

Using a Limberg Flap to Treat Recurrent Pilonidal Disease: A Case Report with a Mini-Review

Nikolaos-Andreas Anastasopoulos^{1*} and Chamzin Alexander²¹First Propaedeutic Department of Surgery, National and Kapodistrian University of Athens, Greece²Department of Surgery, "Hippokrateion" General Hospital of Athens, Greece***Corresponding Author:** Nikolaos-Andreas Anastasopoulos, First Propaedeutic Department of Surgery, National and Kapodistrian University of Athens, Greece.**Received:** April 17, 2020**Published:** April 30, 2020© All rights are reserved by **Nikolaos-Andreas Anastasopoulos and Chamzin Alexander.****Abstract**

Pilonidal disease is a common acquired condition due to the presence of loose hair in the natal cleft. Typically, it presents in younger male patients, and there is an assortment of therapeutic options, with a variable recurrence rate. We present the case of a 32-year old patient with recurrent pilonidal sinuses and abscesses, that came to our outpatient clinic for definitive treatment of his coccygeal disease. We performed a wide surgical excision with a Limberg flap reconstruction, and a negative pressure wound drain. The patient was discharged on the second postoperative pain, with minimal pain and the drain was removed three days after discharge. Sutures were removed on the twentieth postoperative day and follow-up was carried out once weekly for a monthly period. No wound dehiscence or flap necrosis were observed during the immediate follow-up period with a recurrence-free satisfactory aesthetic result, one year after the operation. In an era of numerous available techniques for treating pilonidal disease, the Limberg flap remains an easily reproducible and highly safe technique, even for surgeons with little or no experience regarding flap reconstruction surgery.

Keywords: Pilonidal Disease; Recurrence; Definitive Therapy; Limberg Flap**Abbreviations**

Bid: Twice Daily; EPSiT: Endoscopic Pilonidal Sinus Treatment

Introduction

Pilonidal disease represents a frequent complaint for visiting the outpatient surgical clinics, reaching an incidence of 26 per 100,000 annually, affecting predominantly male teenagers and young adults [1]. Clinical manifestations of disease vary widely with acute-abscess with or without cellulitis, chronic-single or multiple sinuses with various degrees of local inflammation and recurrent presentation. Patients mainly complain of coccygeal pain, purulent discharge, and pruritus [1,2].

Several studies have identified risk factors for disease progression or recurrence after therapy, including family history, smoking, obesity, poorly controlled diabetes, and hirsutism. Novel studies, however, question the role of these risk factors in pilonidal disease natural course. Specifically, the findings of a large German military study, suggest that smoking and obesity do neither impede wound healing nor increase recurrence after excision and primary midline closure [3].

Although the pathophysiology of disease is not yet clearly elucidated, the theory of congenital defective natal cleft closure has been abandoned and the early working theory of Bascom has been adopted, suggesting that a stretched follicle with straight hair becomes embedded under the dermis, collecting debris and keratin, triggering chronic inflammation with foreign body giant cells. This subdermal cystic collection may lead to abscess formation after getting infected by local bacterial flora and form sinuses [4].

In this article, we present the case of a 32-year old male that, having received wide local excision and Limberg flap reconstruction for recurrent pilonidal disease.

CasePresentation

32-year old male patient presented to the General Surgery Outpatient Clinic of the "Hippokrateion" General Hospital of Athens in Greece. The patient complained of severe recurrent pilonidal disease with purulent discharge from sinus exits. Upon examination, apart from the primary sinus tract, a secondary was revealed on the left side of the midline gluteal cleft. Oral amoxicillin/clavulanate was prescribed bid for seven days and a

wide local excision with flap reconstruction was planned. Patient's history was notable for previous pilonidal disease excision and healing by secondary purpose. He also referred smoking (10 pack-years). No drug allergies or systemic medication was referred in patient's history.

On the day of the operation, preoperative shaving of the area was performed, one dose of both intravenous cefoxitin and metronidazole were administered, and the operative field was sterilized using both povidone iodine solution and scrub. A Limberg (or rhomboid) flap was drawn, using permanent skin marker (Figure 1), containing the primary sinus tract and the secondary sinus. Iodine povidone solution was administered, using a peripheral venous catheter, through the primary sinus (Figure 2). Sharp dissection, with scalpel and electrocautery, was used to conduct the wide local excision, in a rhomboid shape, and to undermine the rotational flap, to the level of the presacral fascia. After fully mobilizing the flap and undermining wound edges (Figure 3 and 4), the flap was sutured in place using several Polyglactin 910 simple interrupted subcutaneous sutures, placed in the wound corners, and a negative pressure wound drain was inserted. Skin was closed using interrupted vertical mattress monofilament nylon sutures (Figure 5). Notably, the procedure lasted approximately two hours. The patient showed minimal postoperative pain, well controlled with intravenous paracetamol, as needed. Oral amoxicillin/clavulanate was administered until the seventh post-operative day. The patient was discharged on the second postoperative day with minimal serosanguineous exudate in the drain, which was removed three days after discharge. The patient was advised to avoid placing strain on the flap and a sodium hyaluronate/silver sulfadiazine cream was used during wound dressing changes. Sutures were removed on the twentieth post-operative day (Figure 6) and the patient was again examined twice, one on the thirtieth post-operative day, and a second one, one-year post-operative. No local complications were recorded and there was a satisfactory cosmetic result.

Discussion

Management of pilonidal disease can be divided in two categories: non-operative versus operative. Non-operative management involves hair removal - by shaving or laser epilation, phenol or fibrin glue application and local or systemic antibiotics. These methods require no hospitalization, cause less pain and can control mild disease effectively, or provide and adjunctive to operative management, but higher recurrence rates occur when they are applied. The American Society of Colon and Rectal Surgery guidelines suggest no need for perioperative antibiotics in pilonidal disease. However, a single study has shown lower recurrence rates when

Figure 1: Designing a Limberg flap using a permanent marker. Our technique involves a flap with the inferior pole in the intergluteal cleft.

Figure 2: Iodine povidone solution was administered through a peripheral venous catheter in the primary sinus.

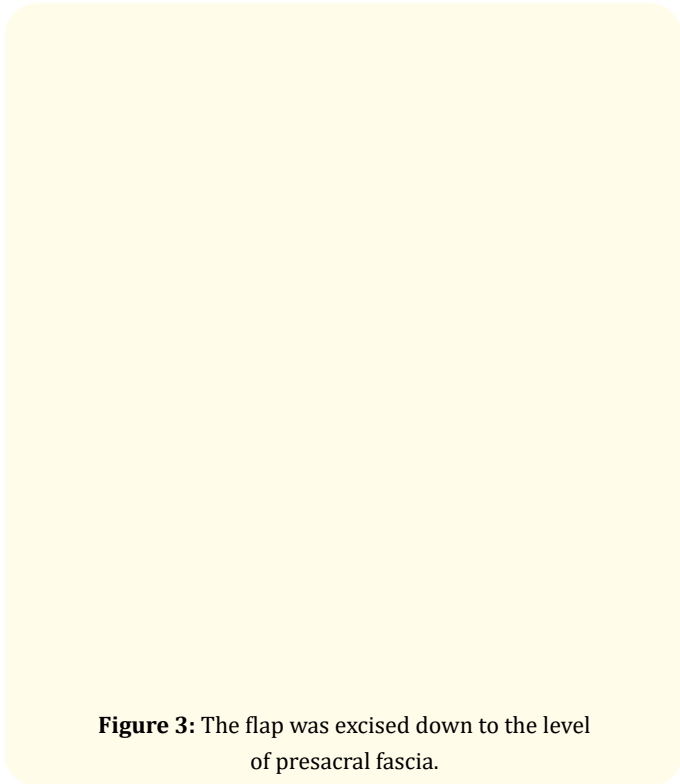


Figure 3: The flap was excised down to the level of presacral fascia.

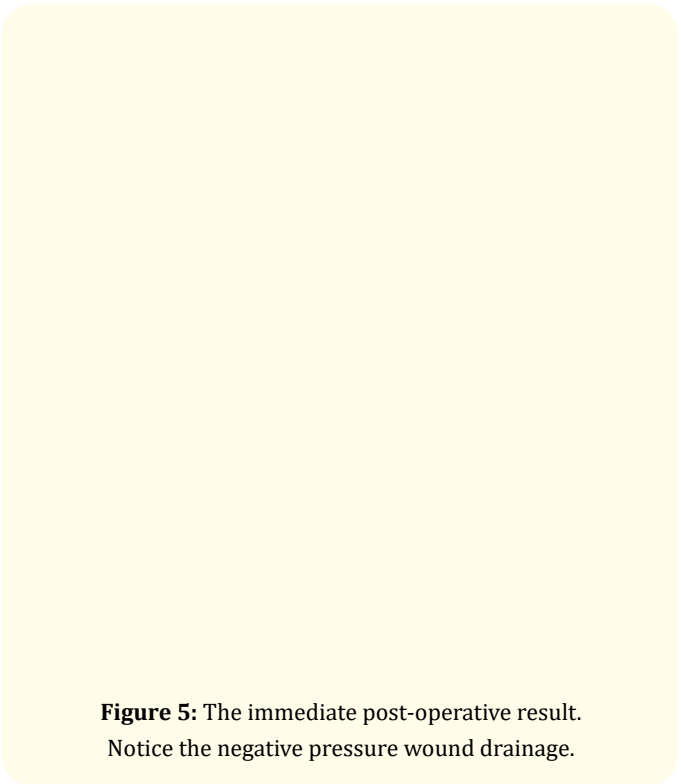


Figure 5: The immediate post-operative result. Notice the negative pressure wound drainage.



Figure 4: The fully mobilized rhomboid flap.

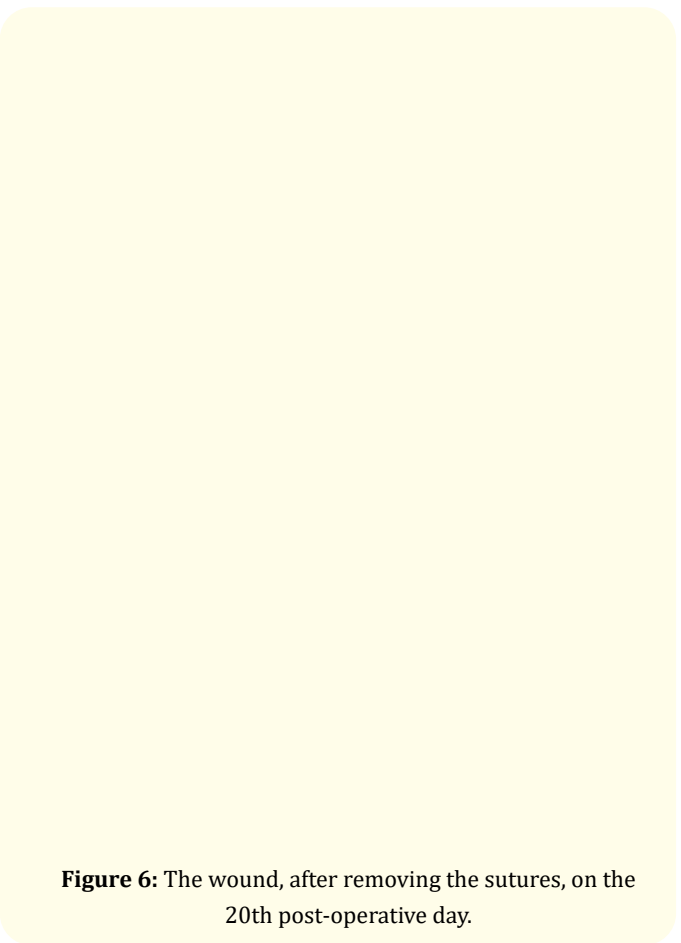


Figure 6: The wound, after removing the sutures, on the 20th post-operative day.

one dose of preoperative metronidazole with or without cefuroxime plus five days of amoxicillin/clavulanate are administered [2].

The armamentarium of the general surgeon treating pilonidal disease includes several techniques. Choosing the appropriate method is largely dependent on the timing of procedure. Acute disease is often managed with incision and drainage of the abscess, which is associated with a 15 - 40% recurrence rate. These patients will require a second definitive procedure. Local excision and secondary healing remain surprisingly popular among surgeons, leading to a prolonged healing time, from one up to three months, and delayed return to activities, with recurrence rate fluctuating among studies from 2% to 35% [2].

Primary midline closure reduces significantly healing time. Nevertheless, wound dehiscence and infection rates are significantly higher in comparison with off-midline closure techniques in an analysis performed by Petersen and colleagues [5]. On the contrary, a prospective cohort analysis of a modified Lord-Millar midline closure versus other techniques yielded different results. Patients treated with the modified Lord-Millar midline closure required less hospitalization, fewer days abstaining from work and reduced postoperative analgesia, indicating rapid wound healing [6].

Off-midline flap reconstruction techniques present a major advantage, that is, levelling of the intergluteal cleft, and diminishing a local factor of recurrence. Several techniques have been employed for reconstruction after wide excision, such as the Karydakias flap, the Limberg flap, the Bascom flap, the V-Y advancement flap, and others. Dr. George Karydakias pioneered in pilonidal disease surgery, proposing a technique of an elliptical excision of the affected area with suturing of the deep flap - after undermining - onto the presacral fascia. His personal series included 7471 procedures with less than 8% wound complication and 2% recurrence rates [7]. Compared with secondary healing after wide excision, Karydakias flap demonstrated superior results, yet higher wound complication and recurrence rates than the original series. A systematic review comparing Karydakias with Limberg flap revealed no significant differences concerning wound infection or dehiscence, haematoma, recurrence, length of hospital stay and patient satisfaction. Modest differences were observed in operative time and seroma formation. However, the impact of operative time difference - which is seven minutes - is of minor clinical importance and no difference was shown between the two groups in seroma formation, after subgroup analysis. Of note, this is the only systematic review, to our knowledge, comparing the two types of flap reconstruction, and it is characterised by high heterogeneity rates [8].

The Limberg or rhomboid flap reconstruction, which we used in the current case, displays several advantages. This transposition flap has multiple applications in surgery and was initially designed by Dr Alexander Limberg in 1948. The initial flap had a rhomboid shape, with its opposite sides equal in length and a pair of 60 degrees and a pair of 120 degrees angles. This design minimized tension produced while mobilizing the flap to its new position. The Limberg flap, and its modifications, have been widely used, as they are associated with extremely low recurrence rates (less than 5% in most studies), minimal complications, and good patient tolerance. It is suitable for treating recurrent disease with multiple sinuses, as it allows a wide excision area [1,2,8]. Moreover, in a recent case series the Limberg flap has been effectively used to treat acute pilonidal disease. The authors presented similar results using the rhomboid flap for both acute and chronic disease, while the need for a larger excision was underlined when treating patients with acute disease [9].

In the modified rhomboid flap, described by Mentés, *et al.* the inferior pole of the flap is relocated 2 cm laterally to the midline, reducing rate of recurrence [10]. A randomized controlled trial showed low recurrence rates, when both the initial and modified Limberg technique were used to treat recurrent disease. Of note, in the group of the initial technique, a higher prevalence of macerations in the inferior pole of the flap and infection was observed, leading to delay in both suture removal and return to workplace [11]. Another factor with a mildly beneficial impact is drain placing, as proven in several case series, in which decreased wound fluid collections and faster healing time were observed [2].

Interestingly, a multicentre evaluation of 228 consecutive patients that received treatment with a presacral perforator flap yielded very satisfactory results. The flap procurement was aided by acoustic Doppler. This technique resulted in reduced operative time, low complications' rate, a highly satisfactory aesthetic result, and a recurrence rate of approximately 1% in a median follow-up of 28 months. Technical complexity consists an obvious drawback of this technique [12].

Several minimally invasive procedures have been recently introduced in the management of pilonidal disease. Endoscopic Pilonidal Sinus Treatment (EPSiT) was firstly described in 2014, in which after excision of the external opening of the sinus tract, a fistuloscope is inserted in order to remove excess hair, rinse the sinus, and cauterise it using monopolar diathermy. Apart from nullifying the need for general anaesthesia, this technique warrants less postoperative pain, return at work on the same day of the procedure and better long-term cosmesis [13]. Its results in recurrent disease are

remarkable, displayed in a series of 122 patients with recurrent disease, in which 95% complete wound healing was observed in a mean time of 29 days [14]. However, in a setting of complicated disease, even if EPSiT yielded less complications, it showed higher recurrence rate, when compared with the Limberg flap [15]. Another, minimally invasive procedure, which has been compared with the Bascom technique in a single centre randomised prospective clinical trial, is the Video-Assisted Ablation of Pilonidal Sinus (VAAPS). This trial showed superior results in terms of early mobilization, wound infection rate, postoperative pain and patient satisfaction in the VAAPS group [16].

Conclusion

Given the fact that pilonidal disease is affecting young productive people in a persistent manner, an "ideal" therapeutic modality would guarantee rapid wound healing, low rate of complications and recurrence and minimal pain. All these parameters lead to fewer days off work and abstaining from any physical activities, thus improving quality of life. There are several options for surgeons operating on pilonidal disease, and operative management should be guided by several factors, such as timing and severity of presentation, recurrence history, patient's medical history and social profile, as well as surgeon's expertise. The lack of a single best operative technique in conjunction with the variability in recurrence rate reports suggest that we are far from identifying the "ideal" treatment modality for pilonidal disease. Flap reconstruction techniques, however time-consuming might be, present as a safe option for patients with disease recurrence. It should be noted that attempting these techniques requires that the surgeon be familiar, and the patient be an appropriate candidate. Novel endoscopic treatment methods are quite promising in all aspects, but more extensive data need to be published in order to establish recurrence rates and cost-effectiveness.

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Conflict of Interest

We declare no conflict of interest.

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