

Surgical interventions for the treatment of sacroccocygeal pilonidal sinus disease in children: A systematic review and meta-analysis

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Abstract

Background

Pilonidal sinus disease (PNS) is not uncommon in children. Controversy remains over the best treatment and there is limited evidence. This systematic review and meta-analysis aims to establish which techniques have the best outcomes in children.

Methods

MEDLINE, EMBASE and CENTRAL databases were searched. Studies reporting treatment outcomes for PNS in children were included.

Results

Open healing has pooled risk of recurrence of 26% (95%CI 15-38%), risk of wound complication of 21% (9-36%) and wound healing ranged from 38-92 days. Midline primary closure has pooled risk of recurrence of 12% (8-18%), risk of wound complication of 30% (19-46%) and wound healing ranged from 8-32 days. Off-midline primary closure has pooled risk of recurrence of 6% (1-15%), risk of wound complication of 14% (6-25%) and wound healing was 27 days. VAC therapy has pooled risk of recurrence of 20% (0-65%) and wound healing ranged from 38-92 days. Minimally invasive techniques has pooled risk of recurrence of 7% (1-16%) and wound healing ranged from 21-30 days. Marsupialisation has pooled risk of recurrence of 6% (0-22%), and wound healing ranged from 6-41 days.

Conclusion

Evidence for management of PNS in children is poor. Off-midline primary closure, minimally invasive techniques, and marsupialisation have the best outcomes.

KEYWORDS

Pilonidal Sinus, Adolescent, Child, Systematic Review, Surgery

Level of Evidence rating: IV

1. Introduction

Sacrococcygeal pilonidal sinus disease (PNS)¹ is not uncommon in children [1,2]. Onset is usually around the time of puberty. Symptoms such as recurrent acute abscess and chronic suppuration may have significant impact on patients' quality of life [4], education and social integration [2,3]. The ideal treatment for this condition should cause minimal disruption or discomfort whilst having good cure and recurrence rates.

Systematic reviews and meta-analyses [4,5] in the treatment of adult PNS have indicated wide excision and healing by secondary intention has the lowest long term recurrence, whilst excision with primary closure via an off-midline or flap technique have quicker healing and only slightly less favourable recurrence rates. These approaches involve significant tissue loss, a hospital admission often of several days[2] and a long time (14-60 days) until return to normal activities [6,7].

More recently minimally invasive techniques including fibrin glue obliteration [8,9], minimal excision [10], endoscopic [11,12] and crystallized phenol [13] and have been described in both adults and children. These treatments reportedly offer quicker healing and return to normal activities with acceptable recurrence rates. However, long term follow-up data is absent.

The treatment of PNS in children has generally mirrored that in adults, but there has been little research in in children [1,14]. It is not well understood whether adult outcomes can be generalised to children; indeed some studies have indicated that long term recurrence rates are higher in patients where surgery is required at an earlier age [15]. This may be due to a genetic predisposition to developing the disease, or that PNS presenting in childhood tends to be more severe or that the conditions predisposing to PNS are present for a longer period. Furthermore, as surgical interventions in children may impact on schooling, sporting

¹ PNS – pilonidal sinus disease

activities and social integration, different outcome priorities may exist for treatment in this age group.

We have therefore carried out a systematic review and meta-analysis of all published evidence regarding the treatment of pilonidal sinus disease in the paediatric population with the aim of establishing which techniques have the best outcomes for recurrence, wound healing, wound complications, and which are most acceptable to patients. We also looked for evidence on quality of life and time to return to normal activities.

2. Method

Study design

This systematic review was registered prospectively with PROSPERO (registration number CRD42018095297) and was carried out in accordance with the PRISMA statement [16]. Any study reporting treatment outcomes for sacrococcygeal PNS in a population with median age of 18 or under were included. The minimum outcome reporting required for inclusion was recurrence rate. Studies which did not report treatment outcomes, reported non sacrococcygeal PNS or reported outcomes in an adult population (median age >18) were excluded.

Literature search

Literature searches were carried out by a trained Clinical Research Librarian using the following databases: MEDLINE, EMBASE and CENTRAL (all searched from their inception to 17th May 2018). No language or date restriction was applied to the searches. The Cochrane Library of Systematic Reviews was searched for relevant reviews and the abstracts from the conference proceedings of the British Society of Paediatric Surgeons for the last 10 years were searched for relevant unpublished studies. Previous systematic reviews of related topics were also searched for relevant studies. References of identified potentially relevant studies were hand-searched for further studies. Finally, all studies citing

the identified potentially relevant primary studies identified on Google Scholar were screened for inclusion. Example search strategies can be found in Appendix A.

Abstracts were screened independently by two authors (EH and PH) with the aid of Rayyan systematic review software (2016, Qatar Computing Research Institute, Doha, Qatar) [17] and considered for full text review if either author deemed them to be potentially relevant. A grey literature search as described above was completed by one author (RR). Full text versions of all potentially relevant primary studies were then independently screened against the inclusion and exclusion criteria by two authors (EH and PH) and agreement to inclusion reached by consensus.

Data extraction

Study characteristics and outcome data were independently extracted and verified by two authors (EH and HP). Risk of bias for included studies was assessed independently by two authors using the Risk of Bias In Non-randomized Studies – of Interventions (ROBINS-I) assessment tool [18] (EH and PH) with any disagreement resolved by consensus.

Statistical analysis

All analyses were conducted in STATA Version 15 (StataCorp LLC, Texas). We used the metaprop user-written. Effect estimates are presented as proportions with 95% confidence intervals (CI) as none of the included studies reported outcomes as time to event. Pooled estimates were calculated using the Freeman-Tukey double arcsine transformation and CIs were calculated using the exact method. We used random effects models due to the clinical heterogeneity between the included studies. Statistical heterogeneity was assessed using the I^2 statistic. To assess the association between follow up time and rate of recurrence we used a maximum likelihood random effects meta-regression with the Knapp Hartung modification. To assess publication bias, if ten or more studies were included we conducted Egger's linear regression test. Due to the previously published problems with conventional

tests for publication bias and proportion outcomes we used study size rather than standard errors on the Y axis [19].

3. Results

A total of 5128 potentially relevant abstracts were screened for inclusion, of which 2664 were unique papers. Of the 2664 unique abstracts screened, 41 studies were identified for full text review. Following full text review a further 15 studies were excluded as they either did not report recurrence rates, did not clearly report which surgical technique was used or were performed in adults. Twenty six studies were included in the qualitative synthesis and 23 in the quantitative analysis (Figure 1).

Study characteristics

The characteristics of included studies are shown in Table 1. No studies published before 2002 met the criteria for inclusion. All studies had full texts published in peer reviewed journals. All included studies were retrospective case series or cohort studies. Eleven case series reported the results of one surgical intervention alone, whilst a further 11 cohort studies compared results of 2 different interventions, and 4 papers reported the results of 3 different surgical intervention. Sample sizes ranged from 8 to 268 patients. Only 4 studies reported outcomes of over 50 patients for any one intervention, and only 3 studies included a total of over 100 patients.

All but 1 study were single centre studies. Ten studies were based in the USA, 5 studies were from Turkey, 2 from each of the UK, Israel, Italy and Canada and 1 each from Spain and Italy. Ethnicity of subjects was not reported in any study.

Median follow-up ranged from 39 days to 54 months. Nine studies reported results with a median follow up of over 12 months.

Risk of bias

Risk of bias was assessed using the ROBINS-I tool (Table 2). Fifteen studies were considered at moderate risk of bias with regard to confounding factors as they had performed no appropriate analysis to adjust for any important confounding factors. The remaining 11 studies were found to be at severe risk in this domain as they had not appropriately controlled for potentially important confounding factors and performed no appropriate statistical adjustment. Eleven studies were judged to be at moderate or severe risk of selection bias as they either did not report selection criteria or selection was based on surgical preference. Two studies were judged to be at serious risk of bias with regard to classification of intervention as they grouped several procedures together in reporting outcomes. One study was at serious risk of bias due to deviation from intended intervention as a random selection of patients receiving laser epilation in addition to the primary intervention, with no reporting of the outcomes for this subset or how these patients were distributed between the 2 groups of primary intervention. All studies were at moderate risk of bias in measurement of outcomes and reporting results as all were non-blinded studies and none had a predefined set of outcomes to be reported.

Overall sixteen studies were found to be at severe risk of bias, whilst the remaining ten studies were at moderate risk.

Data synthesis

All 26 included studies reported recurrence rates, whilst 18 reported wound complication rates, allowing these outcomes to be included in meta-analysis. Only 14 studies reported outcomes for time to wound healing and where reported it was of insufficient detail to allow further analysis. Seven studies reported a measure of patient satisfaction, but the reported measure was highly variable and no studies used any validated quality of life or patient satisfaction measure. Results are summarised in table 3.

Open healing

Six studies reported the outcomes of open healing for 191 patients. All papers were comparative cohort studies, of which 1 study compared open healing to minimally invasive techniques [12], 1 compared open healing to marsupialisation [1], 2 compared open healing to off-midline primary closure [20,21] and 4 compared open healing to midline primary closure [1,21,22]. Overall pooled risk of recurrence (Figure 2) was 26% (95% CI 15-38%). When analysed as a subgroup, studies with median follow-up of over 12 months had a pooled risk of recurrence of 32% (13-53%). There was moderate statistical heterogeneity ($I^2=54.6\%$). Wound complications were reported by 3 studies [12,20,21] and the pooled risk of wound complication was 21% (9-36%, $I^2=36.1\%$) (Figure 3). Average time to wound healing ranged from 38 to 92 days, but was not reported in enough detail to allow pooled analysis. Meta regression analysis, assessing the relationship between length of follow-up and incidence of recurrence, appeared to show a relationship, but this did not reach statistical significance (Fig 4). There was no statistical evidence of publication bias (Egger's test $p=0.996$).

Midline primary closure

Sixteen studies reported the outcomes of midline primary closure for a total of 728 individuals. Five studies were case series which reported the outcome of midline primary closure alone [14,23–26]. Five comparative cohort studies compared midline primary closure to off-midline primary closure [2,8,21,27,28], 4 compared it to open healing [1,21,22,29], 3 compared it to minimally invasive techniques [8,13,30], and 2 to marsupialisation [1,31]. Overall risk of recurrence was 12% (8-18%), and in studies with over 12 months follow up pooled risk of recurrence was 13% (4-24%) (Figure 5). There was substantial heterogeneity between groups ($I^2 =62.2\%$). Wound complications were reported by 13 studies, with a pooled risk of wound complication 30% (19-42%), although there was considerable heterogeneity ($I^2 87.4\%$) (Figure 6). Average time to wound healing was reported by 7 studies and ranged from 8 to 38 days, but was not reported in enough detail to allow pooled analysis. Meta regression analysis assessing the relationship between length of follow-up

and incidence of recurrence appeared to show a relationship, but this was not statistically significant (Figure 7). There was no statistical evidence of publication bias (Egger's test $p=0.889$).

Off-midline primary closure

Nine studies reported the outcomes of off-midline primary closure for a total of 199 patients (including modified Limberg flap [2,8,21,28,32,33], Karydakis [2,8,21,27] and Bascom cleft lift [8,20,34]). Three studies reported the outcomes of off-midline closure alone [32–34], 5 compared it to midline primary closure [2,8,21,27,28], 3 compared it to open healing [2,20,21], and 1 compared it to minimally invasive techniques [8]. Overall pooled risk of recurrence rate for excision and off-midline primary closure is 6% (1-15%) (Figure 8). When analysed as a subgroup, studies with median follow-up of over 12 months had a pooled risk of recurrence of 8% (0-20%). There was substantial statistical heterogeneity ($I^2=67.8\%$). Wound complications were reported by 8 studies with pooled risk of wound complication of 14% (6-25%) with I^2 of 64.81% (Figure 9). Average time to wound healing was reported by only 1 study and was 27 days. There were insufficient studies to perform meta-regression or statistical analysis of publication bias.

Excision + Vac therapy

Two case series reported outcomes of VAC therapy following excision of pilonidal sinus in a total of 29 patients [35,36]. Overall pooled risk of recurrence was 20% (0-65%). There was substantial statistical heterogeneity ($I^2=77.5\%$) (Figure 10). Neither study reported any wound complications. Average time to wound healing ranged from 38 to 72 days, but was not reported in enough detail to allow further statistical analysis. There were insufficient studies to perform meta-regression or statistical analysis of publication bias.

Minimally invasive techniques

Six studies reported the results of minimally invasive approaches in 150 patients. The three studies reported the outcomes of Endoscopic Pilonidal Sinus Treatment (EPSiT) [12,30,37]

in 79 patients overall pooled risk of recurrence was 7% (1-16%, $I^2=24.1\%$) (Figure 11). There was one study reporting each of minimal incision [38], fibrin glue obliteration [8] and crystallised phenol treatment [13]. Recurrence rates for these procedures were 2.5%, 20% and 28.6% respectively. Wound complications were reported in 4 studies and ranged from 0% to 10%. Time to wound healing were reported in all 3 EPSiT studies and ranged from 21-30 days, however there was not sufficient detail reported to allow further statistical analysis. There were insufficient studies to perform meta-regression or statistical analysis of publication bias.

Marsupialisation

Results of marsupialisation were reported in three studies for a total of 53 patients [1,31,39]. All were comparative cohort studies comparing marsupialisation to midline closure and open healing. Overall risk of recurrence was 6% (0-22%) (Figure 12). There was substantial statistical heterogeneity ($I^2=60.6\%$). Wound complications were not reported for this procedure. Average time to wound healing was reported in all 3 studies and ranged from 6 to 41 days, but was not reported in sufficient detail to allow further statistical analysis. There were insufficient studies to perform meta-regression or statistical analysis of publication bias.

4. Discussion

We found that evidence for the management of paediatric PNS is limited. Only 26 publications reporting the outcomes of the management of paediatric pilonidal sinuses in 1399 patients were identified. All of these studies were of low quality, and the majority had low number of participants, short follow-up periods and were of moderate to severe risk of bias.

Off-midline primary closure, marsupialisation and minimally invasive techniques have the best outcomes for recurrence rates (6-7%), wound complications (3-14%) and average time to wound healing (6-41 days).

Open wound healing and VAC therapy had the worst reported outcomes with pooled risk of recurrence of 26% and 20% respectively. They also had the longest reported range of wound healing times (38-92 and 38-72 days respectively) and open healing had the second highest wound complication rate (21%).

Midline primary closure has a better pooled risk of recurrence than open healing (12% vs 26%), and average wound healing rates comparable to marsupialisation and minimally invasive techniques (8-38 days). However, midline primary closure has the highest risk of wound complication (30%).

In general the results reported for children are worse than those reported in adults. Stauffer et al [5] report recurrence rates for off-midline closure of 1.6% at 12 months rising to 6.7% at 10 years in adults. In our study, children with median follow-up of just 25.5 months reported recurrence rates of 8% following off-midline closure. The results also showed different trends in children. Excision and open healing was found to have the worst pooled risk of recurrence (26%) in children, whereas in adults this procedure has the best recurrence rates (1.5% at 12 months to 13% at 5 years [5]). There are several possible explanations for this. Doll et al [15] suggest that PNS in children is often more severe, especially if there is a family history. Furthermore it has been shown that for many other surgical procedures that high volumes improve outcomes [40]. Most of these studies include relatively small numbers of patients and it may be that the low volume of procedures being carried out has an impact on outcomes.

In contrast, our findings that off-midline primary closure had better outcomes than midline primary closure is similar to findings of meta-analysis of PNS treatments in adults [4,5].

Off-midline primary closure, marsupialization and minimally invasive techniques were found to be comparable and have the best outcomes in terms of recurrence, wound complications and time to wound healing in children. With equivalent outcomes the best technique would be the one of these which causes least disruption to the patient, especially as recurrence

and reoperation is more likely in children [15]. However, there is virtually no objective or subjective data to guide children and their parents in which of these procedures would be best for them.

The reporting of time to return to normal activities and patient satisfaction was poor across all studies. This information is especially important in children who may miss school and opportunities for social integration because of prolonged wound healing problems and appointments for wound dressing. Future studies should collect results of issues identified as being important by patients, including quality of life, time to return to normal activity and cosmesis.

Although there is little data from using novel, minimally invasive techniques in children these may address some of the issues around impact of surgery on schooling and socialisation. Our study has shown these procedure to have a low risk of recurrence and following fibrin glue obliteration of pilonidal sinus children returned to school after an average of just 3 days [9]. These procedures therefore show great promise as a generalisable treatment for PNS in children.

Limitations:

Evidence is of poor quality and limited conclusions can therefore be drawn. As there have been no true comparative studies it is not possible to give definite guidance on which technique gives the best result.

There is obvious need for well performed, large, randomised controlled studies to further investigate the optimum treatment of sacrococcygeal PNS in children. It is important that future studies assess operative outcomes including long term recurrence rates, wound complication rates, time to wound healing and return to normal activities. It is equally important that studies assess the effect of techniques on the quality of life and patient satisfaction of the children treated as well as developing specific patient reported outcome

measures to facilitate informed consent and allow children and their families to choose the best treatment for them.

5. Conclusion

Evidence for management of PNS in children is poor. Minimally invasive techniques, off-midline primary closure and marsupialization have the best reported risk of recurrence, wound complications and time to wound healing.

As limited evidence to guide treatment is available the effect of the chosen method on the patient's quality of life should be given equal consideration. Patient counselling and choice should be an important part of choosing the right approach.

Good quality research including randomised control trials is required to establish the best treatment option for PNS in children.

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References

- [1] Nasr A, Ein SH. A pediatric surgeon's 35-year experience with pilonidal disease in a Canadian children's hospital. *Can J Surg* 2011;54:39–42. doi:10.1503/cjs.028509.
- [2] Fike FB, Mortellaro VE, Juang D, Ostlie DJ, St. Peter SD. Experience with pilonidal disease in children. *J Surg Res* 2011;170:165–8. doi:10.1016/j.jss.2011.02.016.
- [3] Allen-Mersh G, Allen-Mersh TG. Pilonidal sinus: finding the right track for treatment 1990;77:12–132.
- [4] McCallum IJD, King PM, Bruce J. Healing by primary closure versus open healing after surgery for pilonidal sinus: systematic review and meta-analysis. *BMJ* 2008;336:868–71. doi:10.1136/bmj.39517.808160.BE.
- [5] Stauffer VK, Luedi MM, Kauf P, Schmid M, Diekmann M, Wieferich K, et al. Common surgical procedures in pilonidal sinus disease: A meta-analysis, merged data analysis, and comprehensive study on recurrence. *Sci Rep* 2018;8:3058. doi:10.1038/s41598-018-20143-4.
- [6] H. Abu Galala, Isam M. A. Salam, Kh K, Salam IM, Abu Samaan KR, El Ashaal YI, Chandran VP, Sabastian M, et al. Treatment of Pilonidal Sinus by Primary Closure with a Transposed Rhomboid Flap Compared with Deep Suturing: a Prospective Randomised Clinical Trial. *Eur J Surg* 1999;165:468–72. doi:10.1080/110241599750006721.
- [7] Lund JN. Less Is More in the Treatment of Pilonidal Sinus Disease. *Dis Colon Rectum* 2017;60:e1. doi:10.1097/DCR.0000000000000727.
- [8] Smith CM, Jones A, Dass D, Murthi G, Lindley R. Early experience of the use of fibrin sealant in the management of children with pilonidal sinus disease. *J Pediatr Surg* 2015;50:320–2. doi:10.1016/j.jpedsurg.2014.11.022.
- [9] Hardy E, Herrod P, Sian T, Boyd-Carson H, Blackwell J, Lund J, et al. Fibrin glue

- obliteration is safe, effective and minimally invasive as first line treatment for pilonidal sinus disease in children. *J Pediatr Surg* 2018.
doi:10.1016/J.JPEDSURG.2018.07.024.
- [10] Speter C, Zmora O, Nadler R, Shinhar D, Bilik R. Minimal incision as a promising technique for resection of pilonidal sinus in children. *J Pediatr Surg* 2017;52:1484–7.
doi:10.1016/J.JPEDSURG.2017.03.040.
- [11] Meinero P, Stazi A, Carbone A, Fasolini F, Regusci L, La Torre M. Endoscopic pilonidal sinus treatment: a prospective multicentre trial. *Color Dis* 2016;18:O164–70.
doi:10.1111/codi.13322.
- [12] Esposito C, Izzo S, Turrà F, Cerulo M, Severino G, Settimi A, et al. Pediatric Endoscopic Pilonidal Sinus Treatment, a Revolutionary Technique to Adopt in Children with Pilonidal Sinus Fistulas: Our Preliminary Experience. *J Laparoendosc Adv Surg Tech* 2017;28:lap.2017.0246. doi:10.1089/lap.2017.0246.
- [13] Ates U, Ergun E, Gollu G, Sozduyar S, Kologlu M, Cakmak M, et al. Pilonidal sinus disease surgery in children: the first study to compare crystallized phenol application to primary excision and closure. *J Pediatr Surg* 2018;53:452–5.
doi:10.1016/J.JPEDSURG.2017.05.012.
- [14] Braungart S, Powis M, Sutcliffe JR, Sugarman ID. Improving outcomes in pilonidal sinus disease. *J Pediatr Surg* 2016;51:282–4.
doi:10.1016/J.JPEDSURG.2015.10.076.
- [15] Doll D, Matevossian E, Wietelmann K, Evers T, Kriner M, Petersen S. Family History of Pilonidal Sinus Predisposes to Earlier Onset of Disease and a 50% Long-Term Recurrence Rate. *Dis Colon Rectum* 2009;52:1610–5.
doi:10.1007/DCR.0b013e3181a87607.
- [16] Moher D, Liberati A, Tetzlaff J, Altman DG, PRISMA Group. Preferred reporting items

- for systematic reviews and meta-analyses: the PRISMA statement. *BMJ* 2009;339:b2535. doi:10.1136/BMJ.B2535.
- [17] M O, Hammady H, Fedorowicz Z, Elmagarmid A. Rayyan- a web and mobile app for systematic reviews. *Syst Rev* 2016;5:210.
- [18] Sterne JA, Hernán MA, Reeves BC, Savović J, Berkman ND, Viswanathan M, et al. ROBINS-I: a tool for assessing risk of bias in non-randomised studies of interventions. *BMJ* 2016;355:i4919. doi:10.1136/BMJ.I4919.
- [19] Hunter JP, Saratzis A, Sutton AJ, Boucher RH, Sayers RD, Bown MJ. In meta-analyses of proportion studies, funnel plots were found to be an inaccurate method of assessing publication bias. *J Clin Epidemiol* 2014;67:897–903.
- [20] Gendy AS, Glick RD, Hong AR, Dolgin SE, Soffer SZ, Landers H, et al. A comparison of the cleft lift procedure vs wide excision and packing for the treatment of pilonidal disease in adolescents. *J Pediatr Surg* 2011;46:1256–9. doi:10.1016/J.JPEDIURG.2011.03.062.
- [21] Zagory JA, Golden J, Holoyda K, Demeter N, Nguyen NX. Excision and primary closure may be the better option in the surgical management of pilonidal disease in the pediatric population. *Am Surg* 2016;82:964–7.
- [22] Lukish JR, Kindelan T, Marmon LM, Pennington M, Norwood C. Laser epilation is a safe and effective therapy for teenagers with pilonidal disease. *J Pediatr Surg* 2009;44:282–5. doi:10.1016/j.jpedsurg.2008.10.057.
- [23] Arda İS, Güney LH, Sevmiş Ş, Hiçsönmez A. High Body Mass Index as a Possible Risk Factor for Pilonidal Sinus Disease in Adolescents. *World J Surg* 2005;29:469–71. doi:10.1007/s00268-004-7533-y.
- [24] Mutus HM, Aksu B, Uzun E, Gulcin N, Gercel G, Ozatman E, et al. Long-term analysis of surgical treatment outcomes in chronic pilonidal sinus disease. *J Pediatr Surg*

- 2018;53:293–4. doi:10.1016/j.jpedsurg.2017.11.031.
- [25] Gonzalez-Temprano N, Sanchez-Vazquez M, Ayuso-Gonzalez L, Pison-Chacon J, Perez-Martinez A. [Are we correctly treating pilonidal disease in children? therapeutic goals beyond preventing recurrence]. *Estamos Tratando Bien La Enferm Pilonidal En Los Ninos? Objet Ter Mas Alla Prev La Recidiv* 2011;24:161–4.
- [26] Serour F, Somekh E, Krutman B, Gorenstein A. Excision with primary closure and suction drainage for pilonidal sinus in adolescent patients. *Pediatr Surg Int* 2002;18:159–61. doi:10.1007/s003830100683.
- [27] Morden P, Geiger JD, Drongowski RA, Teitelbaum DH, Hirschl RB. Comparison of Karydakis versus midline excision for treatment of pilonidal sinus disease. *Pediatr Surg Int* 2005;21:793–6. doi:http://dx.doi.org/10.1007/s00383-005-1543-1.
- [28] Yildiz T, Ilce Z, Küçük A. Modified Limberg flap technique in the treatment of pilonidal sinus disease in teenagers. *J Pediatr Surg* 2014;49:1610–3. doi:10.1016/j.jpedsurg.2014.06.011.
- [29] Ozcan R, Hüseyinov M, Bakır AC, Emre S, Tütüncü C, Celayir S, et al. Which treatment modality for pediatric pilonidal sinus: Primary repair or secondary healing? *Asian J Surg* 2018;41:506–10. doi:10.1016/j.asjsur.2017.08.006.
- [30] Sequeira JB, Coelho A, Marinho AS, Bonet B, Carvalho F, Moreira-Pinto J. Endoscopic pilonidal sinus treatment versus total excision with primary closure for sacrococcygeal pilonidal sinus disease in the pediatric population. *J Pediatr Surg* 2018;53:2003–7. doi:10.1016/j.jpedsurg.2018.02.094.
- [31] Lee SL, Tejrjian T, Abbas MA. Current management of adolescent pilonidal disease. *J Pediatr Surg* 2008;43:1124–7. doi:10.1016/J.JPEDSURG.2008.02.042.
- [32] Afşarlar ÇE, Yılmaz E, Karaman A, Karaman İ, Özgüner İF, Erdoğan D, et al. Treatment of adolescent pilonidal disease with a new modification to the Limberg flap:

- Symmetrically rotated rhomboid excision and lateralization of the Limberg flap technique. *J Pediatr Surg* 2013;48:1744–9. doi:10.1016/J.JPEDIURG.2013.01.029.
- [33] Yamout SZ, Caty MG, Lee Y-HH, Lau ST, Escobar MA, Glick PL. Early experience with the use of rhomboid excision and Limberg flap in 16 adolescents with pilonidal disease. *J Pediatr Surg* 2009;44:1586–90. doi:10.1016/j.jpedsurg.2008.11.033.
- [34] Umesh V, Sussman RH, Smith J, Whyte C. Long term outcome of the Bascom cleft lift procedure for adolescent pilonidal sinus. *J Pediatr Surg* 2018;53:295–7. doi:10.1016/j.jpedsurg.2017.11.036.
- [35] Bütter A, Emran M, Al-Jazaeri A, Ouimet A. Vacuum-assisted closure for wound management in the pediatric population. *J Pediatr Surg* 2006;41:940–2. doi:10.1016/J.JPEDIURG.2006.01.061.
- [36] Caniano DA, Ruth B, Teich S. Wound management with vacuum-assisted closure: experience in 51 pediatric patients. *J Pediatr Surg* 2005;40:128–32. doi:10.1016/J.JPEDIURG.2004.09.016.
- [37] Pini Prato A, Mazzola C, Mattioli G, Escolino M, Esposito C, D'Alessio A, et al. Preliminary report on endoscopic pilonidal sinus treatment in children: results of a multicentric series. *Pediatr Surg Int* 2018;34:687–92. doi:10.1007/s00383-018-4262-0.
- [38] Speter C, Zmora O, Nadler R, Shinhar D, Bilik R. Minimal incision as a promising technique for resection of pilonidal sinus in children. *J Pediatr Surg* 2017;52:1484–7. doi:10.1016/j.jpedsurg.2017.03.040.
- [39] Rouch JD, Keeley JA, Scott A, Sydorak R, DeUgarte D, Lee SL. Short- and long-term results of unroofing and marsupialization for adolescent pilonidal disease. *JAMA Surg* 2016;151:877–9. doi:10.1001/jamasurg.2016.0850.
- [40] Aquina CT, Probst CP, Becerra AZ, Iannuzzi JC, Kelly KN, Hensley BJ, et al. High volume improves outcomes: The argument for centralization of rectal cancer surgery.

Surgery 2016;159:736–48. doi:10.1016/j.surg.2015.09.021.

Legends for figures

Figure 1. PRISMA flow diagram

Figure 2. Risk of recurrence following excision and open healing

Figure 3. Risk of wound complications following excision and open healing

Figure 4. Meta-regression analysis of length of follow-up vs recurrence rate for open wound healing

Figure 5. Risk of recurrence following midline primary closure

Figure 6. Risk of wound complications following midline primary closure

Figure 7. Meta-regression analysis of length of follow-up vs recurrence rate for midline primary closure

Figure 8. Risk of recurrence following off-midline primary closure

Figure 9. Risk of wound complications following off-midline primary closure

Figure 10. Risk of recurrence following VAC therapy

Figure 11. Risk of recurrence following EPSiT

Figure 12. Risk of recurrence following Marsupialisation

Table 1: Characteristics of included studies

Paper	Year	Country	N (total)	Median age (years)	Gender		Procedure	n	Outcome reported	Average follow up (months)
					Male	Female				
Afsarlar <i>et al</i>	2013	Turkey	15	14	8	7	Off midline primary closure	15	Recurrence, complications, length drain in situ, length of stay	4
Arda <i>et al</i>	2005	Turkey	14	15.4	12	2	Midline primary closure	14	Recurrence, complications	3
Ates <i>et al</i>	2018	Turkey	117	15.6	65	52	Minimally Invasive (Crystallized phenol)	40	Recurrence, complications, need for IV analgesia	8.1
							Midline primary closure	77		44.6
Braungart <i>et al</i>	2016	UK	19	15	9	10	Midline primary closure	19	Recurrence, complications	13
Butter <i>et al</i>	2006	Canada	8	NR	NR	NR	Excision + VAC	8	Recurrence, time to wound closure	Not reported
Caniano <i>et al</i>	2005	USA	21	16	NR	NR	Excision + VAC	21	Recurrence, complications	Not reported
Eposito <i>et al</i>	2017	Italy	15	16	9	6	Minimally invasive (Endoscopic)	15	Recurrence, wound healing, pain, length of stay, patient satisfaction	6
							Open healing	15		
Fike <i>et al</i>	2011	USA	120	14.9	50	70	Midline primary closure	74	Recurrence, wound breakdown, no. of follow up visits	Not reported
							Off midline primary closure	18		
							Other (not well defined)	21		
Gendy <i>et al</i>	2011	USA	70	16	49	21	Off midline primary closure	39	Recurrence, complications, primary wound healing incidence	19
							Open healing	37		45
Gonzales <i>et al</i>	2011	Spain	20	13.3	5	15	Midline primary closure	20	Recurrence, length of stay	Not reported
Lee <i>et al</i>	2008	USA	26	16.7	13	13	Marsupialisation	17	Recurrence, Time to wound closure, re-operative rate	6.4
							Primary closure (unspecified)	9		
Lukish <i>et al</i>	2009	USA	28	17.2	17	11	Midline primary closure	17	Recurrence	24.2

							Open healing	8		
Morden <i>et al</i>	2005	USA	68	13.8	30	38	Off midline primary closure	24	Recurrence, complications	49.4
							Midline primary closure	44		
Mutus <i>et al</i>	2018	Turkey	268	146	146	122	Midline primary closure	268	Recurrence, complications	Not reported
Nasr <i>et al</i>	2011	Canada	121	15	64	57	Open healing	90	Recurrence, time to healing	1.2
							Midline primary closure	4		
							Marsupialisation	13		
Ozcan <i>et al</i>	2017	Turkey	47	15.6	22	25	Open healing	11	Recurrence, length of stay, recovery time	43.2
							Midline primary closure	36		
Prato <i>et al</i>	2018	Italy	43	15	20	23	Minimally invasive (Endoscopic)	43	Recurrence, length of procedure, length of stay, reoperation rate	4
Rouch <i>et al</i>	2016	USA	39	16	19	20	Other (not well defined)	16	Recurrence, time to healing, re-operation	Not reported
							Marsupialisation	23		
Sequeira <i>et al</i>	2018	Portugal	84	16.2	61	23	Minimally Invasive (Endoscopic)	21	Recurrence, complications, healing time,	11.9
							Midline primary closure	63		
Serour <i>et al</i>	2002	Israel	34	16.4	20	14	Midline primary closure	34	Recurrence, complications	36
Smith <i>et al</i>	2015	UK	41	15	22	19	Minimally invasive (Fibrin glue obliteration)	10	Recurrence, wound dehiscence incidence	32
							Midline primary closure	5		
							Off midline primary closure	26		
Speter <i>et al</i>	2017	Israel	42		30	12	Minimally invasive (Minimal incision)	21	Recurrence, postoperative functional outcome, Reoperation rate	28
							Other (not well defined)	21		50
Umesh <i>et al</i>	2018	USA	22	16	18	4	Off midline primary closure	22	Recurrence, complications, healing time, reoperation rate, patient satisfaction	44

Yamout <i>et al</i>	2009	USA	16	16	9	7	Off midline primary closure	54	Recurrence, complications	11
Yildiz <i>et al</i>	2014	Turkey	40	15.2	18	22	Midline primary closure	8	Complications, recurrence	5
							Off midline primary closure	32		
Zagory <i>et al</i>	2016	USA	60	15	25	35	Open healing	17	Recurrence, complication, length of stay, re-operation rate	53.8
							Midline primary closure	36		
							Off midline primary closure	7		

Table 2: Risk of bias

Study	Confounding	Selection	Classification of Intervention	Deviation from intended intervention	Missing Data	Measurement of Outcomes	Reported Result	Overall
Afsarlar (2013)	Moderate	Low	Low	Low	Low	Moderate	Moderate	Moderate
Arda (2005)	Moderate	Low	Low	Low	Low	Moderate	Moderate	Moderate
Ates (2018)	Serious	Low	Low	Low	Low	Moderate	Moderate	Serious
Braungart (2016)	Moderate	Low	Low	Serious	Low	Moderate	Moderate	Serious
Butter (2006)	Moderate	Low	Low	Low	Low	Moderate	Moderate	Moderate
Caniano (2005)	Moderate	Low	Low	Low	Low	Moderate	Moderate	Moderate
Esposito (2017)	Serious	Low	Low	Low	Low	Moderate	Moderate	Serious
Fike (2011)	Serious	Serious	Serious	Low	Low	Moderate	Moderate	Serious
Gendy (2011)	Serious	Low	Low	Low	Low	Moderate	Moderate	Serious
Gonzalez-Temprano (2011)	Moderate	Low	Low	Low	Low	Moderate	Moderate	Moderate
Lee (2008)	Serious	Serious	Low	Low	Low	Moderate	Moderate	Serious
Lukish (2009)	Serious	Serious	Low	Low	Low	Moderate	Moderate	Serious
Morden (2005)	Serious	Serious	Low	Low	Low	Moderate	Moderate	Serious
Mutus (2018)	Moderate	Low	Low	Low	Low	Moderate	Moderate	Moderate
Nasr (2011)	Serious	Moderate	Low	Low	Low	Moderate	Moderate	Serious
Ozcan (2017)	Moderate	Low	Low	Low	Low	Moderate	Moderate	Moderate
Prato (2018)	Moderate	Low	Low	Low	Low	Moderate	Moderate	Moderate
Rouch (2016)	Moderate	Serious	Low	Low	Low	Moderate	Moderate	Serious
Sequeira (2018)	Moderate	Serious	Low	Low	Moderate	Moderate	Moderate	Serious
Serour (2002)	Moderate	Low	Low	Low	Low	Moderate	Moderate	Moderate
Smith (2015)	Moderate	Serious	Low	Low	Low	Moderate	Moderate	Serious
Speter (2017)	Serious	Low	Serious	Low	Moderate	Moderate	Moderate	Serious
Umesh (2018)	Moderate	Moderate	Low	Low	Moderate	Moderate	Moderate	Moderate
Yamout (2009)	Moderate	Serious	Low	Low	Low	Moderate	Moderate	Serious
Yildiz (2014)	Serious	Low	Low	Low	Low	Moderate	Moderate	Serious
Zagory (2016)	Serious	Serious	Low	Low	Low	Moderate	Moderate	Serious

Table 3: Summary of results

Procedure	No of studies	Total No of patients	Recurrence		Wound complications		Wound Healing
			Pooled risk (%)	95% CI (%)	Pooled risk (%)	95% CI (%)	Range (median values - days)
Open Healing	6	191	26	15-38	21	9-36	38-92
Midline Primary closure	16	728	12	8-18	30	19-42	8-38
Off-midline primary closure	9	199	6	1-15	14	6-25	27
Excision and VAC	2	29	20	0-65	Not reported	Not reported	38-72
Minimally Invasive Techniques	6	150	7	1-16	Not reported	Not reported	21-30
Marsupialisation	3	53	6	0-22	Not reported	Not reported	6-41