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Worldwide trends in pilonidal sinus disease recurrence rate

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Abstract

Background: With the emergence of new techniques and increasing knowledge about pilonidal sinus disease (PSD), there is curiosity regarding the trends in recurrence rate.
Objective: The objective of this study is to analyze trends in the recurrence rates associated with PSD treatments over the past decades, spanning from the discovery of PSD to the present. This analysis was conducted by examining the proportion of recurrent patients among all patients undergoing pilonidal sinus surgery.
Methods: A comprehensive search was performed in global literature databases, including PubMed, Embase, ScienceDirect, and others, to gather reported data on PSD recurrence rates from 1833 to the present. The collected articles were grouped and analyzed.
Results: The published patient group sizes ranged from $n = 6439$ patients with $n = 130$ cases of recurrent disease in 1930 to $n = 42{,}537$ patients with $n = 3548$ cases of recurrent disease in 2000. While the rate of recurrent patients seeking surgery varied between 2% in 1930 and 14% in 1940, no clear trend could be established. Notably, there was no significant decrease in the raw recurrence rate between 1930 and the past two decades. No limitations were identified in this study.
Conclusion: The findings suggest that the published recurrence rate of PSD is not decreasing. As the incidence of PSD is increasing, it is possible that the effectiveness of superior therapies is being overshadowed by unknown factors influencing the outcomes. Alternatively, it is plausible that more challenging cases are being published or that available superior therapies are not being adequately utilized. The next step would involve analyzing global trends in the choice of treatment for pilonidal sinus disease.

Keywords
Study trends · Pilonidal sinus · Therapy trends · Pilonidal sinus disease · Incidence

Introduction

The incidence of pilonidal disease on a global scale appears to be increasing [1, 2], with variations observed among different countries [3, 4] and genders [5]. This increase seems to be more pronounced in developing countries, although the reasons for this trend remain unknown. In higher-income countries, early onset of puberty has been associated with earlier onset of pilonidal sinus disease [2, 6], which, in turn, is linked to a higher recurrence rate of up to 50% [7]. With advances in understanding the generation and prevention of the disease [8–11], there have been more meta-analyses providing guidance on establishing effective follow-up procedures and identifying therapies that may reduce recurrence rates [12, 13]. These findings have been reflected in national guidelines [14–16]. However, the

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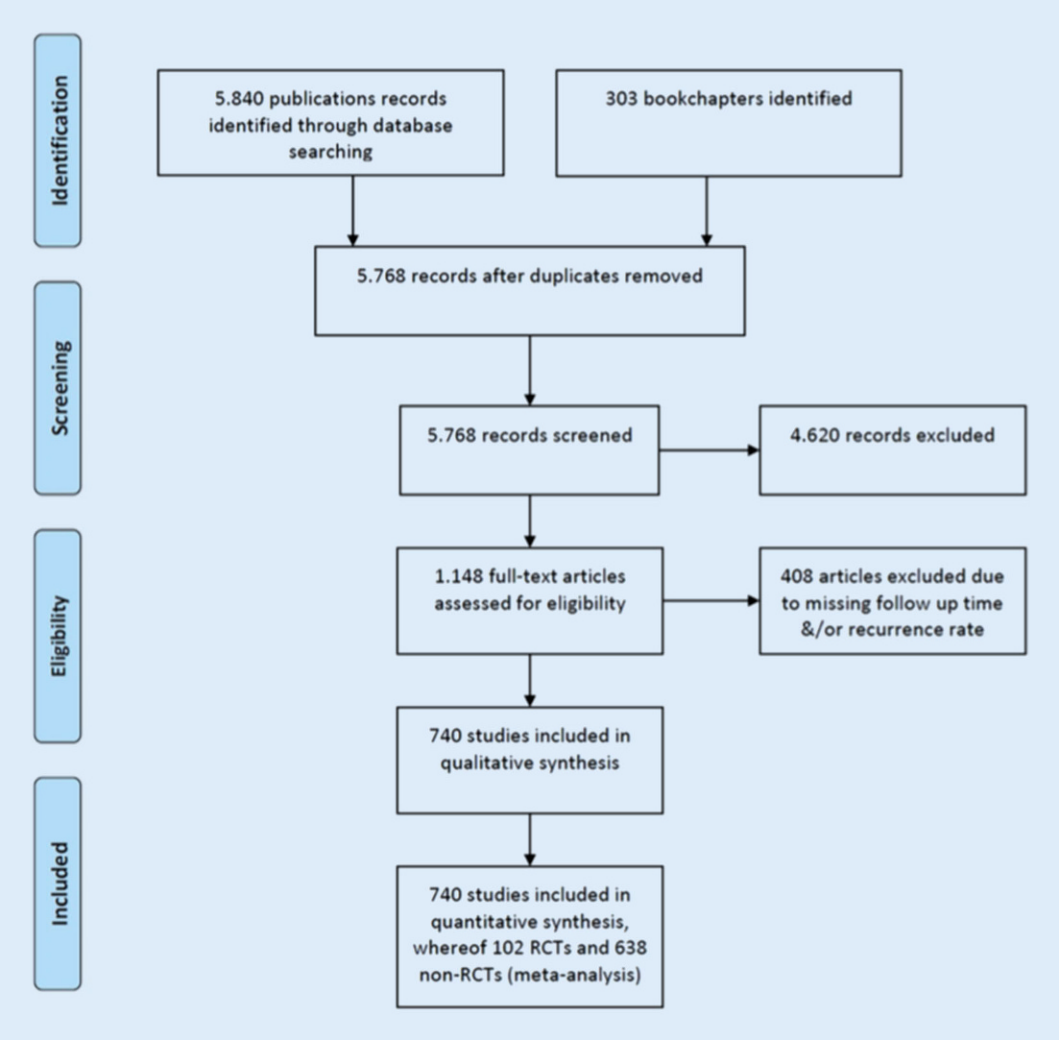

Fig. 1 ◀ PRISMA (Moher D et al. [20]) diagram of data harvesting, resulting in 740 studies with $n = 139{,}337$ patients in reference to Stauffer et al. [13, p. 1–27]

implementation of study knowledge into practice can be a lengthy process, if it occurs at all. Given these circumstances, our objective was to systematically investigate worldwide recurrence trends of pilonidal disease from 1833 to the present day.

Methods

Search strategy

To achieve this objective, we conducted an extensive search across various databases and platforms, including Medline, PubMed, PubMed Central, Scopus, Ovid, Embase, ScienceDirect, the Cochrane Central Register of Controlled Trials (Central), Google Scholar, ResearchGate, AWMF, and free-access websites. We used specific keywords such as "Pilon* AND Recurrence", "Pilonidal AND Sinus AND Recurrence", "Pilon* AND treatment" , "Pilonidal AND Sinus AND treatment", "Pilon* AND therapy" and "Pilonidal AND Sinus AND therapy" in our search. In addition, we performed an onsite Endnote research analysis of 5840 publications on pilonidal sinus disease and 303 book chapters, focusing on terms such as "recurrence" "recurr*", "therap*", "relaps*" and "recidiv*". Articles containing information on pilonidal sinus disease recurrence were carefully studied and analyzed. The obtained results were thoroughly examined and tabulated when appropriate.

Inclusion criteria

Inclusion criteria for the studies encompassed recurrence rates, the country of estimation, the number of patients with pilonidal sinus disease under care, the proportion of recurrent diseased patients undergoing care, and the year of data generation. Additional factors considered were the inclusion of military populations and the Mediterranean origin of the study population.

Exclusion criteria

Exclusion criteria included insufficient minimal dataset (as mentioned above), the presence of neoplastic disease [17–19] within the pilonidal sinus disease population, meta-analyses, inclusion of extrasacral pilonidal sinus disease, conditions mimicking pilonidal sinus disease, and duplicate publications.

Results

A total of 740 studies were analysed and have already been outlined in cooperation with Stauffer et al. [13, p. 1–28]. Equate

Table 1 Percentage of recurrent disease patients of all pilonidal sinus disease (PSD) operations published worldwide, listed for all decades from 1930 to 2020

Decade	Recurrent PSD (*n*)	All patients for surgery (*n*)	Recurrent PSD (%)
1930	150	982	15.3
1940	482	1788	27.0
1950	404	1386	29.1
1960	211	1296	16.3
1970	3079	7024	43.8
1980	460	2455	18.7
1990	1065	7659	13.9
2000	4201	24,330	17.3
2010	4747	25,327	18.7
2020[a]	302[a]	1058[a]	28.5[a]
Total	**15,102**	**7330**	**20.6**
Median	471	2122	18.7
Mean	1510	7331	22.9
SD	1788	9539	9.3

Please note that the decade of treatment, and here the midterm year, was used
[a]2020 decade not fully available

datasets were identified, as depicted in the PRISMA [20] diagram below (**Fig. 1**). These findings were computed and examined.

These studies encompassed a population of 73,309 patients over a span of ten decades, as illustrated in **Table 1**. Their results were tabulated and analyzed.

Study size per decade varies between $n = 982$ (in 1930) and $n = 25{,}327$ persons (in 2010; median $n = 2122$), with from $n = 150$ recurrences (in 1930) up to 4747 recurrences (in 2010) mentioned (median $n = 471$ recurrences). Thus, the share of recurrent disease seems to be varying between 13.9% (in 1990) and 43.8% (in 1970; **Fig. 2**). The largest variations can be seen between 1960 and the 1970 decade, comprising the years from 1950 to 1970.

Even if two decades are calculated together (not shown), the proportion of recurrent diseased patients arriving for therapy seems to undulate between 16 and 31%. According to these numbers, there is no clear upward or downward trend in recurrent disease arriving for further therapy.

Figure 3 investigates the group size and its possible influence on recurrent share. Scatter in recurrent PSD shares is to be seen below a group size of $n = 10{,}000$ patients per decade analyzed; here, shares of between 14 and 44% are evident. Above this benchmark, percentages of 17% (decade of 2000) and 19% (decade of 2010) can be seen. The last decade analyzed is 2020, with a group size of $n = 1058$ patients, as, obviously, time from recruitment/treatment to publication may take years.

Discussion

This study has enabled us to recognize that there was no downward trend in the recurrence rate during recent decades. This is somewhat astonishing, considering that we have become equipped with improved knowledge and therapies during the same period. What are the potential explanations for the lack of improvement in these therapeutic results?

As the incidence of the disease is rising, it is possible that the recurrence rate is also increasing. However, this negative effect may be masked by the improved use of better technologies and therapies. This will be analyzed in the following paragraphs to determine whether therapies have indeed improved.

Is there enough knowledge available regarding the therapies associated with the lowest recurrence rates? Yes, there is a wealth of knowledge on this subject. Off-midline closure was recommended as early as 1990 by Allen-Mersh and reiterated by Petersen in 2002, as well as Al-Khamis in 2010 [21]. The 2019 national guidelines from the USA [14], 2015 from Italy [15], and 2016 from [22] Germany all repeated this knowledge. However, the question remains: have these recommendations been followed? We know that the implementation of scientific and study knowledge into practice may take 10 years or longer, with a study-to-theatre time of about 10 years or more.

German pilonidal sinus disease guidelines were published in 2014 (and repeated in 2020). Research on PSD therapy changes in all three German-speaking countries followed in 2020 and 2021. It was observed that Germany [23], Switzerland [24], Austria [24], Denmark [25], the UK, and Ireland [26] have not fully adopted the best result therapeutic advice thus far. On the other hand, Australia, New Zealand, and Norway [27] have implemented changes from recurrence-prone procedures to modern low-recurrence procedures in a timely manner, despite not having their own national guidelines. The willingness for their adoption of best-known practice is quite impressive. Do procedures have to be modern? While some new procedures may be modern and fashionable, they may vary in terms of being chosen wisely or simply following trendy advice. For example, while pit picking is trendy, it is emerging that only a few studies can reproduce the impressive long-term results from Gips in Israel [28, 29], and worldwide pit-picking rates may have more than 40% recurrences at 5 years. Thus, the results of various surgical trends may be summarized as "keeping the recurrence rate stable" without improvement.

On the other hand, the pressure for cost effectiveness may have played a larger role, leading surgeons to opt for less costly and easy-to-administer outpatient treatments. Phenol is cheap and easy to handle, and pit picking is performed under local anesthesia in an outpatient clinic. Both therapies enable fast patient care but are associated with recurrence rates exceeding 40% at 5 years (phenol) and a 15% recurrence rate after 5 years following surgery (pit picking); both above the desired level of 2% recurrence rate per year of follow-up. So, if more patients have access to these methods as they are available and affordable, this would come with a higher recurrence rate within the population treated.

Pilonidal sinus disease (PSD) primarily affects younger individuals, specifically

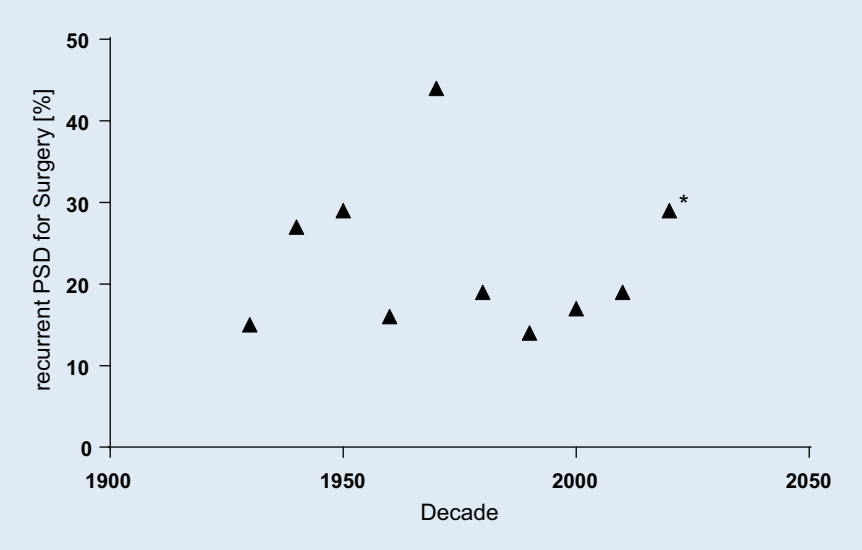

Fig. 2 ▲ Recurrent pilonidal sinus disease (*PSD*) patients arriving for surgery; worldwide published studies pooled per decade. *Asterisk* 2020 decade not fully available

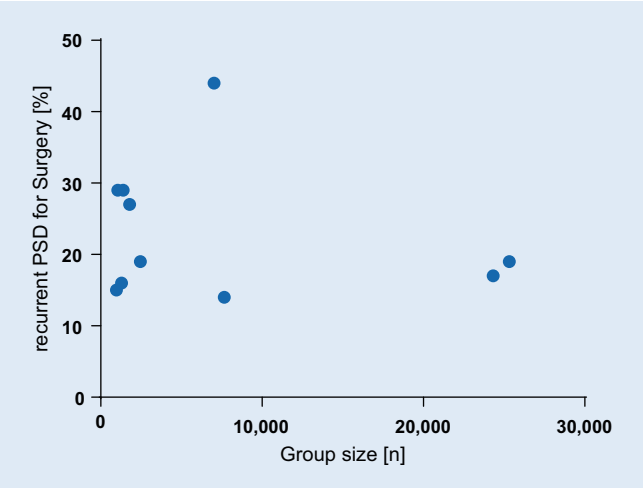

Fig. 3 ◀ Pooled decade group size versus share of recurrent pilonidal sinus disease (*PSD*) patients. Above $n = 10{,}000$ patients, recurrent rate variation seems to decrease

teenagers and those in their twenties, with the highest number of cases observed between the ages of 15 and 25 [2, 30]. A world with a larger population in this age group would consequently have a higher number of patients with PSD. However, the recurrence of the disease would only be influenced by the availability of surgical treatments, as previously documented. If healthcare services were to suddenly improve, the rate of recurrence could temporarily increase due to more patients receiving initial treatment. Subsequently, within a span of 2–5 years, a certain percentage of these patients would experience a relapse [31]. Although this phenomenon may impact the research, it is important to note that it typically occurs once in a given country and is not a continuous occurrence over an extended period. Moreover, the progress in healthcare varies among countries, and not all nations are able to synchronously enhance their healthcare systems. The countries with the highest prevalence of PSD are commonly referred to as developed countries, and their populations are aging and declining, which could potentially lead to a decrease in the occurrence of PSD in the future, but not necessarily in the proportion of recurrent cases.

Working with the recurrence rate necessitates addressing several critical aspects. Firstly, it should be noted that the majority of reported recurrence rates (RR) found in the worldwide literature, as well as those previously mentioned, are crude or raw recurrence rates. Since the recurrence rate is dependent on time, it is preferable to use a Kaplan–Meier analysis as the standard reporting method. This approach effectively avoids complications arising from variations in follow-up rates and follow-up durations.

Secondly, it is important to differentiate between RR which encompass recurrences following previous surgeries or new hair insertions, and wound healing disorders. There exists a subgroup of patients with primary open wounds (resembling ulcers) prior to surgery. These chronic wounds exhibit single or multiple midline defects and contain granulation tissue along with an abundance of hair. Such wounds continue to pose challenges even after surgical intervention, requiring specialized care. When surgical errors are also taken into account, such as unnecessarily large or near-anus excisions, the absence of dyes like methylene blue, or the exposure of the presacral fascia, the closure of wounds may be delayed for several years.

Thirdly, there appears to be a discernible inclination towards the publication of positive articles, particularly over the past three to four decades. Negative results often tend to be attributed to the deliverer (i.e., the author), which may lead to appropriate retribution in the form of rejection. Fortunately, in modern times, no one is subjected to severe consequences or harm for reporting unfavorable news.

In conclusion, our analysis indicates that the proportion of surgeries performed for recurrent PSD has not exhibited a significant reduction over the past 80 years, implying that the rate of recurrence has remained constant. Numerous studies have identified treatment practices that could be replaced with superior alternatives. However, based on our investigation, it remains uncertain whether the lack of progress in this regard is due to limited knowledge, inadequate education, inertia, or the pressure to prioritize cost effectiveness.

To verify whether higher healthcare expenditures in high-income countries result in lower recurrence rates, an analysis of the relationship between recurrence rates (RR) and gross domestic product (GDP) is warranted.

Outlook

It seems that there is still considerable scope for enhancing pilonidal sinus education, particularly in promoting informed decision-making within the surgical community, combined with the freedom to choose the best therapeutic for each patient. It is imperative that treatment results be bettered until a preventative solution for this disease becomes available.

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Declarations

Conflict of interest. D. Doll, M. Braun-Münker, C. Oetzmann von Sochaczewski and I. Iesalnieks declare that they have no conflicts of interest.

For this article no studies with human participants or animals were performed by any of the authors. All studies mentioned were in accordance with the ethical standards indicated in each case.

Zusammenfassung

Weltweite Trends beim Rezidiv eines Pilonidalsinus

Hintergrund: Mit dem Aufkommen neuer Techniken und zunehmendem Wissen über die Erkrankung an einem Pilonidalsinus wäre es interessant zu wissen, welche Tendenzen sich bei der Rezidivrate abzeichnen.
Ziel: Ziel der vorliegenden Studie war es, die Tendenzen bei den Rezidivraten zu untersuchen, die mit den Behandlungen eines Pilonidalsinus über die letzten Jahrzehnte einhergingen, von der Erstbeschreibung des Pilonidalsinus bis in die Gegenwart. Die Auswertung wurde durchgeführt, indem der Anteil von Patienten mit Rezidiv an der Zahl aller Patienten, bei denen eine Pilonidalsinusoperation erfolgte, ermittelt wurde.
Methoden: Dazu wurde eine umfassende Suche in den weltweiten Literaturdatenbanken durchgeführt, einschließlich PubMed, Embase, Science Direct und anderen, um die veröffentlichten Daten zum Pilonidalsinusrezidiv von 1833 bis heute zu sammeln. Die ermittelten Artikel wurden in Gruppen eingeteilt und ausgewertet.
Ergebnisse: Die veröffentlichten Gruppengrößen der Patienten reichten von $n = 6439$ Patienten mit $n = 130$ Fällen eines Rezidivs im Jahr 1930 bis $n = 42.537$ Patienten mit $n = 3548$ Fällen eines Rezidivs im Jahr 2000. Die Rate von Patienten mit Rezidiv, die sich zur Operation vorstellten, variierte zwischen 2% im Jahr 1930 und 14% im Jahr 1940, jedoch konnte keine klare Tendenz ermittelt werden. Es fällt auf, dass es keine signifikante Abnahme der rohen Rezidivrate zwischen 1930 und den letzten beiden Jahrzehnten gab. In dieser Studie wurden keine Einschränkungen festgestellt.
Schlussfolgerung: Den Ergebnissen zufolge nimmt die veröffentlichte Rezidivrate eines Pilonidalsinus nicht ab. Da die Inzidenz für einen Pilonidalsinus ansteigt, ist es möglich, dass die Wirkung überlegener Therapien durch unbekannte Faktoren, die die Ergebnisse beeinflussen, überschattet wird. Alternativ ist es auch plausibel, dass mehr herausfordernde Fälle publiziert werden oder dass verfügbare überlegene Therapien nicht adäquat eingesetzt werden. Der nächste Schritt wäre die Einbeziehung einer Analyse globaler Trends bei der Wahl der Therapie eines Pilonidalsinus.

Schlüsselwörter
Studientrends · Pilonidalsinus · Therapietrends · Pilonidalsinuserkrankung · Inzidenz

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