Gender-specific prevalence of pilonidal sinus disease over time: A systematic review and meta-analysis

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Abstract

Background: Gender-specific risk factors have been suggested to promote a fourfold higher incidence of pilonidal sinus disease (PSD) in male as compared to female patients. However, in recent decades there has been an apparent shift towards an increasing prevalence of PSD in women, as body weight and other risk factors influence the disease. We aimed at determining whether PSD prevalence actually changed in men and women over time.

Methods: Following PRISMA guidelines (PROSPERO ID: 42016051588), databases were systematically searched. Papers reporting on PSD published between 1833 and 2018 in English, French, German, Italian and Spanish containing precise numbers of male and female participants were selected for analysis. Gender-specific prevalence of PSD over several decades was the main outcome measure.

Results: We screened 679 studies reporting on 104 055 patients and found that the male/female ratio in patients with PSD has remained constant over time, with women being affected in about 20% of all PSD cases ($I^2 = 96.18\%$; meta-regression $p < 0.001$).

Conclusion: While the prevalence of PSD has risen over the past decades, the ratio between affected males and affected females has remained constant, with women invariably representing about 20% of patients despite wide ranging socioeconomic and behavioural changes.

INTRODUCTION

Pilonidal sinus disease (PSD) is commonly associated with young men. While the first woman patient with PSD was reported in 1880 by Hodges,[1,2] the disease was classically described as a disease of the 'lower back' occurring in woodsmen with respective gluteal hair-growth.[1,2] In an analysis of a large cohort of PSD patients in the 1930s, the prevalence of women affected with PSD was around 20%.[3] An increasing number of studies reported during World War II noted a high prevalence of PSD in young male soldiers.[4,5] The military reported a lion's share of the disease burden,[6–8] and as mechanised warfare was new, the disease was referred to as 'Jeep disease'.[4] During this time period, the number of women serving in the army was very limited.[4,5]

Over time, PSD was increasingly diagnosed in both genders,[9–11] probably caused by occipital rather than local hair.[12–15] With gender specific dimensions gaining momentum in medicine for staff and patients alike,[16] treatment strategies must go beyond 'one approach for all' and strive to embrace the entire complexity involved.[17,18] While some authors reported a 4:1 male-to-female ratio,[19] others noted a lower number of men being affected with

PSD, at a ratio of 3:1,[20,21] or even 2:1.[22,23] Obedman reported an equal distributed.[24]

In the largest study to date of asymptomatic PSD patients, 365 of 31 497 male students (1.16%) and 24 of 21 367 female students (0.11%) were diagnosed with PSD, resulting in a 15:1 ratio, notably in asymptomatic patients.[24]

The discrepancies in reported results may be a result of factors such as regional differences,[25] study design or selection bias.[26,27] Therefore, we designed a systematic review and meta-analysis to determine whether there has been a shift in the prevalence of PSD reported in men and women since 1833. This knowledge may help to adapt treatment strategies, especially as the relative invasive primary open treatments traditionally used in men, may aesthetically not suit women.

MATERIALS AND METHODS

This meta-analysis was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA).[28] Our original search has been described previously.[25,29] In brief, we searched for the NCBI Medical Subject Heading (MeSH) terms 'pilonid*' and ('cyst' AND 'dermoid') in a number of search engines, including Embase, MEDLINE, Ovid, PubMed, PubMed Central, Scopus, and the Cochrane Central Register of Controlled Trials (CENTRAL).[25,29] Papers published between 1833 and 2018 in English, French, German, Italian and Spanish were included.[25,29] Evidence presented in other languages was included if follow-up occurred at specific times and definitive treatment strategies were provided.[25,29] The study is registered in the National Institute for Health Research's International prospective register of systematic reviews (PROSPERO number 42016051588). Data were handled using Microsoft Excel (Version 2016, Microsoft Corp., Redmond, WA, USA). Only publications with precise numbers of both genders were selected for analysis. No publications mentioned intersex or transsexual patients, so analysis focused solely on binary male or female characterisation and occurrence of the disease. Figure S1 displays the PRISMA diagram to find evidence of pilonidal surgery in men and women since 1833.[25,29]
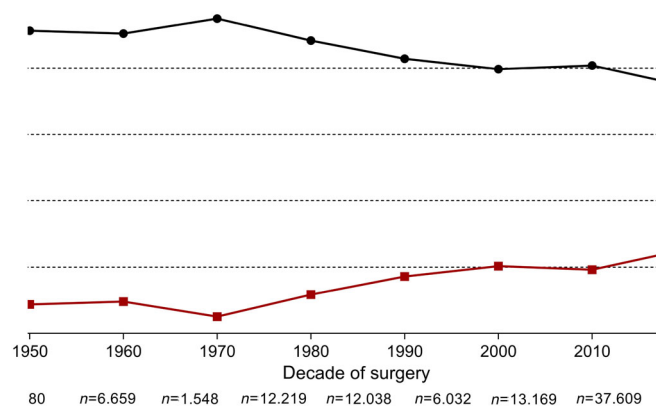

Fig 1. Raw gender distribution based on decade of surgery (proportions of patients). Uncorrected dataset. (—●—), male share of PSD patients between 1950 and 2010; (—■—), female share of PSD patients over 6 decades.

Statistical analysis

Crude proportions of female patients are calculated as the number of reported females divided by the total number of patients with reported gender for each study, across all studies, and stratified by decade. The share of female patients was pooled across studies with a random-effects meta-analysis using the method of DerSimonian and Laird as described previously.[26] Variance was stabilised using the Freeman-Tukey double arcsine transformation, after confirming that back-transformation led to estimates similar to those obtained with the exact likelihood approach.[30,31] Absolute heterogeneity was quantified using between-study variance τ^2. Relative heterogeneity (proportion of observed variation reflecting true variation rather than sampling error) was quantified with the I^2 statistic. Meta-regression was performed on the logit-transformed proportion to explore the relationship between the decade in which the study was performed and female gender, adjusting for the region in which the study was performed. The Cochran-Armitage test was used to examine whether there was a trend for an increase or decrease over time in manuscripts that only included male patients. All analyses were performed in STATA 16.0, using the 'metaprop_one', 'meta-funnel' and 'metareg' programmes.

RESULTS

Analysis of all available publications showed that the percentage of female PSD patients documented in the literature starts at far below 20% (Fig. 1). In the decade around 1950, data collected from more than 100 patients of both sexes were published worldwide. A decade later the number of patients was well over 1000. The female share varied between 5% and 10% until the 1970s, and increased steadily from that point on. The percentage of women doubled, to 20%, by 2010, and provisional numbers from the 2020s showed that, with more than 10 000 published patients, the percentage of women exceeded 25%. Where originally one in ten PSD patients reported were women, it is currently about 20%.
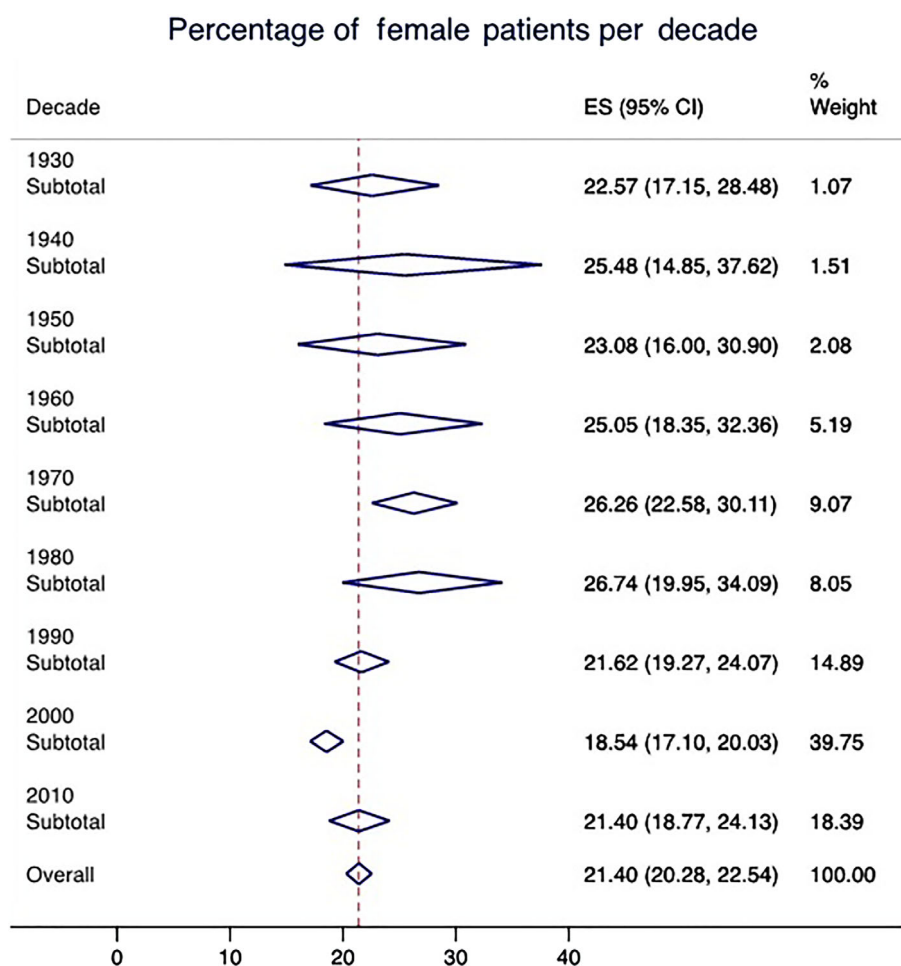
While the share of females suffering from PSD in the Mediterranean seems to have increased over time, it still lags behind the North American numbers, which have been increasing since the 1960s (Fig. S2).

With the exception of the 1930s (in which the estimate is only based on a few studies and is accordingly quite imprecise), there seems to be a trend towards an increase in female PSD patients over time.

As Table S1 illustrates, in the 1940s and 1950s (highlighted), more than half of the PSD studies only included men. This resulted in a lower share of females, as seen in the far-right column, lasting into the 1960s. In the 1990s and thereafter, the percentage of females was consistently above the mean of 17.48% (Table S1).

Meta-analysis showed that across all decades, 15.6% (95% confidence interval [CI] 14.3–17.0) of patients were female, with marked heterogeneity (between study variance $\tau^2 = 0.2$, $I^2 = 96\%$, $p < 0.001$) and with apparent differences between decades. Meta-regression confirmed a significant association between decade and the percentage females ($p = 0.002$), suggesting that the proportion of females indeed increased over time, which may at least in part

Fig 2. Forest plots of the proportion of female pilonidal sinus disease patients reported in publications, listed by decade/the decade's start respective. Our meta-analysis reveals an overall pooled percentage of females of 21.4% (95% confidence interval [CI] 20.3–22.5) across all decades. A priori exclusion of studies selectively reporting on male or female patients only, reveals that there is no evidence of an increase in female patients over time.

Percentage of female patients per decade

Decade	ES (95% CI)	% Weight
1930 Subtotal	22.57 (17.15, 28.48)	1.07
1940 Subtotal	25.48 (14.85, 37.62)	1.51
1950 Subtotal	23.08 (16.00, 30.90)	2.08
1960 Subtotal	25.05 (18.35, 32.36)	5.19
1970 Subtotal	26.26 (22.58, 30.11)	9.07
1980 Subtotal	26.74 (19.95, 34.09)	8.05
1990 Subtotal	21.62 (19.27, 24.07)	14.89
2000 Subtotal	18.54 (17.10, 20.03)	39.75
2010 Subtotal	21.40 (18.77, 24.13)	18.39
Overall	21.40 (20.28, 22.54)	100.00

explain the between-study heterogeneity. However, studies including only male patients may have biased this result, as the percentage of females is zero by study design. The same is true for the few studies reporting only women. The number of studies reporting only males has decreased over time (Mann–Whitney U test for the comparison of the time-distribution between studies reporting only males versus the other studies $p = 0.0051$, Cochran-Armitage test for a trend in studies reporting only males across decades $p < 0.001$). This may explain why the number of females seems to increase over time.

As a consequence, all $n = 163$ studies reporting only men ($n = 155$) or only women ($n = 8$) were excluded.

There is no longer an evident trend (Table S2). The percentage of females over the decades hovers around the total mean % of females, without any evident trend over time.

Meta-analysis now shows an overall pooled percentage of females of 21.4% (95% CI 20.3–22.5) across all decades. Excluding studies that led to a biased estimate by study design halved the between-study variance ($\tau^2 = 0.1$), and meta-regression no longer shows a significant association between decade and the percentage of females ($p = 0.908$). Thus, after a priori excluding studies selectively reporting only males or females, there is no evidence of an increase in female patients over time. However, despite a marked

reduction in the absolute dispersion of percentages across studies, the range of reported female proportions is still wide, and a high relative heterogeneity remains ($I^2 = 93\%$). This might in part be explained by the fact that the proportion of female patients differs by geographic region.[25]

Figure 2 displays the Forest Plot, in which subgroups (decade) are summarised. In addition, our data show that the proportion of PSD in female patients is not distorted by study size (Fig. S3). Please note that a separate x-log is used to enlarge the area of studies from 1 to 200 patients, where most PSD studies are located.

DISCUSSION

We found that the distribution of PSD between men and women remains constant. Over the past decades, the prevalence of PSD has risen in both sexes in parallel, with studies especially with larger involvement of women now being increasingly reported.[32] Our data indicate that there is an PSD increase both in North American and Mediterranean countries.[25]

A number of factors may play a role in greater awareness of PSD among women.[26] First, an increased level of health awareness in both sexes might have led to a higher number of women reportedly

seeking surgical therapy for PSD.[26] While women's and men's life-styles might become more similar, no lifestyle related factors have indicated an increase in PSD prevalence over the short or long term.[27,33,34] Yet, data from early studies might be biased by the fact that the incidence of the disease is not growing over time but rather women are able to access care and be diagnosed/ treated with greater frequency in the modern era.[26] Next, the number of women serving in the military, where PSD is common, has risen over the past two decades.[7,35,36] In contrast, in a 2017 survey of 19 013 Turkish participants aged between 17 and 28 years, Duman found that only 6% were women, which is 3–4 times lower than in the other reports.[35] In Israel, military service is mandatory for women as well—which is represented in Moshe Gips' large study cohort from 2008, in which 15.7% of the women reported that they suffered from PSD.[37]

Women differ from men in terms of their attitudes about health, their behaviour,[38] as well as their modes of communication and their coping mechanisms,[39–41] thus, there may also be differences between men and women in terms of lifestyle, physiology, prevalence, severity and perception of disease.[26] Primary open wound treatment is still widely used,[42] despite better methods being available in terms of aesthetics and recurrence rate.[43,44] The type of surgery itself may decrease quality of life in PSD patients.[45] Recurrence free outcome is of primary interest in terms of life quality and patient satisfaction,[46] while especially less compromising surgery with same outcome results matters for female patients in most cultures.[25,26] The knowledge of the substantial female participation in pilonidal disease now mandates more surgical reflection on the subject of recurrence free outcome plus non discomforting surgery results. Men will benefit from this more careful view and emerging trend as much as women with PSD.

A limitation is that the number of patients included in published data might not reflect the disease's prevalence. Given this epidemiological dilemma inherent in retrospective analyses, we studied a long period. We may have missed some data published in other sources. Still, we were able to assess the large main body of literature. In addition, even if it were different, the information not included would hardly affect the reliability of data from a large database exceeding 89 000 patients. Thus, we believe that the main trends stand fast. We further depended on the correctness of the diagnosis PSD by the authors of the assessed studies. Also, differences in access to care may exist for women often referred to as the social determinants of health even in developed countries and further studies will have to address social determinants of health in this regard. Finally, heterogeneity is relatively high for some data—too high to allow a meaningful estimate of the percentage of females—but there is no other way to estimate s than using the available studies. Heterogeneity is fine as long as it reflects real differences between studies that can be explored or discussed (such as regional differences). In fact, we do not know much about inclusion and exclusion criteria, and differences in females may simply reflect differences in sampling rather than 'true' differences. We addressed this by excluding all patients in studies that only reported males or females, but even then, there could be selection bias with respect to gender.

In summary, it is conceivable that women value quality of life and cosmetic outcome differently than men, which might influence the therapeutic approach and treatment decisions for PSD. While the prevalence of PSD has risen over the past three decades,[10,47] the ratio between affected males and affected females has remained constant, with women representing about 20% of patients.

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Author contributions

Markus Luedi: Conceptualization; data curation; formal analysis; methodology; supervision; visualization; writing-original draft; writing-review & editing. **Patrick Schober:** Conceptualization; data curation; formal analysis; investigation; methodology; resources; software; validation; writing-original draft; writing-review & editing. **Verena Stauffer:** Conceptualization; data curation; methodology; writing-original draft; writing-review & editing. **Maja Diekmann:** Conceptualization; data curation; formal analysis; methodology; project administration; writing-original draft; writing-review & editing. **Lukas Andereggen:** Conceptualization; writing-original draft; writing-review & editing. **Dietrich Doll:** Conceptualization; data curation; formal analysis; investigation; methodology; project administration; software; supervision; validation; visualization; writing-original draft; writing-review & editing.

Data availability statement

All data and calculations are available to readers upon request to the corresponding author.

Conflict of interest

None declared.

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Supporting information

Additional Supporting Information may be found in the online version of this article at the publisher's web-site:

Figure S1: Flow diagram based on the PRISMA checklist, documenting the approach used to find evidence of pilonidal surgery in men and women since 1833 as described previously.

Figure S2: Proportion of women from North America vs. women from the Mediterranean depending on decade of surgery (smoothed for four decades. Uncorrected dataset.

Figure S3: The proportion of PSD in female patients is not distorted by study size (note that a separate x-log is used to enlarge the area of studies from 1 to 200 patients, where most PSD studies are located).

Table S1: All patients with known gender, by decade. Total studies and male-only studies. Uncorrected dataset. 'Decade' indicates the first year of the period analysed.

Table S2: All patients with known gender, by decade; DATASET with all studies with evident selection bias excluded.