Int Surg 2018;**103**:443–451
DOI: 10.9738/INTSURG-D-15-00077.1

Pilonidal Sinus—Plenty of Literature yet Lack of Direction: A Pooled Analysis

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A systematic analysis was conducted to compare various surgical treatment modalities and evaluate the short- and long-term outcomes in the management of sacrococcygeal pilonidal disease. Pilonidal sinus is a common and debilitating condition often requiring a surgical approach to treatment, of which many are available. Despite numerous treatment methods, no universal decision has been made as to the most efficient and effective surgical treatment. All randomized control trials published between January 1, 2003 and January 8, 2013 were accessed. Interventions included classic and modified Limberg flap, Karydakis flap, primary closure, and healing by secondary intention. Primary outcome measures included rate of surgical site infection, recurrence, and length of hospital stay. Secondary outcome measures were hematoma or seroma, and return to work. The search identified 22 articles suitable for inclusion in this review totaling 3693 patients. Analysis showed a 50% reduction in the rate of infection and recurrence in patients receiving closure with flaps compared with primary midline closure and healing by secondary intention. The Karydakis flap was shown to be more favorable cosmetically; however, it failed to compete with the classic and modified Limberg flap in treatment of complicated disease. No significant difference was noted between the modified Limberg and Limberg flap. These results show that the Karydakis flap, classic, and modified Limberg flap demonstrate significant benefits over healing by secondary intention and primary closure.

Key words: Pilonidal sinus – Complications – Colorectal

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Sacrococcygeal pilonidal sinus disease (PNS) is a relatively common and chronic condition occurring in approximately 26 out of 100,000 people.[1] It is 3 times more prevalent in males and affects teenagers and young adults, leading to significant pain, morbidity, and body image issues in an otherwise healthy population.[2]

The first published case of PNS was reported in 1833 by Herbert Mayo identifying the hair containing sinus in the sacrococcygeal region.[3] Few surgeons were aware of the disease and little literature was published until 1944 during World War II when Buie reported 77,000 cases of PNS among army personnel.[4]

Despite almost 2 centuries passing since this disease was first noted, no standard management for pilonidal disease exists, but numerous methods of treatment are available. All treatments are weighed against the optimal treatment, one of low cost, fast recovery, minimal time off work, as well as a low complication and recurrence rate with a satisfactory cosmetic result.[5] Due to the lack of consensus among the surgical community, various methods have been trialed ranging from conservative treatment to wide excisions and extensive flap procedures with inconsistent outcomes.[6,7]

Peterson *et al* published a pooled analysis including over 10,000 participants surgically treated for PNS. It was concluded that asymmetric oblique closure techniques and the use of flaps following disease excision had a significant benefit and more favorable surgical outcomes in comparison with closure in the midline.[8] Since this article, increasing numbers of publications comparing asymmetric closure, various flap procedures for easy closure, and complete excision have been reported. No treatment has shown consistent benefit over another and it has been suggested that introducing a stepwise individualized approach may be appropriate.[9,10]

This systematic data analysis aims to summarize data published over the last decade looking at the current surgical treatments for pilonidal sinus disease comparing rates of postoperative complications, hospital stay, return to work, and recurrence between different procedures, while attempting to identify causes of variation within the data.

Materials and methods

Relevant studies published between January 2003 and September 2013 were identified using MEDLINE, Scopus, and Google Scholar. The search terms "pilonidal," "sacrococcygeal sinus," and "complications" were used. A language restriction was placed, searching for articles in English only. Additional articles were identified via screening the references of all studies reviewed for inclusion. Any articles that could not be obtained were referred to the librarians for retrieval.

Two authors independently reviewed the abstracts for relevance and suitability for inclusion. Randomized control trials (RCT) comparing various surgical procedures for the treatment of chronic pilonidal sinus were included. Studies were excluded if they failed to report postoperative complications and the recurrence of pilonidal disease or if they focused on the adolescent or pediatric population. One reviewer extracted study data on a standardized data extraction form, which was further assessed by the second reviewer. Any inconsistencies were discussed and resolved.

A PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flowchart is included (Fig. 1). The articles were analyzed for risk of bias using the Cochrane risk of bias assessment tool.[11] Each of the included studies was critically appraised based on their sequence generation and allocation concealment, whether they addressed incomplete data and whether they were free of selective reporting and other bias.[11]

The principal outcomes included rate of recurrence, surgical site infection, seroma or hematoma, and return to work. The secondary outcomes included other postoperative complications and patient satisfaction. Each outcome was compared quantitatively.

Results

Study flow and characteristics

Electronic searching identified 1026 citations from Medline and Scopus through January 1, 2003 to September 2013. On screening of articles for inclusion criteria 741 publications were excluded. Of the 48 articles identified and obtained for full text review; 13 were excluded as they were retrospective or nonrandomized studies, and 2 articles were not accessible in full text.[12,13] A further 10 studies were not included in the analysis because of insufficient or noncomparable data, and 2 articles were excluded due to data being published twice (Fig. 1).[14–24]

Twenty-two randomized control trials were included, published between 2005 and 2013, and compromising a total of 3693 participants. In terms of geographical location, 18 of the articles in this

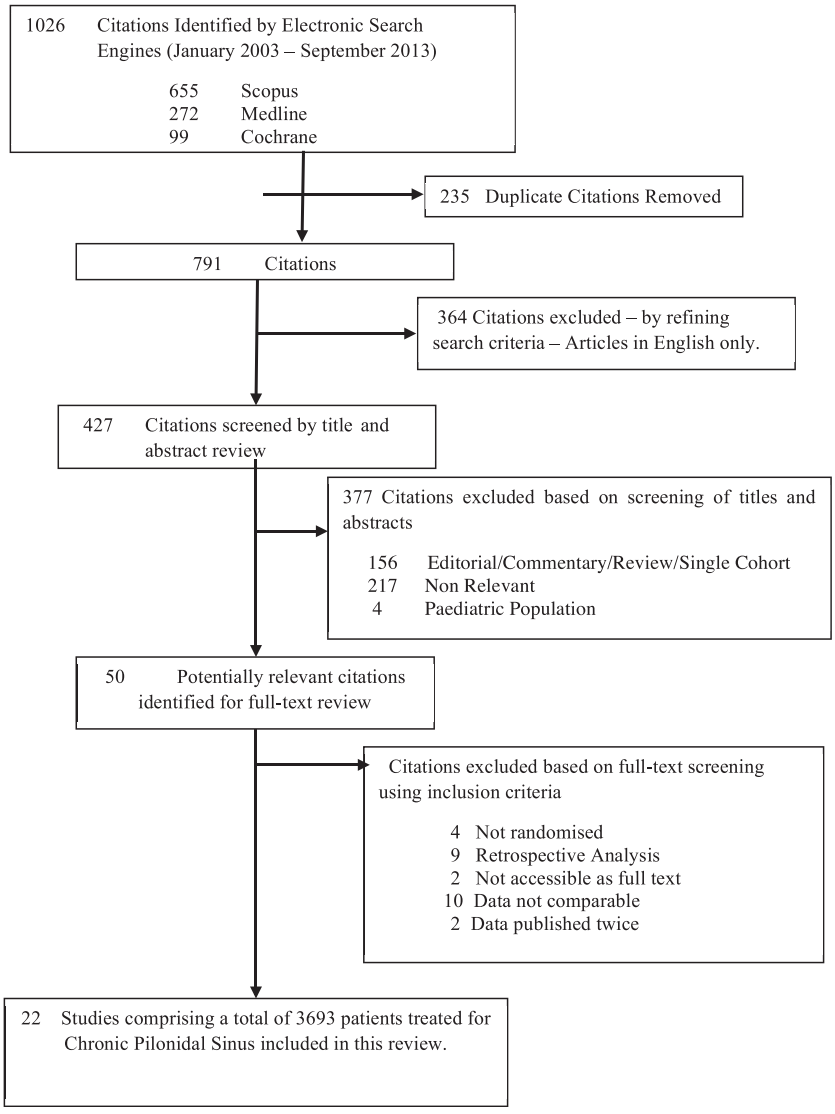

Fig. 1 Flow diagram showing identification, screening, eligibility, and inclusion in this review.

study were published in the Middle East. The overall sample size within individual studies ranged from 40 to 803 participants. Males represented 81.7% of the pooled data population, and the average age was 25.3 years.

All articles included patients with pilonidal sinus disease; in 4 cases this inclusion criteria was further specified with only chronic PNS included.[25–28] In a further 3 studies only symptomatic PNS was included. Elshazly *et al* specified only uncomplicated sinuses, and Hosseini *et al* specified uncomplicated and symptomatic PNS.[29–32] No articles provided reasoning or definitions for specified criteria.

In 14 publications, acute abscesses were excluded.[25,27,30–41] In the remaining 8 trials, acute infection was included but treated prior to further operation.[26,28,29,42–46] In 11 trials, persons with recurrent pilonidal sinus were excluded and Marco *et al* (2011) further specified this criteria excluding only those previously treated with flap surgery.[27,31,33–37,42–44,46] Complex PNS was excluded in 2 articles. Only Ates *et al* (2011) provided definition of the criteria: sinuses extending laterally from the midline or close to the anus.[33,36] Three articles excluded sinuses lateral to the midline, some defining a specified distance, ranging from exclusion of any sinuses lateral to the midline, to up to 5 cm from the midline.[33,35,38] Arslan *et al* used the American Society of Anesthesiologists (ASA) score to categorize their patients excluding any classed as >ASA III.[42] One study explained their reasoning, describing it as an attempt to simplify and standardize the treatment group.[38]

Nineteen articles discussed patients' presentation with the most common symptoms being pain and discharge. The length of symptoms prior to presentation was recorded in 6 RCTs with a pooled average of 22 months.[32,35,37,39–41] Ertan *et al* further defined their patients according to the presence of single or multiple sinuses within the midline or lateral to the midline.[40]

Excision and rhomboid flap (Limberg/Duforemental) was discussed in 12 articles totalling 14 cohorts and 810 participants.[29,31,32,34–36,38,40–42,44] Thirteen studies focused on primary midline closure (PMC) with 1680 participants.[25–29,31–34,37,39,40,43] Karydakis flap (KF) was used in 6 publications with a pooled cohort of 478 participants.[28,30,36,38,42,46] Finally modified Limberg flap (MLF) and excision with healing by secondary intention were discussed in 4 and 5 studies totaling 278 and 347 participants, respectively.[25–27,30,33,39,41,42,45]

Surgical site infection

Surgical site infection (SSI) was an end point in all 22 studies with only 5 studies defining this term. A significant range in criteria for inclusion was present: extending from redness and or edema of the skin to purulent discharge.[24,25,33,41,43,44] Pooled data analysis showed the MLF had the lowest rate of SSI occurring in an average of 4% (3%–5.2%) of participants, followed by the LF with SSI occurring in 5.5% (0–22.9%) then the KF in 6.4% (0–26%). Infection occurred in 12.5% of the PMC cases and in 16.3% of cases healing by secondary intention.

Antibiotic use was reported in all but 2 studies.[27,38] They were used preoperatively in 14 studies, postoperatively in 1 study, and pre0 and postoperatively in 3 studies. Antibiotics were not used in 2 of the studies.[38,46]

Hospitalization time

The length of hospital stay was recorded in 18 studies. The RF had the shortest hospitalization time when performing pooled analysis with an average stay of 2.7 days. MLF and KF had an average stay of 3 days and PMC had a stay of 3.8 days. Participants with defects healing by secondary intention had the longest hospital stay of 3.9 days. For cohorts with drains *in situ*, the length of hospital stay was prolonged.

Seroma and hematoma formation

Rates of seroma and hematoma were reported in 19 trials with secondary closure reporting the lowest rate of occurrence in 1.3%. The groups who received PMC, MLF, and LF had rates of 3.4%, 3.5%, and 4.7%, respectively. The cohort that received the KF reported the highest rates of collections, occurring in an average of 9.9%.

All studies reported the presence or absence of a drain *in situ*, with 29 individual trial cohorts receiving drains, 10 cohorts with no drains *in situ*, and 5 cohorts undergoing secondary closure thereby not requiring drainage.

More than a 50% reduction in the rate of postoperative seroma and hematoma formation was seen in those with drains in situ, in both the LF and KF cohort.

Time to return to work

The average time to return to work or normal activity was recorded in 17 studies and was defined as from the time of surgery until the return to employment or leisure.[29,33] The fastest return to work was in those receiving the LF reporting an average of 12.7 days. In those receiving the MLF, KF, or PMC the average number of days to return to work was 16.7, 15.4, and 18.8, respectively. Those healing by means of secondary intention had a substantial increase in the number of days off work at 29.3 days.

Recurrence rate

The rate of recurrence was measured in all but 1 RCT.[38] The highest rate of recurrence among the pooled data was in those healing by secondary intention with a rate of 10.2% measured at an average follow-up period of 31.8 months. This was closely followed by PMC with 9% recurrence at an average of 27.2 months. In those receiving the KF and LF, it occurred in 3.6% and 3.7%, respectively, at an average follow-up time of 24.7 months. The lowest rate of recurrence was in those receiving the MLF with 2.3% recurrence at 25.2 months.

Other

Other postoperative outcomes measured included early bridging, dehiscence, maceration, flap edema, and necrosis.

Patient satisfaction was discussed in 8 publications, with 2 articles using the visual analog scale score to judge satisfaction and the other 5 publications rating satisfaction on a scale from bad/unsatisfied to excellent/highly satisfied. One article

monitored patient satisfaction via the Cardiff wound impact schedule, comparing preoperative with postoperative scores.

Study risk of bias

Due to the surgical nature of these procedures blinding of participants and surgeons was impossible and, therefore, this criteria was not included. Of the 22 studies included, only 2 studies (9%; n = 260 participants) were judged low risk on all risk of bias criteria considered. In 7 studies (31.8%), the cohorts were selected using random or consecutive sampling. Selective reporting of results was noted in 1 article, and according to author's judgment, the endpoints were joined and overall results were not clearly described. Other potential threats of bias were noted in 3 articles, 1 due to the significant differences in follow-up times between cohorts and the other 2 due to the shifting of patients from 1 group to another following randomization.[25,28,30]

Discussion

Pilonidal sinus disease is a significant condition occurring in 26 per 100,000 people.[47] It occurs in the young and otherwise healthy population leading to significant body image issues and loss of productivity for the patient. It is reported to be more prevalent in males, and, in this study, it compromised 81.7% of the participants.[3]

Numerous surgical procedures have been proposed for the treatment of pilonidal sinus, ranging from simple excision and healing by secondary intention, or midline closure to advanced flap mobilization. Despite these many procedures, no clear consensus as to treatment has been reported.

Healing by secondary intention

Excision and healing by secondary intention was the first reported technique used for the treatment of pilonidal sinus.[48] It has remained an option despite its prolonged hospital stay, patient discomfort, and loss of patient productivity due to the low rate of SSI and recurrence.[49] Rates of recurrence, however, have not shown to be consistent with reports of recurrence and failure ranging from 0–21%.[50,51] Interestingly, in this study, healing by secondary intention showed not only a significant increase in hospital stay, surgical site infection, and return to work, but it also identified an increased rate of recurrence of 10.2% when compared with off midline and flap techniques. It should be noted, however, that this cohort also reported the longest follow-up period in comparison with all other groups, as well as a significant infection rate in the specific studies, which have been previously linked to increased recurrence. The average time to return to work was 29.3 days, a considerable loss in working days in such a young population. These results suggest that the complications and negative impact on the patient may well outweigh the suggested benefits from this procedure.

Primary midline closure

Excision and PMC have previously reported a healing time that is significantly shorter than secondary intention, with decreased length of hospitalization.[52] Our pooled analysis showed similar results with a slightly shorter hospital stay (3.8 versus 6.25 days) and time to return to work (18.8 versus 29.3 days) when compared with healing by secondary intention. PMC, however, is reported to have its downfalls. Most importantly it has been shown to leave the patient with a midline scar and a continuing presence of the natal cleft, which is associated with an increased rate of infection and recurrence due to vulnerability and persisting natal forces.[9] Vulnerability worsens and forces increase exponentially with increasing size of the wound leading to increased postoperative complications and pain. In this study infection, occurred in 12.5% of the pooled group and was associated with a recurrence rate of 9% with significant range between studies from 1.9%–17.5%.

Flap techniques

The Karydakis flap was introduced as a means by which to remove the scar from the midline and obliterate the natal cleft.[1] This procedure has been shown to eliminate the risk of increased vulnerability to the natal cleft and decreased natal height and forces, reducing infection and the rate of recurrence of PNS. Our study showed a 50% reduction in the rate of SSI when compared with PMC, and patients returned to work over 3 days earlier. However a 2-fold increase in the rate of seroma and hematoma formation occurred.

Karydakis reported rates of recurrence as low as 1% whereas others have shown rates of up to 10% recurrence.[1,13] Our pooled results showed a mean recurrence rate of 3.6%, again less than 50% of that of PMC. Although the KF shows excellent results

Table 1 Pooled data—postoperative outcomes following pilonidal surgery

	No. of participants	LOS[a]	SSI %	Collection[b] %	RTW[c]	# Reviewed[d]	% Re-occur	Date measured[c,e]
Rhomboid flap	810	2.7 (1–5.7)	5.7 (0–22.9)	4.9 (0–22.2)	13.1 (8.2–20.8)	811	3.8 (0–11.1)	25.5 (6–38.7)
Modified Limberg flap	278	3 (1.3–4.8)	4 (3–5.2)	3.5 (0–10.2)	16.7 (9.3–21.5)	274	2.3 (0–5.4)	25.2 (16.8–32.9)
Karydakis flap	478	3 (1.3–5.5)	6.4 (0–26)	9.9 (0–32)	15.4 (8.5–19.1)	418	3.6 (0–11)	24.7 (16.8–33.3)
Primary midline closure	1680	3.8 (1–6)	12.5 (0–34)	3.4 (0–14.3)	18.8 (10.4–28.5)	1680	9 (1.9–17.5)	27.2 (6–60)
Healing by secondary intention	347	6.3 (4–5.7)	16.3 (0–43.2)	1.3 (0–4.0)	29.3 (3–42.2)	347	10.2 (1.4–2.8)	31.8 (18–49.9)

LOS, length of stay; RTW, return to work; SSI, surgical site infection.

[a]Days.

[b]Seroma or hematoma.

[c]Months.

[d]Total number to attend follow-up appointments.

[e]Date measured is the date at which patients' pilonidal sinus reoccurrence was noted.

with less severe pilonidal sinus, it has been shown to have its limitations. As the size of the sinus increases, the tension within the closed wound increases, leading to increased rates of failure. In some cases the use of KF is impractical due to the size of the lesion. In this analysis 1 trial was forced to change patients postrandomization from the KF group into a cohort receiving flap procedures, as the laterally extending sinus orifices prevented them from using this treatment.[30]

The Limberg flap is used to close the resultant natal defect, using well vascularized tissue of the same thickness and color.[52,53] It has shown to allow for decreased tension on the wound, reducing the rate of complications and increasing the rate of healing.[54] In our study this cohort had the lowest pooled hospital stay of 2.7 days and shortest time to return to work—13.1 days. Results showed almost 5% of the population had a fluid collection; however, in pooled analysis of those with drainage *in situ*, it was seen in 3.4% as compared with 9.7% without drainage.

This flap, however, has also been shown to have its pitfalls in that the scar crosses the natal cleft allowing for increased susceptibility to infection and recurrence. The use of flaps has had outstanding results; however, the risks are higher with risks of flap necrosis and large deficits to heal by secondary intention if the flap fails. The resultant scar is also a concern with many persons being affected by the cosmetic outcome following this surgery with 1 study showing up to 63% of patients to be unhappy with the cosmetic appearance of their scars.[55] Few studies looked at patient satisfaction in our study, but reports were much improved on those discussed already, with Roshdy reporting 65.7% of those receiving Limberg flap reported their satisfaction of the procedure as excellent and only 2.9% of reports being unsatisfied with the procedure.[32]

A small modification to the LF allows for complete closure of the wound without sutures intervening with the natal cleft. When combining this with the proposed advantages of the LF discussed already it allows for a reduction in the amount of postoperative outcomes as well as a reduced recurrence rate. Our pooled analysis showed similar to others with MLF to have the lowest rate of SSI (4%) and recurrence (2.25%; Table 1).

This article would not be complete without mentioning a few other well vascularized, imported flaps for the repair of pilonidal defects. Due to the small number of randomized control trials currently published with regard to these flaps, they were unable to be included in the pooled analysis. They are, however, becoming increasingly popular in the repair of pilonidal defects.

V-Y advancement flaps have been used in the treatment of pilonidal sinus disease for many years. The rate of acute and chronic complications has been shown to be favorable with rates as low as 5% for wound dehiscence and infection, comparable with those of the Limberg flap.[56] This flap has, however, been reported to have a significantly increased delay in return to daily activities when compared with other methods of repair with studies showing a mean return to work of 32.7 (18–47) days. It also has mixed reports on cosmetic outcomes and

is therefore mostly reserved for closure of extensive defects.[56,57]

The Keystone flap was designed by Felix Behan in 1994, first published in 2003, and is now routinely used in various regions of the body.[58] This island flap integrates 2 V-Y advancement flaps orientated side by side. The geometrics of this design allows for a larger range of mobility and a smaller resultant lateral defect when compared with the V-Y flap thereby allowing for more ease in the closure of the initial defect.[59] This fasciocutaneous flap has also been shown to be quite versatile with Behan recently discussing the use of 2 Keystone flaps named the Yin Yang flap for the repair of extensive pilonidal defects that were previously left to heal by secondary intention.[60]

Various other perforator flaps are becoming increasingly more common as they too allow for flexibility in design and movement. Based on the local vascular network they provide a simple, efficient and expeditious way of repairing surgical defects. Perforator flaps have reduced donor-site morbidity to a minimum, and refined techniques have resulted in collection of less tissue with optimal cosmetic result. Patient recovery time has been minimized and patients experience less pain and faster rehabilitation.[61–63]

Limitations

It can be seen from the aforementioned data that a variety of procedures can be used to treat pilonidal sinus disease highlighting the variable complexity of pilonidal sinus disease and its unpredictable postoperative outcomes. This makes it difficult to make a comparison between strategies where the patient cohorts and disease severity vary considerably. Secondly treatment is a surgeon-dependent technique, and this makes it difficult to compare treatments among studies. Due to the nature of surgical treatment for pilonidal sinus, it was impossible for blinding to occur among doctors and patients for this treatment; therefore, bias may exist. Trial sizes were small in size and notable discrepancy existed in inclusion and exclusion criteria as well as criteria definitions between studies with many studies excluding complex disease or sinus lateral to the natal cleft. Several small variations in surgical techniques also existed among studies including method of incision and drainage, the use of various antibiotics, and suture materials. Inconsistencies in follow-up times for measurement of recurrence were also present.

Conclusion

Pilonidal sinus disease remains a challenging and controversial surgical topic with various treatments and techniques available. The condition itself is complex and presents with varying degrees of severity from minor asymptomatic sinuses to widespread, lateralized recurrent disease.

This pooled data analysis shows an advantage of off-midline and flap procedures over standard primary midline closure and healing by secondary intention with more than 50% reduction in surgical site infections and recurrence rates.

The Karydakis flap proved equally as effective as other flap procedures in this analysis for less severe lesions and is favored by patients cosmetically; however, data suggests it fails to perform for more extensive lesions. It is recommended that larger flaps, including the modified Limberg and Limberg flap, be reserved for more extensive lesions.

Unfortunately it is difficult to make a significant comparison between the current surgical treatments available for pilonidal sinus disease due to the wide variability of lesion severity at presentation and postoperative outcomes. The authors note that no standard universal classification method currently exists for the severity of pilonidal sinus disease. It is proposed that this is needed as a means to allow for appropriate comparison of surgical treatments as well as a standardized approach to data collection.

Acknowledgments

Jemma Kate Porrett notes that the material presented in this thesis is her own work. All information derived from the work of others, published and unpublished, has been acknowledged in the text and references given. Assistance and guidance of others in preparing this thesis has been duly noted. Ms. Porrett states that this work complies with the Australian National Statement on Ethical Conduct in Human Research and the Australian code for the care and use of animals for scientific purposes where applicable. The authors declare no conflict of interest.

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