



## Pilonidal Sinus Disease - A Literature Review

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### Abstract

Pilonidal Sinus Disease (PSD) is a common condition that has had controversies surrounding its aetiology and treatment since its first description in the mid-19<sup>th</sup> century. The prevalence in the UK has been estimated at 0.7% with peak age of incidence at 16 years to 25 years. Males are more commonly affected than females and risk factors include stiff body hair, obesity, and a bathing habit of less than two times a week, and a sedentary occupation or lifestyle (i.e. those who sit for more than six hours a day). Pilonidal sinus disease is best managed by specialists with an interest in the disease such as a colorectal or plastic surgeon experienced in treating recurrent cases. Emergency treatment should primarily consist of off-midline incision and drainage with subsequent referral to a specialist should the condition recur. The aim of this article is to summarise the current practice for treatment of pilonidal sinus disease including difficult modalities used and their limitations.

### Introduction

Pilonidal Sinus Disease (PSD) was previously referred to as Jeep disease when it was noticed amongst American soldiers driving the eponymous vehicles in World War II [1]. It is a common condition that has had controversies surrounding its aetiology and treatment since its first description in the mid-19<sup>th</sup> century [2]. Due to its high recurrence rate, PSD has previously been ascribed to a congenital origin such as a caudal remnant of the neural tube or sequestered ectodermal tissue during development [3,4].

The current accepted aetiology is that PSD is an acquired pathology with multiple contributing factors [5]. Hairs, either loose or in the skin are thought to grow inwards due to a combination of local forces and friction acting on the topography of the natal cleft which cause an inflammatory reaction [6]. It is unknown whether hairs (either loose or native to the region) are the primary cause of PSD or whether hair follicles become affected by the physical environment which leads to micro-abscesses and PSD [7].

### Epidemiology

The prevalence in the UK has been estimated at 0.7% with peak age of incidence at 16 to 25 years [8]. PSD does affect children as shown by an American case series of 120 patients with a mean age of 14.9 years (range 1 to 19) [9]. Exact incidence and prevalence figures for the UK have not been calculated and previous estimates are historical or not applicable to the general populace. For example, a prevalence of 4.6% to 8.3% (clinically apparent to silent PSD) was found in a Turkish population [10]. With regards to incidence, it has previously been estimated at 26 per 100,000 in a Norwegian population more than 20 years ago [11]. A more recent estimate states an incidence range of 0.11% in women at college to 8.8% in Turkish soldiers [12]. What is known is that males are more commonly affected than females and risk factors include stiff body hair, obesity, a bathing habit of less than two times a week, and a sedentary occupation or lifestyle (i.e. those who sit for more than six hours a day) [13].

Long term recurrence in PSD has been estimated at 22% with the majority occurring in the first year but recurrences 20 years after treatment has been reported [14]. Recurrence rates should be tracked over a minimum of 5 years as a long term study of PSD in a German cohort found that 60% of recurrences occurred within that time frame [15].

Risk factors for recurrence include black ethnicity, young age at presentation, established recurrent disease, and a family history of PSD, sinus discharge on physical examination, post-operative hair shaving and post-operative Surgical Site Infection (SSI) [15-17]. Counter-intuitively, a German military cohort study found that a high Body Mass Index (i.e. 25 and higher) and smoking were not risk factors for recurrence [16,18]. However, Incision and Drainage (I&D) of PSD abscesses before definitive surgery and use of methylene blue in definitive surgery were found to reduce the

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risk of recurrence [15].

## Signs and Symptoms

Half of all PSD patients present as an emergency with an abscess in the natal cleft but implantation of hairs can theoretically occur anywhere on the body [19]. One well known (but little seen) example affects men's barbers who develop sinuses in the web spaces between fingers (interdigital PSD) [20,21]. Other reported sites include the umbilicus, chest wall, axilla and scalp [22]. PSD abscesses most commonly present in the midline of the natal cleft, occasionally with the abscess pointing or protruding laterally [23].

Patients with chronic or recurrent PSD have a range of presentations from a simple cyst to multiple pits in the midline of the natal cleft [24]. Occasionally there is no visible opening but other times, hairs may be extruded from primary or secondary openings [25]. There is no widespread or recognized system of classification for PSD [26], however we propose a pragmatic system to consider the disease entity in terms of severity:

Mild - Sinus disease (which may be multiple)

Moderate - Presence of fistulae (which may be multiple)

Severe - Cicatrization or involvement of tissue outside the natal cleft

## Differential Diagnosis

Few other conditions mimic the clinical picture but occasionally, other skin conditions may cause midline skin pits such as hidradenitis suppurativa, pyoderma gangrenosum, syphilis or tuberculosis [27]. Pathology from nearby structures may also be the underlying cause such as Crohn's anal fistulae or a congenital presacral sinus. Cancer may arise from within long standing inflamed PSD tissue but this is rare and is estimated to involve 0.1% of chronic PSD patients [28]. This is usually a squamous cell cancer akin to the pathogenesis of a Marjolin's ulcer where longstanding inflammation triggers carcinogenesis [29,30], however, other cancers such as basal cell carcinoma and adenocarcinoma have also been found in PSD [31].

## Management

The management of PSD can be divided into emergency and elective settings.

### Emergency

The treatment of PSD abscesses is straightforward with I&D over the pointing lesion [32]. However, in a retrospective case series, an off-midline approach has been reported as healing approximately 3 weeks quicker than incisions over the midline [33]. PSD patients who only present with cellulitis may be treated with antibiotics but there is no strong evidence that this will arrest abscess formation [22,28]. An improvement on the I&D technique has been reported by Khalil et al. [34] who showed that aspiration of the PSD abscess followed by injection of a local anaesthetic and then incision allowed acute PSD to be treated without a general anaesthetic [34].

### Elective

Treating PSD in an elective setting is a more contentious issue. The mainstay of treatment has been removal of diseased tissue via surgery but the defining variables have been the amount of tissue excised and the method of closing the defect.

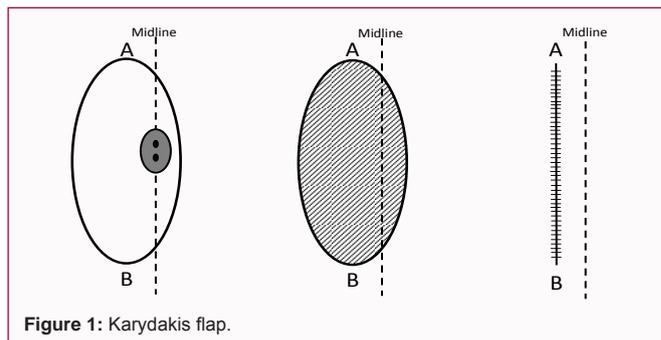


Figure 1: Karydakis flap.

### Limited vs. wide excision

Excision of all diseased tissue is the most commonly performed procedure in the UK according to a snapshot audit done in 2011 [8]. However, the removal of all affected tissue necessitates closure of large defects and does not guarantee cure. The most established less invasive technique of treating PSD is the simple Bascom's procedure which involves removal and sutured closure of midline hair pits along with drainage and curettage of the underlying abscess cavity via a lateral incision [35].

Another limited excision techniques is de-roofing and curettage of pilonidal sinuses (rather than excision) which was the subject of a recent systematic review in 2016. This review included 13 studies (n=1145) and showed that the technique is effective with a recurrence rate of 4.47%, a low complication rate and the additional advantage of being done under local anaesthetic [36]. The authors postulated that the results of their review suggested that this technique should be first line for all types of PSD cases both acute and chronic. A relatively new procedure- sinusectomy-has also been reported which involves excision of PSD sinuses alone identified by methylene blue. It can be performed under local anaesthetic, has a relatively low morbidity and a recurrence rate of 7% [37].

These two approaches to PSD however, have been compared in part of a recent meta-analysis which included 4 randomized controlled trials (n=153) and found that there was no significant difference in time to healing and recurrence rate. Limited excision was found to have an earlier return to work and less pain than wide excision. However, it must be noted that in this meta-analysis, sinusectomy was combined with sinotomy (i.e. unroofing) and that these two techniques were compared against wide excision left to heal by secondary intention (i.e. not primary closure). Furthermore, the authors warn that none of these trials were registered and were of poor quality [38].

### Secondary vs. primary closure

After excision of affected tissue, the next decision for the surgeon concerns whether to close the defect primarily or to allow healing to occur by secondary intention. This has recently been the subject of several systematic reviews. The earliest review in 2008 (and updated in 2010) concluded that although no clear benefit was found in primary closure versus secondary intention healing, a clear benefit in terms of healing time and recurrence rate was shown in off-midline rather than midline wound closure [39].

A further meta-analysis in the same year however, showed that in primary closure versus secondary intention healing, the former heals more quickly but at the expense of increased recurrence [40]. The authors also found that where primary closure was performed, an off-midline closure had lowered surgical site infection and recurrence

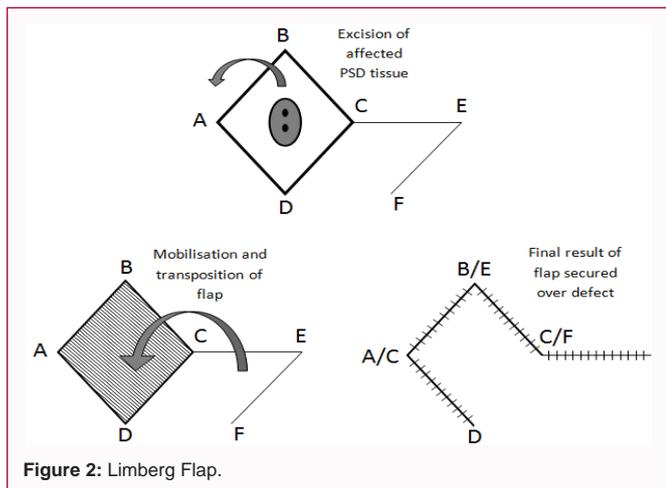


Figure 2: Limberg Flap.

rate [40]. The latest meta-analysis by Enrique-Navascues et al. [38] on different techniques of primary closure identified ten trials which showed that off-midline closure had significantly lower rates of dehiscence and wound infection [38]. The argument for off-midline closure has been strengthened by a large retrospective review of 569 patients which compared midline closure against asymmetric closure that crosses the midline (as compared to a Karydakias flap where closure is entirely off the midline) [41]. This study by Limongelli et al. [41] showed a significantly lower recurrence rate in the asymmetric closure group.

### Flap closure

In the UK, the most common flap technique for closing defects after PSD excision was the Karydakias flap (18%) followed by the Limberg flap (5%) [8]. The Karydakias flap consists of a para-median elliptical incision incorporating affected PSD tissue with the final suture line lateral to the midline (see Figure 1) [42]. The Limberg flap (named after its early 20<sup>th</sup> century inventor-Prof. Aleksandr Limberg of Leningrad) is a rhomboid transposition flap (Figure 2) was first published as a treatment for PSD by Azab et al. [43]. It has been used in PSD patients with reported lower recurrence rates and shorter hospital stays compared to primary closure [44,45]. Variations of the Limberg flap have been described such as the modified Limberg flap (lateralisation of the inferior apex) to flatten the natal cleft [46], a superiorly based Limberg flap or an oval head rhomboid flap to improve viability [47,48]. A meta-analysis comparing primary closure and the Limberg flap in 2010 found significantly lower rates of dehiscence, infection and recurrence for the latter and concluded that the Limberg flap should be used for elective treatment of primary PSD [49]. That being said, further trials not included in that meta-analysis have had varying conclusion.

Muzi et al. [50] found no difference in recurrence rate or wound dehiscence but did find that primary closure had a higher rate of wound infection and a lower reported post-operative pain compared with the Limberg flap [50]. Tavassoli et al. [51] did not show any significant difference in early complication or recurrence rate but did show that post-operative pain, patient satisfaction and return to work was improved with the Limberg Flap [51]. Okuş et al. [52] did not find any difference between the two techniques in terms of wound infection or recurrence rate [52]. Elshazly et al. [53] compared the modified Limberg flap against primary closure and had similar findings and in addition, that the modified Limberg flap patients had fewer post-operative complications and a lower rate of recurrence

[53]. Khan et al. [54] utilized the Limberg flap and showed that the technique had better outcomes overall when compared against primary closure [54].

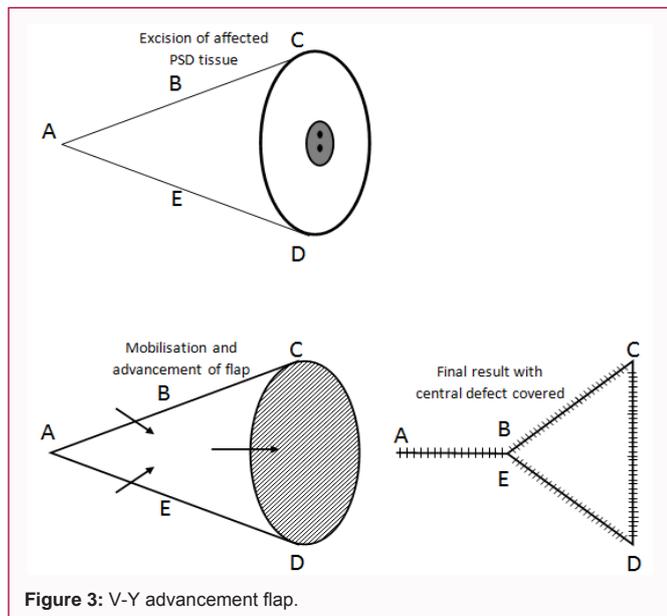
The two flaps have been compared against each other with varying results. A prospective randomized controlled trial compared the Limberg flap with the Karydakias flap and showed that the Limberg flap had a lower complication rate, a shorter length of hospital stay but there was no difference in recurrence rate [55]. A multicentre prospective randomized controlled trial in 2010 compared the modified Limberg flap against the Karydakias flap which showed that both techniques had similar outcomes except with laterally situated openings/pits where the Karydakias flap had limited applicability [56]. However, Karaca et al. [57] retrospectively compared the modified Limberg flap (n=35) with the modified Karydakias flap (n=46) modified in that the base of the asymmetric flap is not fixed to the sacrococcygeal fascia as described by Bessa et al. [58]. The modified Limberg flap group reported less pain, had lower complication rates, recurrence rates and higher patient satisfaction scores [57,58]. In terms of reports in the literature, the V-Y advancement flap (Figure 3) seems to be the next most common flap technique after the Limberg flap and is a third option in closing defects after surgical excision of PSD. This technique was previously used to close sacral pressure sores and offers the advantage of flattening the deep natal cleft groove [59].

A randomized controlled trial comparing V-Y advancement flap over primary closure did not show any significant advantage in terms of SSI, dehiscence or recurrence. In this series by Nursal et al. [17], logistic regression analysis showed that post-operative SSI along with younger age; recurrent disease and discharge on physical exam were independent predictors of recurrence [17]. The Limberg flap has been compared with the V-Y advancement flap in two retrospective studies with differing results. Unalp et al. [60] found that outcomes were not significantly different for either technique except that patients with the Limberg flap had a significantly lower recurrence rate [60]. Öz et al. [61] however, found that hospital stay and time off work were significantly shorter with the Limberg flap [61]. Other outcomes such as post-operative complications and recurrence rate were similar and the authors recommended that whilst the Limberg flap is of use in recurrent PSD, the V-Y advancement flap should be used preferentially in larger defects that need to be closed. Other flap based but less commonly used options include a parasacral perforator based flap elliptical island flap [62], a bilobed fasciocutaneous flap [63], an elliptical rotation flap [64], or a lateral advancement flap with Burrow's triangles [65].

### Adjuncts to surgery

Post-operative infection is a concern in the surgical treatment of PSD but it has not been suggested as a risk factor for recurrence. The use of antibiotics as an adjunct to surgery to reduce Surgical Site Infection (SSI) was the subject of a systematic review which reviewed 12 suitable trials involving 1,172 patients. The review suggested that there was no benefit with single dose prophylactic or long course antibiotics in promoting healing, reducing SSI or reducing recurrence rates [66]. The authors did caution that the heterogeneity in the included studies did not allow a meta-analysis to be performed and that higher grade evidence was needed to come to a firm conclusion.

Tritapepe and Di Padova reported good results with antiseptic flushing and drainage after excision and primary closure for PSD [67]. Their minimum follow-up period was 5 years and they reported no recurrences. However, their study was a case series and did not



specify the severity of PSD in their patients. The use of drains after excision and primary closure for PSD has been reviewed by a meta-analysis which included 8 studies involving 1,252 patients [68]. It found that drainage offered no significant improvement over any drainage in terms of post-operative infection or recurrence rates although the authors did acknowledge that drains did have a trend towards improvement.

A systematic review on Negative Pressure Wound Therapy (NPWT) identified only 5 suitable studies which suggest that NPWT is a viable adjunct to surgery for chronic PSD (the studies followed patients who had wide local excision) [69]. Although there were minimal complications in all studies (mainly wound separation), the lack of randomized controlled trials hamper recommendation of NPWT in closing wounds following surgery for chronic PSD.

#### Less well-established treatments

The use of fibrin in PSD has been the subject of two systematic reviews-although the earlier review in 2012 suggested better healing times than other therapies, the more recent review in 2016 showed no benefit in preventing seroma formation after surgery or reduced recurrence rates after sealing sinus tracts [70,71].

Phenol is an alternate treatment option where it is injected directly into PSD sinuses or introduced with the aid of a small incision which ablates the diseased tissue. This technique was the subject of a systematic review by Kayaalp and Aydin who concluded that despite its low morbidity, there was insufficient high grade evidence to recommend its use in place of surgery in PSD [72]. A later study in 2015 comparing phenol (n=44) with the modified Limberg flap (n=37) found no significant difference in SSI or recurrence rate but this was a retrospective trial and thus not randomized or controlled trial [73].

In a small randomized trial of 30 patients, radiofrequency sinus excision was compared against excision and marsupialisation for PSD with the reported advantage that the radiofrequency technique causes less morbidity and post-operative pain [74]. Radiofrequency excision thus seems a feasible option for PSD and the report hypothesized that the lower temperatures generated by RF (as compared to

electrocautery) allow for greater accuracy, less damage to surrounding tissues and thus less morbidity. A minimally invasive technique for PSD has been reported with the use of a fistuloscope and destruction of the fistula tracts under direct vision [75]. After removal of hair and debris, electrocautery is directly applied to the tract followed by debridement and irrigation. A case series of 27 patients with this technique showed only 1 recurrence after a year of follow-up but the report failed to describe the severity of disease or whether patients had been previously treated for PSD [76].

Other less invasive treatment options include cryosurgery combined with incision of PSD sinuses which has the advantages of needing only local rather than general anaesthetic, improved healing rates, lack of haemorrhage and simplicity of use [77]. Laser depilation has also been described to treat recurrent disease and has been postulated to work by removing the hairs which gather in the natal cleft causing epithelial disruption and an inflammatory reaction [78,79].

## Conclusion

The difficulty in interpreting studies on PSD has been the lack of an agreed or widely used classification or grading system for the severity of the condition. This has repeatedly been acknowledged in meta-analyses or systematic reviews concerning PSD [80]. A staging system with corresponding treatment recommendation has been recently proposed [81]. Whilst this is a step in the right direction, the problem is that such a system would have to be widely adopted, recognized in order to facilitate research, communication and meaningful comparisons amongst the many treatment modalities. Pilonidal sinus disease is best managed by specialists with an interest in the disease such as a colorectal or plastic surgeon experienced in treating recurrent cases. Emergency treatment should primarily consist of off-midline incision and drainage with subsequent referral to a specialist should the condition recur. There is no standard or recommended elective treatment for PSD but evidence suggests that there is no one perfect technique to address all the variables involved (e.g. secondary healing may have a lower recurrence rate but a longer time to healing and flap-based options require in-patient care as opposed to day surgery for primary excision and closure).

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