infliximab) remains the first-line intervention and the gold standard[8]. Prior to commencement of biologic therapy, seton placement is the current practice standard. Pelvic MRI with anal fistula protocol defines the course of the fistula tracts and can identify the presence or absence of abscesses. Seton placement, with or without abscess drainage, is important to maintain patency of the tract, and minimizes the formation of other tracts and recurrent abscess formation, all essential while the patient is undergoing medical treatment[9].

Two patients (6.8%) in the current study had no endoscopic nor radiologic signs of inflammation. There are literature reports of perianal disease that sometimes precede intestinal Crohn's disease[10]. Isolated perianal Crohn's disease (iPCD) is used to describe patients with complex fistula in the absence of luminal inflammation, with a series reporting a 14.5 percent occurrence[11]. It is unclear whether the condition represents a truly isolated cryptoglandular fistula, or early presentation of an undiagnosed CD. For iPCD, the best treatment option is still a clinical dilemma.

IBD and its subtypes, are emerging health problems in the country. Because the pathogenesis is still unclear, current treatment modalities are mainly towards symptoms control, rather than achieving cure. Patient's informed consent included a thorough explanation of the nature of the disease, its recurrent and progressive nature, and that seton placement is the first step (a bridge) to subsequent interventions. Biologic therapy will commence after the procedure, and a surveillance period will follow to monitor response to treatment. It is important that patients understand that seton placement is not considered as definitive treatment.

Cryptoglandular infection remains the most common cause of anal fistulas[12]. This paper presents an additional differential diagnosis to consider for complex anal fistulas. The presence of IBD can provide an alternative explanation to why some perianal fistulas are "resistant" to multiple surgical attempts, and provide a way out from the usual runabout of anal fistula procedures. Finally, a

Table 4. Atypical features observed in patients with perianal fistulizing Crohn's disease (PFCD)*

On physical examination:	
1. The external opening does not have the usual "heaped-up" appearance	
2. Presence of inflammation at the perianal skin	
3. External openings that do not appear to drain into one common internal opening	
At time of surgery: 1. Unusual location of internal opening 2. More than one internal opening	

*Above findings may not be consistently present in all patients.

sensible advice in the surgical approach to anal fistulas: "if there is any doubt … no attempt should be made to treat the fistula along conventional lines"[13].

CONCLUSION

Awareness of the possible association of IBD to complex anal fistula offers an additional differential diagnosis to this challenging problem, and may alter approaches to disease management.

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