

Pilonidal disease mimicking anterior anal fistula and associated with posterior anal fistula: a two-step surgery. Case report

C. EBERSPACHER, D. MASCAGNI, L. FRALLEONE, G. GRIMALDI, P. ANTYPAS, P. MASCAGNI, A. MATURO, F.M. DI MATTEO, S. PONTONE, D. PIRONI

SUMMARY: Pilonidal disease mimicking anterior anal fistula and associated with posterior anal fistula: a two-step surgery. Case report.

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Aim. Anal fistula is a common disease originated from abscess according to the cryptoglandular theory. A rare etiology is the pilonidal disease. In our case we observed a pilonidal disease mimicking an anterior perianal fistula, associated with another posterior anal fistula.

Case presentation. A 36-year old man was referred to our department with an anal fistula with an anterior opening. Despite the clinical examination and the endoanal ultrasound, only during the

surgery we discovered the origin of the anterior fistula from a misdiagnosed pilonidal sinus. There was also a posterior anal fistula in communication with the same abscess of the anterior one. We performed a two-step surgery with a first fistulectomy of the anterior tract, a drainage of abscess and the positioning of a seton for the posterior fistula. After about one month and the fall of the seton we evaluate the good healing of posterior anal fistula and excised the residual pilonidal sinus.

Conclusion. This misdiagnosed pilonidal disease created in our clinical report a true challenge. Our goal was to eliminate as much disease as possible, but also to avoid major complications or recurrences. We refused an aggressive approach and chose a two-step surgery, with in the first approach not only a demolitive time but also a reconstruction to facilitate healing, and in the second time the complete eradication of the pathology.

KEY WORDS: Pilonidal disease - Anal fistula - Surgery - Fistulectomy.

Introduction

Anal fistula is a common disease, whose development involves interaction of many histological, microbiological and molecular factors (1). Most of them are justified by the cryptoglandular theory: the first step is the perineal abscess, which evolves in fistula in anus, when a hollow tract connects an internal opening inside the anal canal to one or more external openings. Fistulas can also be a common complication of inflammatory bowel disease (IBD), radiation or in rare cases the first onset of malignancy (2, 3). In literature it is also reported another condition from which can arise an anal fistula: the pilonidal disease. This etiology occurs very infrequently, when the sinus reaches the

anal region and simulates an anal fistula (4-6). Almost all the cases have reported a posterior localization of the perianal opening (7).

We report a rare misdiagnosed pilonidal disease not only mimicking an anterior anal fistula but also associated with a posterior one. The surgical correction of this condition required a two-step procedure: the first consisted of the excision of anterior fistula tract, the drainage of the abscess, the curettage of the cavity and the positioning of a seton for the posterior fistula; during the second time an evaluation under anesthesia (EUA) for the posterior fistula was performed, with the evidence of a good healing after the fall of the seton, associated with the excision of residual pilonidal disease.

Case presentation

A 36-year old man with an anal fistula was re-

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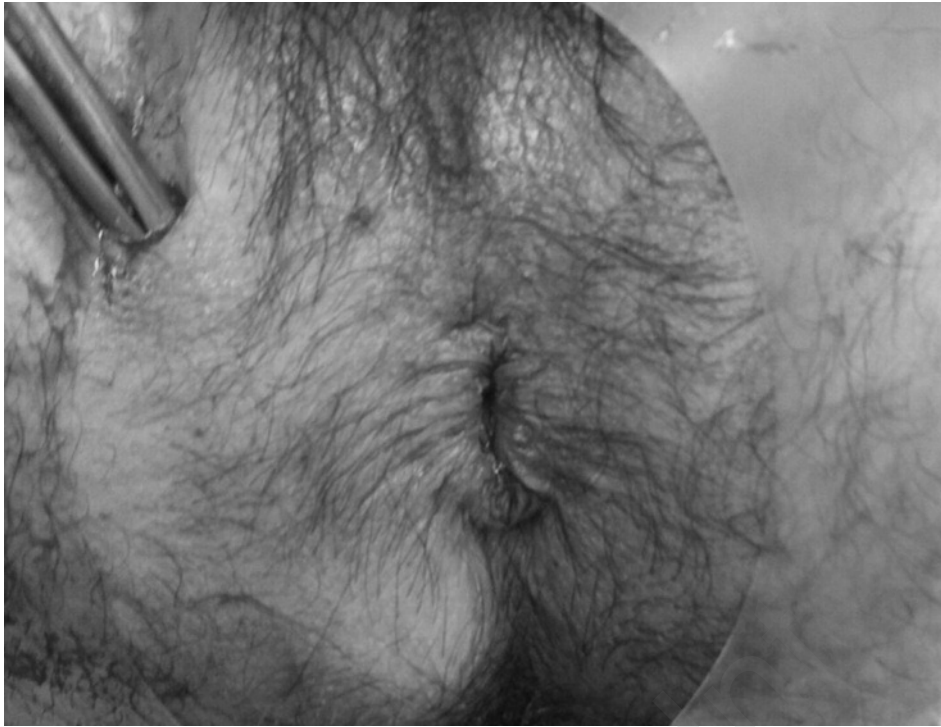


Figure 1 - The anterior external opening with a specillum shows no direct communication with the anal canal, but only with a posterior abscess cavity.

ferred to our Department for evaluation and management. His past medical history was unremarkable. His symptoms were pain and discomfort in perianal region. Clinical examination revealed a discharging opening in the anterior left quadrant, 2,5 cm from anus. It was not palpable a tract leading to the anal canal, but the patient had already been undergone an endorectal ultrasound, which apparently showed an anterior transphincteric fistula. Despite the huge amount of hair in the perianal area it was not identified any pilonidal disease during the inspection. The patient underwent surgical operation according our idea to perform a fistulectomy with sphincteroplasty and anoplasty. During the sampling of the anterior external opening with a specillum it was evident that there was not a direct communication with the anal canal, but only with a posterior abscess cavity (Figure 1). The anoscopic examination showed no primary anterior opening of the fistula as suggested by ultrasound, but a posterior fistula in communication with the same abscess previously described. We changed our plan and decided to perform a fistulectomy of the anterior tract of the fistula (Figure 2), to put a seton in the posterior transphincteric anal fistula, and to drain the abscess. In the cavity we discovered the presence of hair (Figure 3) and so the primary etiology of the disease: a pilonidal sinus misdiagnosed for the absence of evi-



Figure 2 - Fistulectomy of the anterior tract of the fistula.

dent orifices in the sacrococcygeal region. In order to avoid an aggressive surgical approach and a painful postoperative period for the patient we decided to postpone the excision of the sinus and to perform a reconstruction in the anterior region of the fistulectomy, to facilitate healing (Figure 4a, b). After forty days of medication and the fall of the seton we evaluated under anesthesia the good healing of posterior perianal fistula and we removed the

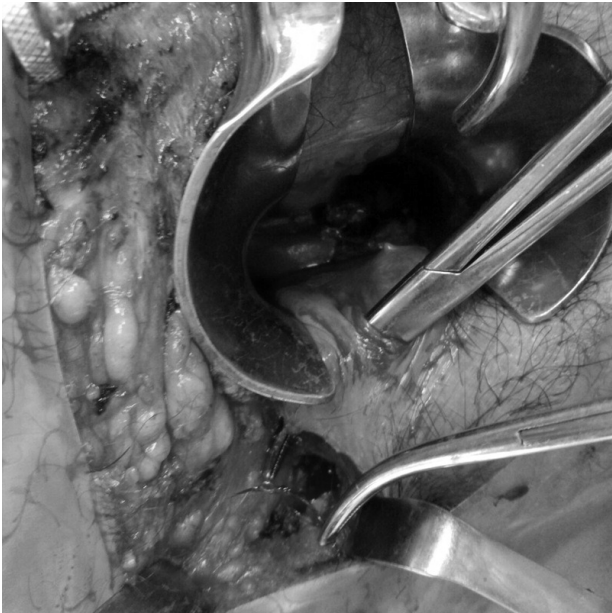


Figure 3 - Placement of a seton in the posterior transphincteric anal fistula and drainage of the abscess. Presence of hair in the cavity.

residual pilonidal disease in the sacrococcygeal region, with a primary closure and complete recovery in two weeks.

Discussion and conclusions

Pilonidal cysts typically present with abscess or recurrent pain and drainage of the sacrococcygeal region. Physical examination usually reveals pits in the midline of this area. In many cases the correct diagnosis of anal fistula is simple: there is an orifice near the anal canal, associated with pain and swelling and often a palpable tract leading from the secondary opening to the anus. Various treatment strategies for managing pilonidal disease have been employed. It is recommended to initially approach pilonidal disease with less-extensive procedures, reserving more complicated approaches for patients who develop chronic pilonidal disease. What seems obvious after the first clinical examination can become totally dif-

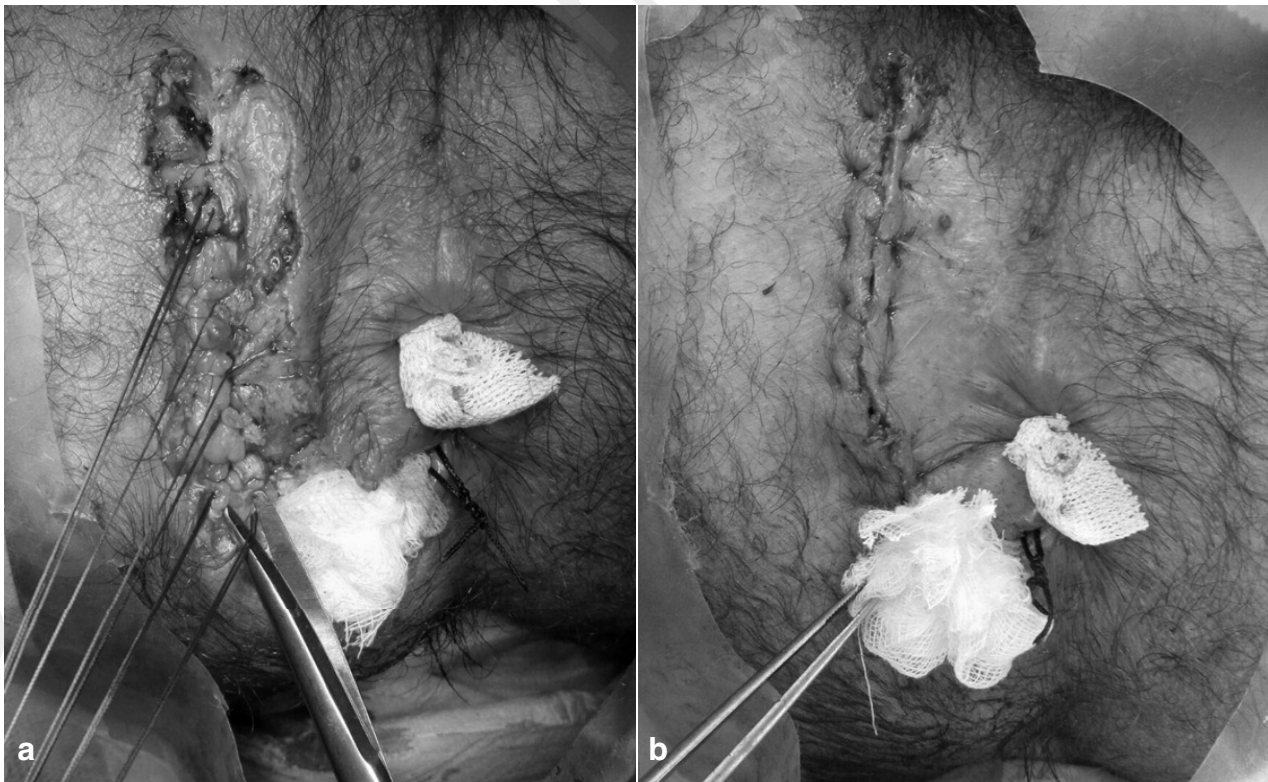


Figure 4 - Reconstruction in the anterior region of the fistulectomy to facilitate healing.

ferent during surgery (8). Chronic pilonidal disease is associated with sinus tract formation which is almost exclusively limited to the sacrococcygeal region. The principals of management in this setting require excision of the sinuses and associated tracts. This can be done through incision and curettage of the tracts with or without marsupialization with recurrence rates of up to 19%. Wide local excision with primary closure is usually associated with more wound complications and reported recurrence rates of 11–29% (9). Karydakakis modified radical excision by proposing a curvilinear incision lateral to the midline with the creation of a gluteal flap and debridement of the sinus tracts. This allows creation of a shallower cleft and keeps the wound off of the midline. Reported recurrence rates with this technique are less than 5%. More extensive reconstruction procedures have been described as well including the rhomboid flap, V-Y flaps, and Z-plasty. These techniques are typically reserved for patients with multiple recurrences or nonhealing wounds (9–11).

Pilonidal disease with fistula and drainage around the anus represents an atypical presentation that is more likely to raise a suspicion of fistula-in-ano rather than pilonidal disease. Four cases were initially reported in 1948 (12).

In our report a misdiagnosed pilonidal disease was the extra anal source of a left anterior fistula, but it was also in communication through a common abscess with a posterior perianal fistula. The presence of hair in cavity clarified the origin of the anterior fistula tract. Likely in all the perianal disease our challenge was to remove all the disease minimizing complications and above all the possibility of recurrence. So that in the first time we decided to perform only the surgical toilette and the removal of the fistula tract, plus the positioning of the seton, and in the second time we checked healing of the posterior fistula and removed the sinus. In the period between the two surgical steps the good reconstruction (13) performed during the first operation reduced pain and discharge of the patient, who returned to work after ten days and came for check-up and medication once a week.

Usually the coexistence of anal fistula and pilonidal disease, even if possible, it is uncommon; the two process are often limited to their respective locations. Only sometimes pilonidal disease can produce an abscess which reaches anus, mimicking an anterior anal fistula. In our case report we observed this phenomenon associated with the presence of a posterior anal fistula, with a common abscess's cavi-

ty. The treatment can be complex (14), but we recommend to avoid any aggressive surgical approach. Our goal was to remove as much disease as possible minimizing complications or recurrences, so we decided a two-step surgical approach as the more appropriate choice.

Declaration

All Authors have read and approved the final manuscript. The Authors have no conflict of interests.

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