Management of multiple cryptoglandular anal fistulas: evaluation of combined fistulotomy and seton application techniques

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Aim
The aim of this study was to evaluate combined fistulotomy and/or seton application for the management of multiple cryptoglandular anal fistulas in terms of recurrence and postoperative fecal incontinence.
Patients and methods
This study was carried out in Alexandria Main University Hospital, Alexandria, Egypt, by revising the medical files of all patients with multiple anal fistulas, who underwent fistulotomy, seton application or combined techniques, during the period spanning from December 2013 to June 2016.
Results
Twelve (10 male patients and two female patients) patients were reviewed, with a mean age of 41.75±7.75 years. Number of multiple fistulas had a mean of 2.58 ±0.90. For 11 patients, fistulotomy was performed for one or two fistulas (inter-sphincteric or low trans-sphincteric). The rest of the fistulas were treated by two-stage seton fistulotomy or draining seton application. One patient had two high trans-sphincteric anteriorly located fistulas, and both were treated by application of a draining seton. Among 31 fistulas in our patients, recurrence was encountered in two anterior high trans-sphincteric fistulas (6.4%) in two (16.7%) patients. Twelve months after the last intervention, our patients expressed acceptable continence status.
Conclusion
The condition of multiple cryptoglandular anal fistulas is an uncommon category of anal fistula. MRI is a perfect tool to diagnose the condition preoperatively. Combined fistulotomy and seton application seem a safe strategy for management of multiple anal fistulas with low postoperative recurrence and good postoperative continence. Further studies are required to explore more details about this neglected category of anal fistula.

Keywords:
cryptoglandular, fistulotomy, multiple anal fistulas, seton application

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Introduction

An anal fistula is a common troublesome pathology that may cause marked disability. Most of the anal fistulas are of cryptoglandular origin, but sometimes other pathologies are implicated, such as inflammatory bowel disease (IBD), tuberculosis, actinomycosis or anorectal cancer [1]. Anal fistulas could be of iatrogenic origin following anal or obstetric procedures. The estimated prevalence of cryptoglandular anal fistulas is 8.6–10/100 000 of the population per year, with a male to female ratio of 1.8 : 1.0 [2].

According to the cryptoglandular hypothesis, inter-sphincteric gland infection and abscess formation are the initiating events in the development of perianal fistulas [3]. An infected tract forms, starting at the mouth of one anal gland and ends externally on the perianal skin, discharging pus. Traditionally, an anal fistula has two openings, one internal and one external. In some cases, multiple infected tracts form and lead to multiple external openings, but still with the same internal opening. This is termed complex anal fistula [4].

Surgery is the basic treatment of anal fistula, aiming mainly at healing the fistulous tracts, eradication of infection, and preservation of anal sphincter muscles [5]. Thorough understanding of the anatomy of the anal sphincter and the pathology of perianal suppuration is essential for proper planning of management of anal fistulas [6].

The standard management of anal fistula is to lay open the infected tract between the internal and external

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openings, which is termed fistulotomy [1]. However, especially in complex and multiple fistulas, fistulotomy can be associated with significant risk of fecal incontinence in about 30% of patients. The severity of fecal incontinence increases with the complexity of the fistula [7,8]. This is why, in such cases, the surgeon can resort to other sphincter-saving procedures, the most commonly being used is seton placement [1].

In the literature, the condition of 'multiple cryptoglandular fistulas' is very poorly described or classified [9]. Moreover, management of such a condition is challenging, as aggressive surgical management carries the risk of multiple sphincteric injuries with subsequent fecal incontinence.

Therefore, the aim of our study was to evaluate combined fistulotomy and seton application for the management of multiple cryptoglandular anal fistulas, in terms of recurrence and postoperative fecal incontinence.

Patients and methods

This study followed a retrospective design, that included all patients complaining of multiple cryptoglandular anal fistulas, admitted to The Colorectal Surgery Unit, Alexandria Main University Hospital, during the period spanning from December 2013 to June 2016.

Inclusion criteria were as follows: patients with multiple anal fistulas who had undergone fistulotomy, seton application or combined techniques (Figs 1 and 2). We considered multiple anal fistulas as coexisting fistulas, each having its own external and internal openings, as proved by preoperative MRI or intraoperative examination. Exclusion criteria included patients with complex fistulas but with a single internal opening, patients with recurrent fistulas and patients with fistulas secondary to another pathology (e.g. malignancy, IBD, trauma, radiation, etc.), as proved by history and/or postoperative histopathology.

Files of patients who met the inclusion criteria were reviewed, and the following data were retrieved: demographic data, clinical presentations, number of fistulas, location and types of fistulas, preoperative MRI (if done), preoperative Wexner score [10], intraoperative findings, procedure performed, postoperative pathological report, recurrence, and postoperative Wexner score [10].

Outcomes

Primary endpoints

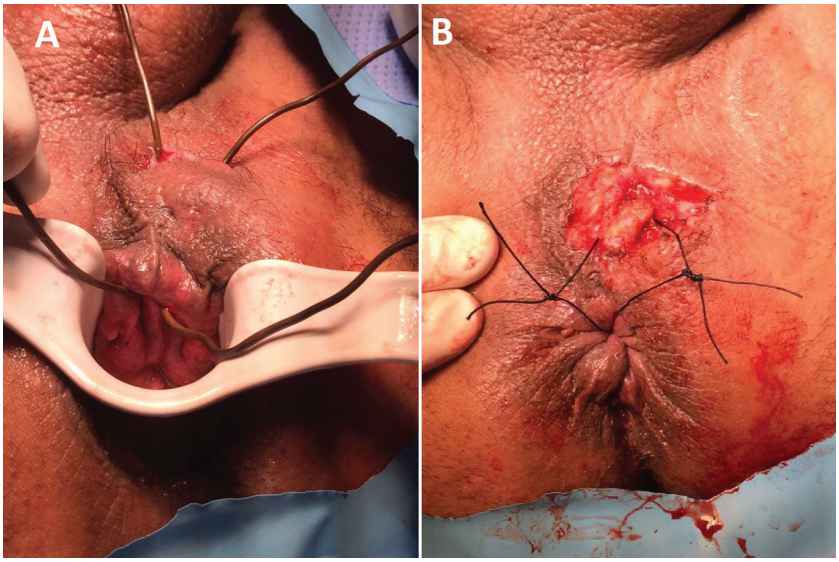
Recurrence: as detected by the surgeon in the outpatient clinic at 6, 9 and 12 months after the operation. Patients who missed follow-up were personally contacted.

(1) Incontinence reassessment, in comparison with the preoperative status using Wexner score [10] at 3, 6 and 12 months after the surgery during outpatient clinic follow-up visits. Patients who missed follow-up were personally contacted.

Statistical analysis

Collected data were entered into a computer using the statistical package for the social sciences (version 23),

Figure 1



(a); A case with two anterior high trans-sphincteric fistulas. (b); Draining seton application for both fistulas.

The International Business Machines Corporation (IBM), New York, United States. Descriptive statistics (frequency, percentage, range, median, mean, and SD) were calculated.

The manuscript was written in accordance with items of the PROCESS checklist [11].

The research was approved by the Ethical Committee of College of Medicine, Alexandria University (IRB 00007555).

Results

The study included 12 (10 male patients and two female patients) patients with the diagnosis of multiple cryptoglandular anal fistulas, who had undergone fistulotomy, seton application or combined technique. The age of patients ranged from 29 to 56 years, with a mean of 41.75±7.75 years.

Patients were complaining for periods ranging from 6 to 18 months, with a mean of 11.25±3.76 months. All patients had more than one fistula; however, some fistulas were minimally active at the time of presentation. The number of coexisting fistulas in our patients ranged from 2 to 5, with a mean of 2.58 ±0.90 fistulas. Details of anal fistulas with regard to their number and location are shown in Table 1.

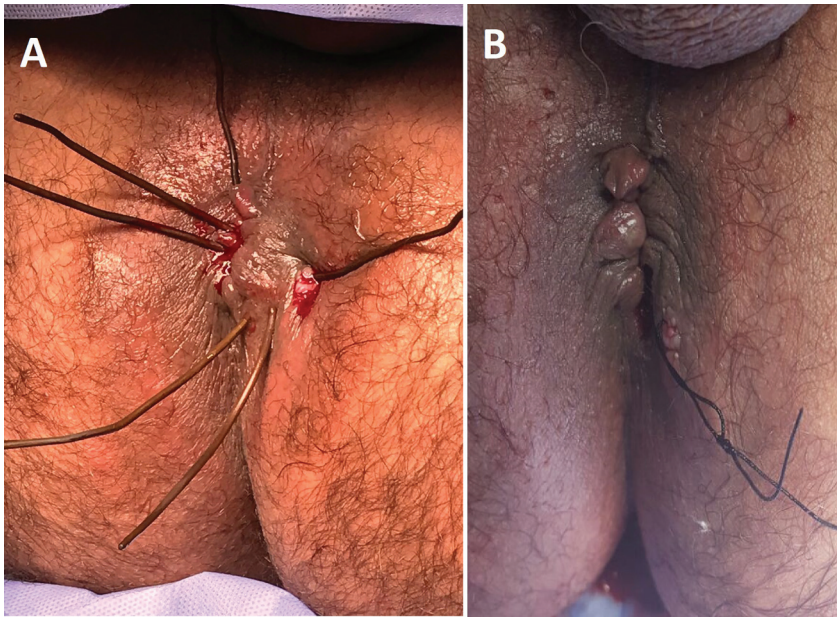
Preoperative MRI was performed in eight (66.7%) patients. MRI accurately diagnosed multiple anal fistulas in all of them. The other four (33.3%) patients were diagnosed intraoperatively. Preoperative Wexner score showed perfect continence for all patients.

All patients were carefully examined intraoperatively. For most patients, fistulotomy was performed for one or two fistulas (usually inter-sphincteric or low trans-sphincteric). The rest of the fistulas were treated by two-stage seton fistulotomy or draining seton application. One patient had two high trans-sphincteric, anteriorly located fistulas, and both were treated by draining seton, and no fistulotomy was done. Patients with two-stage seton fistulotomy were subjected to a second operation after 8 weeks to lay open the fistula and to remove the seton. For patients with draining seton, the setons were removed in the outpatient clinic after 8 weeks. Details of the operative management are shown in Table 2.

Recurrence was encountered in two patients, 6 and 7 months after the last surgical procedure (seton removal). Both had anterior high trans-sphincteric fistulas and were treated using draining seton application. They were offered alternative LIFT procedure, which was successful for one patient, while the other patient had another recurrence and was treated successfully by mucosal advancement flap.

On the basis of the Wexner score, our patients reported good results with regard to postoperative continence that showed further improvement with time. Wexner scores of our patients are shown in Table 3.

Figure 2



(a); A case of three multiple fistulas. (b); After laying open of inter-sphincteric and one low trans-sphincteric fistula and loose seton application for one trans-sphincteric fistula (as a step of two-stage seton fistulotomy).

Table 1 Details of clinical types and location of anal fistulas

Serial	Number of fistulas	Type of fistula 1	Location of external opening[a]	Type of fistula 2	Location of external opening[a]	Type of fistula 3	location of external opening[a]	Type of fistula 4	Location of external opening[a]	Type of fistula 5	Location of external opening[a]
Patient 1	2	High trans-sphincteric	1	High trans-sphincteric	2						
Patient 2	3	Low trans-sphincteric	3	Low trans-sphincteric	5	Inter-sphincteric	6				
Patient 3	3	High trans-sphincteric	5	Low trans-sphincteric	6	Inter-sphincteric	7				
Patient 4	2	Low trans-sphincteric	4	Low trans-sphincteric	6						
Patient 5	2	High trans-sphincteric	1	Inter-sphincteric	3						
Patient 6	2	Low trans-sphincteric	3	Low trans-sphincteric	5						
Patient 7	3	High trans-sphincteric	7	Low trans-sphincteric	9	Inter-sphincteric	6				
Patient 8	2	Low trans-sphincteric	6	Low trans-sphincteric	5						
Patient 9	3	Low trans-sphincteric	7	Inter-sphincteric	9	Inter-sphincteric	10				
Patient 10	2	High trans-sphincteric	6	Low trans-sphincteric	7						
Patient 11	2	Low trans-sphincteric	1	Inter-sphincteric	2						
Patient 12	5	Low trans-sphincteric	2	Low trans-sphincteric	11	Inter-sphincteric	12	High trans-sphincteric	7	Inter-sphincteric	6

[a]Described as O'clock location while a patient is in the lithotomy position.

Table 2 Operative management of patients with multiple anal fistulas in our study

Serial	Number of fistulas	Operative management
Patient 1	2	Draining seton for both fistulas
Patient 2	3	Laying open of low trans-sphincteric and inter-sphincteric fistulas and two-stage seton fistulotomy for low trans-sphincteric fistula
Patient 3	3	Laying open of inter-sphincteric fistula, Draining seton for high trans-sphincteric fistula and two-stage seton fistulotomy for low trans-sphincteric fistula
Patient 4	2	Laying open of low trans-sphincteric fistula and two-stage seton fistulotomy for low trans-sphincteric fistula
Patient 5	2	Laying open the inter-sphincteric fistula and two-stage seton fistulotomy for high trans-sphincteric fistula
Patient 6	2	Laying open of low trans-sphincteric fistula and two-stage seton fistulotomy for low trans-sphincteric fistula
Patient 7	3	Laying open of inter-sphincteric fistula, draining seton for high trans-sphincteric fistula and two-stage seton fistulotomy for low trans-sphincteric fistula
Patient 8	2	Laying open of low trans-sphincteric fistula and two-stage seton fistulotomy for low trans-sphincteric fistula
Patient 9	3	Laying open of two inter-sphincteric fistulas and two-stage seton fistulotomy for low trans-sphincteric fistula
Patient 10	2	Laying open of low trans-sphincteric fistula and two-stage seton fistulotomy for high trans-sphincteric fistula
Patient 11	2	Laying open of inter-sphincteric fistula and two-stage seton fistulotomy for low trans-sphincteric fistula
Patient 12	5	Laying open of two inter-sphincteric fistulas, draining seton for high trans-sphincteric fistula and two-stage seton fistulotomy for two low trans-sphincteric fistulas

Discussion

The term 'multiple anal fistulas' was scarcely mentioned in the literature. Akiba *et al.* [12] mentioned multiple anal fistulas to be included within the category of complex anal fistulas, but with no further explanation.

Multiple cryptoglandular anal fistulas seem to be neglected in the literature. Parks' Classification [13] divides anal fistulas into four types: inter-sphincteric, trans-sphincteric, supra-sphincteric and extra-sphincteric. However, Parks *et al.* [13] overlooked multiple anal fistulas in the classification. In addition, the American Society of Colon and Rectal Surgeons did not mention multiple cryptoglandular fistulas in the published practice parameters for the management of perianal abscess and fistula-in-ano in 2016 [6].

Table 3 Postoperative Wexner score for patients included in the study

	Wexner score
After 3 months (n=12)	
Range	0–4
Mean	1.58
SD	1.01
Median	1.00
After 6 months (n=12)	
Range	0–3
Mean	0.75
SD	0.97
Median	0.50
After 12 months (n=10)[a]	
Range	0–2
Mean	0.30
SD	0.67
Median	0.00

[a]After exclusion of the recurrent cases.

The number of coexisting fistulas in our patients ranged from 2 to 5, with a mean of 2.58±0.90 fistulas. In the literature, multiple anal fistulas were usually attributed to specific causes, particularly Crohn's disease [14,15]. This may be explained by the fact that the inflammation in Crohn's disease tends to penetrate the whole thickness of the bowel wall. This creates ulcers, leaks, and abscesses. Consequently, they develop holes that can start to form, which then become tunnels, creating fistulas [16].

In contrast, it may be difficult to explain the pathogenesis of multiple cryptoglandular fistulas. In our study, 11 of the 12 (91.7%) patients had their fistulas located within one quadrant around the anus (within 3 consecutive O'clock locations). According to the cryptoglandular hypothesis, inter-sphincteric gland infection and abscess formation are the initiating events in the development of perianal fistulas [2]. The nearby location of the multiple fistulas in our patients may suggest a regional anatomical cause, such as a possible aggregation of anal glands that leads to the occurrence of infection in more than one anal crypt.

Duration of complaints in our study ranged from 6 to 18 months, with a mean of 11.25±3.77 months. Delay in the management of anal fistulas usually leads to the development of extensions from the existing infected gland with subsequent abscess formation and creation of complex anal fistula with multiple external openings but with the same internal opening [17]. In our study, six (50%) patients presented for the first time, complaining of more than one fistula, which implies that delay in the management may not be the cause of multiplicity. Moreover, five (41.7%) patients had

nondischarging anal fistulas at the time of presentation. We suggest that the susceptibility of these patients to develop cryptoglandular infection may be the cause of their condition.

Preoperative MRI was performed in eight (66.7%) patients in our study. Diagnosis of multiple anal fistulas was successfully described in all of them. The other four patients were diagnosed on a clinical basis preoperatively to be complex anal fistulas. Their fistulas were diagnosed to be low fistulas with nearby external openings. Surgeons preferred to go directly for examination under anesthesia that revealed multiple anal fistulas intraoperatively.

It is strongly recommended that MRI should be considered in patients with complex anal fistulas [6]. The use of MRI for the identification and classification of perianal fistulas can provide essential information with both diagnostic and prognostic values [18]. As experienced in our patients, the existence of external openings of anal fistulas in nearby positions can be commonly misdiagnosed as 'complex anal fistulas', rather than 'multiple anal fistulas'. Moreover, during surgery, some anal fistulas are minimally active and may be overlooked by the surgeon, which will be wrongly considered as recurrence postoperatively. Therefore, the MRI is of utmost importance for preoperative accurate diagnosis and identification of multiple anal fistulas.

In our study, all patients with multiple anal fistulas had inter-sphincteric and/or trans-sphincteric anal fistulas. Nevertheless, performing fistulotomy for all fistulas in a single operation may carry a high risk of incontinence. Fistulas that are high trans-sphincteric and those with anterior location have a more postoperative risk of incontinence if they underwent fistulotomy [7,8]. Therefore, staged management was performed for our patients to reduce the incidence of postoperative incontinence. Pujahari *et al.* [19] reported safe bilateral simultaneous internal sphincterotomy for their patients with anal fissure. Following the same principle, we found that fistulotomy of up to two inter-sphincteric and/or low trans-sphincteric fistulas can be carried out safely in the first operation without the high risk of postoperative incontinence. Seton was applied for the rest of the existing fistulas.

Despite the current technological advances nowadays, it is not possible to formulate a gold standard management algorithm for anal fistulas among the different surgical options [2]. Geltzeiler *et al.* [15] reported that fistulotomy is generally not recommended in patients with complex fistulas, as fistulotomy has an unacceptably high risk for incontinence. Caution should also be used in women with anterior fistulas, and in patients with short anal canals [1,2].

We agree with Geltzeiler *et al.* [15] as regards classic complex anal fistulas. However, we think that there should be a clear classification of multiple cryptoglandular anal fistulas, whether they should be considered as complex or not. In fact, plan of management of this uncommon condition is usually challenging, although it is not technically difficult compared with complex anal fistulas.According to Wexner score, our patients expressed acceptable continence status in the postoperative period. By the end of 12 months postoperatively, only two patients reported rare involuntary passage of gas. They had high trans-sphincteric anal fistulas. Management of high anal fistulas is usually difficult and is associated with higher risk of incontinence [8]. Although it was reported that cutting through up to two-third of the external anal sphincter is safe and has no significant effect on postoperative continence [20], no reports described the effect of sphincterotomy in more than one site of the external sphincter. Continence status usually improves with time after healing of the wound, provided that the muscles were only incised. It is suggested that evaluation of the final state of incontinence should be carried out after at least 6 months after the operation [5].

Postoperative anal incontinence after surgical management of anal fistulas had been reported in the literature in a wide range [8,21]. Likewise, recurrence rates after fistula management have also a very wide range of results between different reports [8,21,22]. On the basis of our findings, it is to be noted that, as each fistula has its own peculiar location and branching, results of anal fistula management may be confusing. In addition, other anatomical factors may affect the outcome of fistula management, for example, the length and tone of the anal sphincter. Moreover, the patient's characteristics (including age, diabetic state, and smoking status) lead to different immunity status and healing. The surgeon's experience has an undeniable role in determining the outcome of anal fistula management. All these variables can explain the wide range of incidence of postoperative complications after anal fistula management.

Limitation

Being a retrospective and single-center study may be the main disadvantage of this study.

Conclusion

The condition of multiple cryptoglandular anal fistula is an uncommon category of anal fistula. MRI is a perfect tool to diagnose the condition preoperatively. Combined fistulotomy and seton application seem a safe strategy for management of multiple anal fistulas with low postoperative recurrence and good postoperative continence. Further studies are required to explore more details about this neglected category of anal fistula.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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