Limberg flap versus Karydakis flap for treating pilonidal sinus disease: a systematic review and meta-analysis

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Background: The Limberg flap reconstruction and the Karydakis flap reconstruction are the 2 most used off-midline closure techniques in pilonidal sinus surgery. The current evidence is inconclusive as to which is the optimal technique. The aim of this systematic review and meta-analysis was to compare differences in outcomes between these 2 flap-based techniques.

Methods: We identified studies by a systematic literature search of the Embase, MEDLINE (PubMed), Cochrane Library and Google Scholar databases and studies selected as per the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) checklist. Only randomized controlled trials (RCTs) that compared the Limberg flap (standard or modified) and the Karydakis flap were included in this review.

Results: Operative time was shorter by 7 minutes in the Karydakis group than in the Limberg group (mean difference 7.00 min, 95% confidence interval [CI] 0.53 to 13.48). The seroma formation rate was significantly higher in the Karydakis cohort (odds ratio [OR] 0.36, 95% CI 0.24 to 0.56); however, after excluding studies with a high risk of bias, the sensitivity analysis showed no significant differences in seroma formation rate between the 2 techniques (OR 0.76, 95% CI 0.31 to 1.85). Other outcomes of interest showed no significant differences between the Limberg and Karydakis techniques.

Conclusion: There were no significant differences between the Limberg and Karydakis techniques. Future RCTs with strict adherence to CONSORT guidelines will further elucidate the efficacy of these surgical procedures.

Contexte: Les reconstructions à l'aide de lambeaux de Limberg et de Karydakis sont 2 des techniques de fermeture décalées de la ligne médiane les plus utilisées pour la chirurgie du sinus pilonidal. Les preuves actuelles ne permettent pas de conclure à la supériorité de l'une par rapport à l'autre. Le but de la présente revue systématique/méta-analyse était de comparer les différences de résultats entre ces 2 techniques de lambeaux.

Méthodes: Nous avons recensé des études au moyen d'une interrogation systématique des bases de données Embase, MEDLINE (PubMed), bibliothèque Cochrane et Google Scholar et les études sélectionnées à l'aide de la liste de vérification PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses). Seuls les essais randomisés et contrôlés (ERC) qui comparaient les lambeaux de Limberg (standard ou modifié) et de Karydakis ont été inclus dans cette revue.

Résultats: Les interventions ont duré 7 minutes de moins dans le groupe Karydakis que dans le groupe Limberg (différence moyenne 7,00 min, intervalle de confiance [IC] de 95 % 0,53 à 13,48). Le taux de formation de séromes a été significativement plus élevé dans la cohorte Karydakis (rapport ces cotes [RC] 0,36, IC de 95 % 0,24 à 0,56); par contre, après avoir exclu les études comportant un important risque de biais, l'analyse de sensibilité n'a montré aucune différence significative quant au taux de formation de séromes entre les 2 techniques (RC 0,76, IC de 95 % 0,31 à 1,85). Les autres paramètres d'intérêt n'ont montré aucune différence significative entre les techniques de Limberg et de Karydakis.

Conclusion: On n'a noté aucune différence significative entre les techniques de Limberg et de Karydakis. De prochains ERC strictement conformes aux lignes directrices CONSORT permettront de préciser davantage l'efficacité de ces interventions chirurgicales.

he reported incidence rate of pilonidal disease is 25 per 100 000 people. Initially, the pathogenesis was considered to be congenital.² Today, however, the theory that it is acquired is more widely accepted. Work by Georgios Karydakis,^{3,4} who highlighted 3 main factors contributing to pilonidal disease (loose hair, an external force that facilitates insertion of hair into the skin and an underlying vulnerability of natal cleft skin), had a pivotal role in this paradigm shift. The first 2 factors are related to personal hygiene and lifestyle, and their modulation can influence the initiation, development and recurrence of pilonidal disease. Armstrong and Barcia⁵ reported that improved hygiene, an active lifestyle and hair control in the natal cleft area decreased the need for surgical procedures and resulted in faster return to work. However, the third factor can be modified only surgically. The rhomboid, well-vascularized transposition Limberg flap was first proposed by the Russian maxillofacial surgeon Alexander Limberg (1894-1974), author of the fundamental work in plastic surgery "Mathematical principles of local plastic procedures on the surface of the human body."6,7

Patients with chronic pilonidal sinus disease are usually candidates for flap procedures. A chronic disease can be defined as any condition with constant or recurrent symptoms observed over several months, without any indication of spontaneous healing.8,9 Flap-based Limberg and Karydakis techniques both aim to achieve off-midline closure of the surgical defect to flatten the natal cleft. In both techniques, methylene blue is injected in the pilonidal sinuses to guide the excision. The Karydakis technique consists of an asymmetric elliptical excision of the affected area (Fig. 1). The upper and lower poles of the ellipse are placed about 2 cm to the side of the midline. Subsequently, after full-thickness mobilization of the contralateral surgical margin and fixation of the base of the flap to the sacral fascia, the skin edges are sutured off the midline. In the modified Karydakis technique, fixation to the sacral fascia is not required. The Limberg technique consists of a rhomboid-shaped excision of the affected area down to the sacral fascia (Fig. 1). The cephalic and caudal apex of the rhombus are placed about 2 cm to the side of the midline. A fasciocutaneous rhomboid flap is then created and transposed so the defect can be covered without any tension. In contrast, the classic Limberg flap consists of a symmetric rhomboid excision with its apices placed on the midline.3,6-8 Karydakis reported a wound complication rate of 8% and a recurrence rate of 2% for 7471 procedures.^{3,4} The reported recurrence rate with the Limberg technique is 5%.8

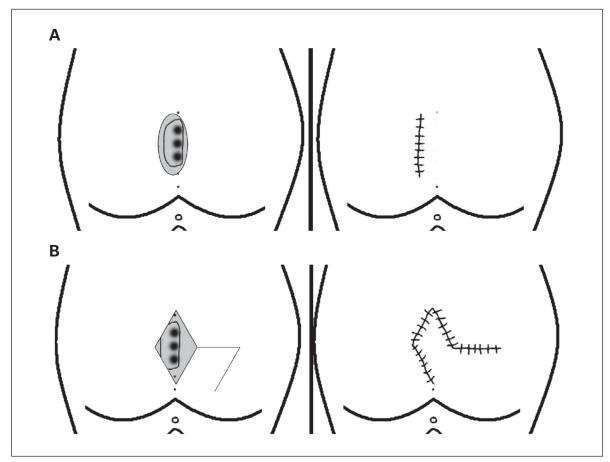


Fig. 1. (A) Karydakis technique. (B) Limberg technique with rhomboid transposition flap.

The existing evidence is inconclusive as to which technique is the optimal treatment for pilonidal sinus disease. To help clarify this issue, we performed a systematic review to compare the Limberg and Karydakis flap techniques through meta-analysis of randomized controlled trials (RCTs). The primary outcomes investigated were wound infection, wound dehiscence and time to return to work. Hematoma formation, length of hospital stay, recurrence and patient satisfaction rate were secondary outcomes.

METHODS

This systematic review and meta-analysis was carried out in accordance with the guidelines set out in the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) checklist.

Literature search

We performed a systematic literature search of articles published in the last 20 years using the Embase, MEDLINE (PubMed), Cochrane Library and Google Scholar databases and free text and MeSH search terms (standard or modified Limberg flap; standard or modified Karydakis flap; pilonidal sinus surgery and disease; randomized or nonrandomized controlled trial). We also performed a grey literature search on the ClinicalTrials.gov Web site. We manually checked references of the retrieved articles for further analysis. Disagreements between the authors were resolved by consensus.

Study selection and inclusion and exclusion criteria

Only RCTs that compared standard or modified Limberg flap and standard or modified Karydakis flap in pilonidal sinus surgery were included in the study. All nonrandomized studies, reviews and narrative articles were excluded.

Data extraction and outcomes

Two reviewers (P.G. and E.B.) independently extracted the following summary data for the included studies: name of authors, patient age, operative time, wound infections, wound dehiscence, hematoma, seroma, recurrences, length of hospital stay, time to return to work and patient satisfaction.

Risk of bias assessment of included studies

The 2 authors independently assessed the risk of selection bias, attrition bias, detection bias, performance bias and reporting bias for each included study. We categorized the risk of bias according to the *Cochrane Handbook for Systematic Reviews of Interventions*¹⁰ as high, low or unclear.

Definitions

We defined operative time as time from scalpel to the skin until the last skin stitch. Wound infections and seromas were reported based on clinical findings. Culture results and wound dehiscence (partial or complete) were defined by the individual study authors. We defined length of hospital stay as the number of days from the operation day until the day of discharge. Patient satisfaction was reported as described by each of the included studies' authors.

Statistical analysis

We conducted statistical analysis using Review Manager 5.3 software (Cochrane Collaboration). Heterogeneity was assessed through the I^2 test, and cut-off values of 25%, 50% and 75% were considered low, moderate and high, respectively. For I^2 values above 25%, both fixed- and random-effects models were used and the results compared between them. In cases in which the I^2 value was 25% or less, we used fixed-effects models throughout.

We analyzed dichotomous variables based on odds ratios (ORs) with 95% confidence intervals (CIs). For the analyzed outcomes, the reference categories were selected so that OR < 1 favoured the Limberg technique. We combined continuous variable based on both the mean difference and standardized mean difference. For studies that did not report the means and variances for the 2 groups, we estimated these values from the median, range and sample size where possible, using the technique described by Hozo and colleagues. In all analyses, the significance level was set at p < 0.05.

Sensitivity analysis

We conducted analyses of primary and secondary outcomes using both random-effects and fixed-effect models in order to assess the impact of heterogeneity on the results. In addition, we performed subgroup analysis of the studies in which the modified flap technique was used. We did not estimate publication bias because fewer than 10 studies were included in the review.¹³

RESULTS

Characteristics of included studies

The search strategy yielded 93 papers. A further 3 papers were obtained via manual reference searching, for a total of 96 studies. After removal of duplicates, 87 studies remained. Their abstracts were screened and 41 studies were removed as they did not fulfill the inclusion criteria. The full articles of the remaining 46 studies were read. Twenty-six 1-arm studies and 11 retrospective analyses were excluded, leaving 9 studies¹⁴⁻²² that met the criteria

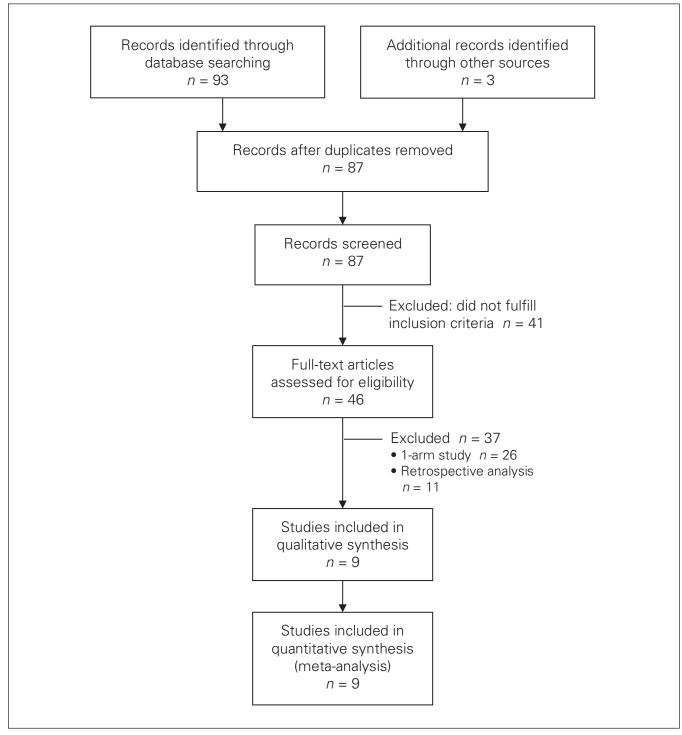


Fig. 2. Flow diagram of literature search strategy.

for inclusion in this systematic review and meta-analysis (Fig. 2).

A total of 1421 patients were involved in the selected studies, of whom 773 (54.4%) underwent Limberg and 648 (45.6%) underwent Karydakis flap reconstruction. Demographic characteristics were similar between the 2 groups (Table 1). All studies included patients with chronic pilonidal disease. Five studies compared the modified Limberg

flap to the Karydakis flap;^{15,17–19,21} those results were similar to those for the total sample (Table 2). Bali and colleagues¹⁹ enrolled only patients with recurrent disease. The most commonly used excluding criteria of the RCTs were recurrent disease, American Society of Anesthesiologists physical status class 3 or greater, diabetes, drug or alcohol addiction, renal failure and immunosuppression. Patients with laterally extended sinuses unsuitable to be included in

Table 1. Study characteristics*	character	ristics*										
Study/year/ country	No. of patients	Age, yr, mean ± SD	Operative time, min, mean ± SD	No. (%) with wound infection	No. (%) with wound dehiscence	No. (%) with hematoma formation	No. (%) with seroma formation	Type of drain	Length of hospital stay, d, mean ± SD	Time to return to work, d, mean ± SD	Patient satisfaction rate, mean score ± SD or no. (%)	No. (%) with recurrence
Ahmed et al., ²² 2017, Pakistan	75 75	32.2 ± 9.8 33.6 ± 9.7 p = 0.2	N W	E Z	E S	<u>د</u> 2	N W	Redivac	3.97 ± 0.71 2.93 ± 0.66 p < 0.001	15.53 ± 1.22 13.13 ± 1.15 \$\rho < 0.001\$	<u>د</u> 2	œ Z
Khan et al., ²¹ 2016, Pakistan†	06 06	42.7 ± 14 43.4 ± 16 p = 0.2	E N	11 (12) 30 (33) p = 0.001	N N	Œ Z	6 (7) 20 (22) p = 0.003	E N	N R	æ Z	œ Z	æ Z
Bali et al., ²⁰ 2015, Turkey	37	25 ± 5 24 ± 5 p = 0.2	54 ± 5 48 ± 5 $\rho = 0.001$	4 (11) 8 (24) p = 0.001	1 (3) 2 (6) p = 0.6	8 (22) 3 (9) p = 0.004	3 (8) 4 (12) $\rho = 0.8$	B-Vak	1.44 ± 0.5 3 ± 0.75 p = 0.001	8 ± 1.5 17 ± 1.5 p < 0.001	2.88 ± 0.16 1.43 ± 0.4 p < 0.001	(0) 0
Tokac et al., ¹⁹ 2015, Turkey†	46	29.28 ± 85 28.35 ± 85 p > 0.05	44.5 ± 6.6 42.9 ± 6.2 p > 0.05	3 (7) 3 (7) p > 0.05	N N	Œ Z	Z Z	Placed, type NR	1.06 ± 0.3 1.03 ± 0.17 p > 0.05	20.61 ± 7.89 23.29 ± 6.42 p < 0.05	œ Z	3 (7) 2 (4) p > 0.05
Arslan et al., ¹⁸ 2014, Turkey‡	96	26.5 ± 5.9 24.7 ± 5.1 p = 0.08	51.1 ± 6.8 50.9 ± 7.3 p = 0.08	2 (2) 6 (7) p < 0.05	2 (2) 14 (15) ρ < 0.001	4 (4) 3 (3) $\rho = 0.9$	5 (5) 18 (20) ρ < 0.05	Jackson-Pratt	1.3 ± 0.5 1.3 ± 0.4 p = 0.5	20.8 ± 6.5 19.1 ± 3.4 ρ = 0.7	71 (74) 64 (70) $\rho = 0.8$	6 (6) 10 (11) p = 0.003
Arslan et al., ¹⁸ 2014, Turkey†	108	24.7 ± 5.1 24.7 ± 5.1 $\rho = 1.0$	52.9 ± 7.3 50.9 ± 7.3 p = 0.08	5 (5) 6 (7) p = 0.3	4 (4) 14 (15) ρ < 0.002	3 (3) 3 (3) $\rho = 1.0$	8 (7) 18 (20) p = 0.002	Jackson-Pratt	1.3 ± 0.4 1.3 ± 0.4 $\rho = 1.0$	19.8 ± 4.6 19.1 ± 3.4 ρ = 1.0	84 (78) 64 (70) $\rho = 0.7$	2 (2) 10 (11) $\rho = 0.002$
Bessa, ¹⁷ 2013, Egypt†	09	23 ± 2.25 23 ± 6.25 p = 0.4	52 ± 3.5 33 ± 3 p = 0.001	3 (5) 2 (3) $\rho = 1.0$	11 (18) 6 (10) $\rho = 0.003$	Œ Z	0 (0) 3 (5) $\rho = 0.2$	18 French	Z Z	Œ Z	43 (72) 58 (97)	2 (3) 1 (2) $\rho = 0.6$
Ates et al., ¹⁶ 2011, Turkey	134	W.	50.14 ± 6.964 2.32 ± 8.64 p = 0.001	8 (6) 4 (3)	14 (10) 8 (6)	3 (2) 1 (1) $\rho = 0.9$	3 (2) 1 (1) $\rho = 0.9$	Placed, type NR	3.80 ± 1.19 3.40 ± 0.94 $\rho = 0.03$	8.55 ± 1.18 8.45 ± 1.16 p = 0.5	3.16 ± 1.40 7.08 ± 1.7 p = 0.001	9 (7)
Can et al., ¹⁵ 2010, Turkey†	77	22 ± 5.25 22 ± 2.75 p = 0.6	52.8 ± 164 0.4 ± 2.5 p = 0.01	3 (4) 3 (4) $\rho = 1.0$	1 (1) $1 (1)$ $p = 1.0$	$1 (1)$ $1 (1)$ $\rho = 1.0$	3 (4) 3 (4) p = 1.0	On surgeon's preference	4.8 ± 2.6 5.5 ± 2 p = 0.1	21.5 ± 8.5 18.8 ± 7.7 p = 0.09	70 (91) 60 (88) p = 1.0	4 (5) 3 (4) $\rho = 1.0$
Ersoy et al., ¹⁴ 2009, Turkey	20	27.8 ± 6.5 25.8 ± 6.4	Z.	4 (8) 13 (26) p = 0.02	AZ AZ	æ Z	Z Z	Placed, type NR	N R	14 ± 10.25 15 ± 10 p = 0.3	æ Z	a Z
Pooled difference $(n = 1421)$	773	MD 0.47 (95% CI 0.18 to 1.11)	MD 7.00 (95% CI 0.53 to 13.48)	OR 0.6 (95% CI 0.41 to 1.4)	OR 0.60 (95% CI 0.20 to 1.81)	OR 1.6 (95% CI 0.76 to 3.51	OR 0.36 (95% CI 0.24 to 0.56)	I	MD -0.07 (95% CI 0.45 to 0.30)	MD 0.33 (95% CI 0.99 to 1.66)	OR 0.83 (95% CI 0.34 to 2.01)	OR 0.88 (95% CI 0.38 to 2.04)
CI = confidence interval; MD = mean difference; NR = not reported; OR = odds ratio; SD = standard deviation.	val; MD = me	an difference; NR =	not reported; $OR = c$	odds ratio; SD = sta	andard deviation.							

CI = confidence interval: MD = mean difference; NR = not reported; OR = odds ratio; SD = standard dev Throughout table, first value is for Limberg flap, second value is for Karydakis flap.

†Modified Limberg flap.

‡Standard Limberg flap.

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Outcome	No. of studies/ patients	Estimated effect (95% CI)	P, %
Total sample			
Age ^{13–21}	9/1244	MD 0.47 (-0.18 to 1.11)	0
Operative time ^{14–19}	7/1082	MD 7.00 (0.53 to 13.48)	99
Wound infection ^{13–21}	8/1182	OR 0.65 (0.41 to 1.04)	18
Wound dehiscence ^{14–19}	6/990	OR 0.60 (0.20 to 1.81)	72
Hematoma ^{14–19}	5/870	OR 1.63 (0.76 to 3.51)	0
Seroma ^{14–20}	7/1170	Peto OR 0.36 (0.24 to 0.56)	19
High-quality seroma studies ^{14–16,19}	4/604	Peto OR 0.76 (0.31 to 1.85)	25
Recurrence ^{14–19}	6/1011	OR 0.88 (0.38 to 2.04)	94
Length of hospital stay ^{14–19}	7/1112	MD -0.07 (-0.45 to 0.30)	97
Time to return to work ^{14–19}	8/1212	MD 0.33 (-0.99 to 1.66)	94
Patient satisfaction rate ^{14,19}	6/990	OR 0.83 (0.34 to 2.01)	75
Subgroup analysis, modified Limb	erg flap		
Wound infection ^{14,17,18,20}	4/556	OR 0.91 (0.42 to 1.94)	0
Wound dehiscence ^{14,17,18}	3/464	OR 0.70 (0.13 to 3.84)	75
Seroma ^{14,17,18,20}	4/644	OR 0.33 (0.99 to 0.57)	0
Recurrence ^{14,17,18,20}	4/556	OR 0.77 (0.22 to 2.63)	48
Length of hospital stay ^{14,17,18}	3/436	MD -0.00 (-0.12 to 0.12)	45
Time to return to work ^{14,17,18}	3/436	MD 0.37 (-2.04 to 2.77)	72

Table 3. Risk o	of bias			
		Type of bia	ns; risk	
Study	Sequence generation	Allocation concealment	Incomplete outcome data	Blinding
Ahmed et al. ²²	Unclear	High	High	High
Khan et al. ²¹	Unclear	High	High	High
Bali et al. ²⁰	High	High	High	High
Tokac et al. ¹⁹	Unclear	Low	High	High
Arslan et al. ¹⁸	Unclear	Low	High	High
Bessa ¹⁷	Low	Low	Unclear	Low
Ates et al.16	Unclear	Low	High	High
Can et al. ¹⁵	Unclear	Low	High	High
Ersoy et al.14	High	High	High	High
Pooled data	Low 1	Low 5	Low 0	Low 1

the fusiform excision of the Karydakis procedure were transferred to the Limberg procedure.¹⁵

Study quality

Generally, RCT quality was poor (Table 3). There was a widespread high risk of inadequate blinding and incomplete outcome data. Allocation was adequately concealed in 5^{16–19} of the 9 studies. Only 1 study¹⁷ adequately addressed the sequence generation and blinding of outcome assessors, and none of the studies performed an intention-to-treat analysis for the missing data.

Outcomes of interest

Operative time was 7 minutes shorter in the Karydakis group than in the Limberg group (mean difference

7.00 min, 95% CI 0.53 to 13.48), although heterogeneity was extremely high (P = 99%). The seroma formation rate was significantly higher in the Karydakis group than in the Limberg group (OR 0.36, 95% CI 0.24 to 0.56, P = 19%) (Fig. 3). However, subgroup analysis including only studies with a low risk of bias showed no statistically significant difference in seroma formation rate between the 2 techniques (OR 0.76, 95% CI 0.31 to 1.85, P = 25%) (Fig. 3). For all other outcomes (wound infection/dehiscence, hematoma, recurrence, length of hospital stay and patient satisfaction), no significant differences were found between the 2 procedures (Table 2).

Sensitivity analysis

Subgroup analysis of the studies including the modified Limberg flap did not show any differences compared to the total sample of studies. The differences found in seroma formation rates disappeared when studies with high risk of bias were excluded (Fig. 3, Table 2). No discrepancies were found between the results produced with fixed-effects and random-effects models.

Discussion

This systematic review and meta-analysis showed no remarkable differences between the Limberg and Karydakis procedures in rates of wound infection/dehiscence, hematoma, recurrence and patient satisfaction, or length of hospital stay. There were only 2 differences between the 2 techniques: operative time and seroma formation rate. In the analysis, a modest difference in operative time was

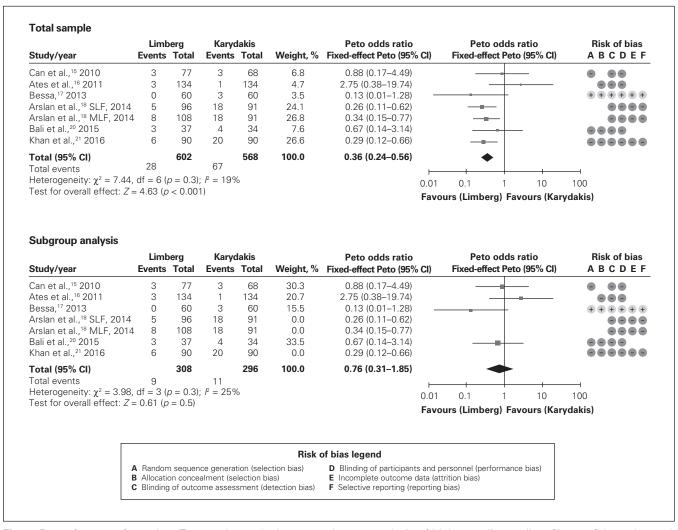


Fig. 3. Rate of seroma formation. Top: total sample; bottom: subgroup analysis of higher-quality studies. CI = confidence interval; SLF = standard Limberg flap; MLF = modified Limberg flap.

noted: the procedure time was 7 minutes shorter for the Karydakis group than for the Limberg group. In a clinical setting, however, this is unlikely to be of any importance. Previous authors have attributed this difference in operative time to 2 factors: first, the classic Karydakis flap does not require extensive mobilization, and second, it does not require fixation at the sacral fascia, ^{15,17} thereby reducing the amount of operative time required. One potential confounding factor with this finding may be the type of anesthesia used in each study. However, it was not possible to retrieve this information adequately for every study, which made it impossible to determine whether anesthetic type did indeed influence operative time.

Seroma formation rates were slightly lower in the Limberg group than in the Karydakis group. However, a subgroup analysis including only higher-quality studies did not show differences in seroma formation rates between the 2 procedures. Arslan and colleagues¹⁸ reported the highest difference in seroma formation rate between the Limberg and Karydakis procedures (5% and 20%, respec-

tively). However, their study contained incomplete data regarding dislodgement of drains, which made it difficult to control for confounders. In the same study, there is contradictory information regarding the length of hospital stay and the duration of drainage and follow-up. Overall, the patients had a mean length of hospital stay of 1.3 (standard deviation 0.4) days and a duration of drainage of 3.1 (standard deviation 1.3) days. The authors reported that some patients were discharged with their drain in situ. However, this proportion of patients was not reflected in the study's mean difference and standard deviation of the duration of drainage. Milone and colleagues'23 meta-analysis of the role of drainage after excision and primary closure revealed that the routine use of drains does not have a statistically significant advantage regarding rates of seroma formation, infection or recurrence.

All other outcomes (wound infection, wound dehiscence, hematoma, recurrence, length of hospital stay and patient satisfaction) in the current study were similar between the Limberg and Karydakis groups.

Enriquez-Navascues and colleagues²⁴ compared different surgical techniques for chronic pilonidal disease using traditional meta-analysis. By default, traditional meta-analyses can be used only to compare 2 treatment approaches. When 3 or more treatments are to be compared, network meta-analysis using either Bayesian or frequentist statistics must be used.²⁵

Limitations

The main limitation of this review is that, although the included studies were RCTs, most were of relatively low quality and had an unclear or high risk of bias: of the 9 studies, only 1 was graded as having a low risk of bias. Also, the studies' populations were likely heterogeneous, as shown by the high I^2 value in the analysis of outcome of interests. The protocols, inclusion criteria, surgical technique and follow-up periods also differed, which make the findings not only statistically but also clinically heterogeneous. In this context, the results of this meta-analysis must be interpreted with caution.

CONCLUSION

Even taking the study limitations into account, the Limberg and Karydakis flap operations in adequately selected patients with pilonidal disease do not show any significant differences between them. Future RCTs should have strict selection criteria, predefined surgical outcome measures, blind outcome assessors, common methods of outcome assessment and follow-up periods of 2–3 years, and must show strict adherence to standards recommended in the CONSORT guidelines.²⁶ These features will help further elucidate the efficacy of these 2 surgical procedures.

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Competing interests: None declared.

Contributors: P. Gavriilidis designed the study and wrote the article. Both authors acquired and analyzed the data, reviewed the article and approved the final version for publication.

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