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ORIGINAL ARTICLES

Study Size Impact on Accuracy of the Worldwide Incidence of Pilonidal Sinus

Dietrich Doll [1, 2 *], Matthias Maak [3], Philipp Mörsdorf [4], Myriam Braun-Münker [5], Christina Oetzmann von Sochaczewski [6]

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Abstract

Background: Significant variations in incidence rates have been observed in the analysis of anecdotal Pilonidal Sinus Disease (PSD) incidents worldwide.

Objective: This study examines the accuracy of PSD incidence estimates and the variations associated with study size from 1833 to the present.

Material and Methods: To gather any PSD incidence data reported between 1833 and 2023, a comprehensive search was conducted in global literature databases, including PubMed, Embase, Science Direct, and others.

Results: The study sizes ranged from 26 to 82,217,837 individuals, with incidence rates varying from 8 to 30,000 cases per 100,000 persons. Notably, in study populations below 200,000 individuals, the incidence rate ranged from 8 to 30,000 cases per 100,000 persons. However, this range narrowed when studying populations exceeding 200,000 persons, with incidence rates ranging from 7 to 300 cases per 100,000 persons.

Limitations: No limitations were identified in this study.

Conclusion: The findings suggest reliable PSD incidences can be calculated with study populations exceeding 200,000 individuals. In such cases, the variability of incidence rates decreases as study size increases, although other known and unknown factors continue to influence the outcomes.

Keywords: study size, pilonidal sinus, disease incidence, variation in incidence, PSD

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* ***Corresponding author:***
Prof. Dr. Med. Dr. Phil. Dietrich Doll,
Dietrich.Doll@kk-om.de

1 *Department of Procto-Surgery & Pilonidal Sinus, St. Marienhospital Vechta, Academic Teaching Hospital of the MHH Hannover, Vechta, GERMANY*
2 *Pilonidal Research Group @ Vechtaer Research Institute VIFF; Vechta, GERMANY.*
3 *Department of Surgery, University Hospital Erlangen of the Friedrich-Alexander University Erlangen-Nuremberg, Erlangen, GERMANY.*
4 *Department of Trauma, Hand and Reconstructive Surgery, Saarland University, GERMANY.*
5 *Fulda University of Applied Sciences, Department of Food Technology, Fulda, GERMANY.*
6 *Department of Surgery, University Hospital of Bonn, Venusberg-Campus Bonn, GERMANY*

Introduction

The global incidence of pilonidal sinus disease (PSD) appears to be increasing[2,3], with variations observed among different countries[4,5] and sexes[6]. While there is limited published data on PSD from Africa, South America, and Asia, there has been a significant increase in publications from Mediterranean regions in 1970[7]. The reported incidences of PSD have been sporadic and highly variable, often differing by a factor of 1000 or more.

Given the considerable variability in reported incidence rates and the lack of comprehensive data, our study aimed to systematically investigate PSD's worldwide incidence figures from 1833 to 2023. By comprehensively analyzing available literature, we sought to better understand this disease's global incidence patterns.

Material and Methods

To achieve our objective, we conducted an extensive search across multiple 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. The search utilized specific keywords such as "Pilon* AND Incidence," "Pilonidal AND Sinus AND Incidence," "Pilon* AND population," "Pilonidal AND Sinus AND population," "Pilon* AND prevalence," and "Pilonidal AND Sinus AND prevalence."

In addition to the database search, we performed an on-site analysis of Endnote, which included 5,840 publications on PSD and 303 book chapters. We focused on the terms "incidence,"" incident*," "population," "prevalence," and "frequency." Relevant articles containing information on PSD incidence were carefully reviewed and analyzed.

The obtained results were thoroughly examined, and relevant data were tabulated as appropriate for further analysis.

Our inclusion criteria encompassed studies reporting incidence rates, the country of estimation, the number of PSD patients under care, the corresponding population size, 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 were applied to ensure the quality and relevance of the included studies. Excluded studies comprised those with an insufficient minimal dataset (as mentioned above), studies including the presence of neoplastic disease within the PSD population, meta-analyses, studies including extrasacral pilonidal sinus disease, conditions mimicking pilonidal sinus disease, and duplicate publications.

Statistical analysis: The study data was recorded in an Excel spreadsheet (Excel 2016, Microsoft Corporation, Redmond, WA, USA). Continuous variables were presented as mean ± standard deviation. All statistical analyses were conducted using SPSS version 26.0 (IBM Corp., Armonk, NY, USA).

Results

A total of 740 studies were identified, out of which n=97 provided suitable source data for calculating PSD incidence. However, we excluded 20 incidences from the German Statistisches Bundesamt and analyzed them separately to mitigate German dominance. This resulted in n=77 studies available for further analysis. Table 1 presents the details of these selected studies.

Table 1: Compilation of publications with PSD incidences between 1833 and 2017, sorted for year of study.

Author	Year of publication	Journal	Country	Study year	Study size	Incidence per 100.000	Soldiers	Mediterranean
Heurtaux[8]	1882	Soc de Chirurgie	France	1882	960	4.500	no	no
Wette[9]	1894	Arch f klin Chir	Not known	1894	15.000	47	no	no
Winkler[10]	1912	Wien Klin Wochenschr	Not known	1912	19.000	167	no	no
Breidenbach[11]	1935	Ann Surg	USA	1930	481.384	59	no	no
McKirdie[12]	1938	Ann Surg	USA	1932	149.913	64	no	no
Kooistra[13]	1942	Am J Surg	USA	1935	313.285	112	no	no
Buie[14]	1937	W.B. Saunders Publishing	USA	1937	3.700	27	not known	no
Johnson[15]	1938	J Nerv Ment Dis	USA	1938	1.000	3.400	no	no
Karydakis[16]	1973	Hell Arm Forces Med Rev	Greece	1940	26	8	yes	yes
Clark[17]	1946	J Indiana State Med Assoc	USA	1941	700.000	48	no	no
Pickett[18]	1942	Am J Surg	USA	1941	30.000	100	yes	no
Pickett[19]	1942	Am J Surg	USA	1942	k.A.	900	no	no
Pickett[19]	1942	Am J Surg	USA	1942	k.A.	500	yes	no
Favre[20]	1964	Mem Acad Chir (Paris)	USA	1944	2.900.000	300	yes	no
Parker[21]	1951	Am J Proctol	USA	1948	10.992	4.000	yes	no
Dwight/Maloy[22]	1953	New Engl J Med	USA	1952	31.497	1.158	no	no
Dwight/Maloy[22]	1953	New Engl J Med	USA	1952	21.367	112	no	no

U.S. Navy[23]	1956	Statistics of Navy Medicine	USA	1953	k.A.	411	yes	no
Franckowiak[24]	1962	Dis Colon Rectum	USA	1954	3.700	27	no	no
Hopping[25]	1954	Am J Surg	USA	1954	k.A.	3.800	no	no
Davage[26]	1954	Am J Pathol	USA	1954	k.A.	200	no	no
Sebrechts[27]	1961	Dis Colon Rectum	USA	1957	5.477	15.400	yes	no
Blackwell[28]	1959	Bull Tulane Univ Med Fac	USA	1959	1.407	1.600	no	no
Karydakis[16]	1973	Hell Arm Forces Med Rev	Greece	1960	23.400	4.900	yes	yes
Favre[20]	1964	Mem Acad Chir (Paris)	France	1963	k.A.	160	yes	no
Favre[20]	1964	Mem Acad Chir (Paris)	France	1964	k.A.	970	yes	no
Karydaki[16]s	1973	Hell Arm Forces Med Rev	Greece	1968	1.029.540	135	yes	yes
Karydakis[16]	1973	Hell Arm Forces Med Rev	Greece	1971	7.500	25.600	yes	yes
Karydakis	1973	Hell Arm Forces Med Rev	Greece	1971	8.704	14.800	yes	yes
Karydakis[16]	1973	Hell Arm Forces Med Rev	Greece	1972	21.800	15.298	yes	yes
Karydakis[16]	1973	Hell Arm Forces Med Rev	Greece	1973	42	57	yes	yes
Karydakis[16]	1973	Hell Arm Forces Med Rev	Greece	1973	7.500	30.000	yes	yes
Powell[29]	1975	J Pediatr	USA	1974	1.997	1.100	no	no
Campbell[30]	2002	Ann R Coll Surg Engl	UK	1974	281.000	7	no	no
Karydakis[31]	1992	Aust N Z J Surg	Greece	1974	210.000	25.800	yes	yes
Campbell[30]	2002	Ann R Coll Surg Engl	UK	1978	288.285	10	no	no
Obedman[32]	1993	Int J Adolesc Med Health	Spain	1981	14.960	1.070	no	yes
Campbell	2002	Ann R Coll Surg Engl	UK	1982	295.850	14	no	no
Campbell[30]	2002	Ann R Coll Surg Engl	UK	1984	300.938	28	no	no
Hori[33]	1984	Nihon Geka Hokan	Japan	1984	3.000	33	no	no
Evers[2]	2011	Zhonghua Wai Ke Za Zhi	Germany	1985	478.000	30	yes	no
Doll	1985	unpublished	Germany	1985	478.000	30	yes	no
Nazarian[34]	1987	Sov Med	Russia	1987	5.132	1.440	no	no
Vazquez[35]	2013	J Invest Dermatol	USA	1988	268	6.000	no	no
Sondenaa[36]	1995	Int J Colorectal Dis	Norway	1989	250.000	26	no	no
Campbell[30]	2002	Ann R Coll Surg Engl	UK	1990	319.651	27	no	no
Karydakis[37]	1992	Aust N Z J Surg	Greece	1992	168.000	31.500	yes	yes
Yücesan[38]	1993	Eur J Epidemiol	Turkey	1993	19.750	263	no	yes
Campbell[30]	2002	Ann R Coll Surg Engl	UK	1994	326.457	28	no	no
Akinci[39]	1999	Eur J Surg	Turkey	1996	1.000	8.800	yes	yes
Evers[2]	2011	Zhonghua Wai Ke Za Zhi	Germany	1996	363.500	100	yes	no

Carrasco[40]	2000	Ambul Surg	Spain	1996	832	23.200	no	yes
Campbell[30]	2002	Ann R Coll Surg Engl	UK	1998	335.485	32	no	no
Doll[41]	2015	PSJ	Germany	2000	k.A.	50	no	no
AFHSC[42]	2013	Medical Surveillance Monthly Report MSMR	USA	2006	35.517	190	yes	no
Evers[2]	2011	Zhonghua Wai Ke Za Zhi	Germany	2007	249.000	240	yes	no
Ardelt[43]	2017	Chirurg	Germany	2007	82.217.837	31	no	no
Doll[44]	2010	Unpublished	Germany	2007	249.000	240	yes	no
Doll[44]	2010	Unpublished	Germany	2007	24.200.000	45	no	no
Levinson[45]	2016	Mil Med	Israel	2007	k.A.	18	yes	yes
Sekmen[46]	2010	Turk J Surg	Turkey	2008	1.000	6.100	yes	yes
Evers[47]	2009	Amb Chir	Germany	2008	247.619	300	yes	no
Ardelt[48]	2017	Chirurg	Germany	2008	82.002.356	33	no	no
Ardelt[48]	2017	Chirurg	Germany	2009	81.802.257	34	no	no
Akbari[49]	2012	Iran J Public Health	Iran	2010	105.912	141	no	yes
Ardelt[48]	2017	Chirurg	Germany	2010	81.751.602	35	no	no
Aysan[50]	2013	Surg Today	Turkey	2010	432	9.400	no	yes
Ardelt[48]	2017	Chirurg	Germany	2011	80.327.900	37	no	no
Nasr[51]	2011	Can J Surg	Canada	2011	k.A.	120	no	no
Ardelt[48]	2017	Chirurg	Germany	2012	80.523.746	39	no	no
Doll[41]	2015	PSJ	Germany	2012	k.A.	98	no	no
Duman[52]	2016	Asian J Surg	Turkey	2013	19.013	6.600	no	yes
Ardelt[48]	2017	Chirurg	Germany	2013	80.767.463	38	no	no
Khodakaram[53]	2016	World J Surg	Sweden	2014	243	36	no	no
Ardelt[48]	2017	Chirurg	Germany	2014	81.197.537	38	no	no
Ardelt[48]	2017	Chirurg	Germany	2015	82.175.684	37	no	no
Zimova[54]	2015	Czech Medical Association J.E. Purkyne	Czechia	2015	k.A.	26	no	no

Research efforts have been conducted from 1882 to the present day. Among these investigations, 19 studies were carried out within the Mediterranean region, while the remaining 58 studies were conducted elsewhere. In the comprehensive analysis of these 77 studies, 48 studies involved participants from civilian populations, while 28 studies specifically focused on soldiers. Unfortunately, one study did not specify whether its participants were of civilian or military origin.

The research study populations varied significantly, ranging from a modest sample size of 26 individuals to an impressively large cohort of 82,217,837 persons. Additionally, the reported incidence rates in these studies showed a wide range, ranging from 7 cases per 100,000 individuals to 31,500 cases per 100,000 individuals. Figure 1 provides a graphical representation of the relationship between study size and incidence rate.

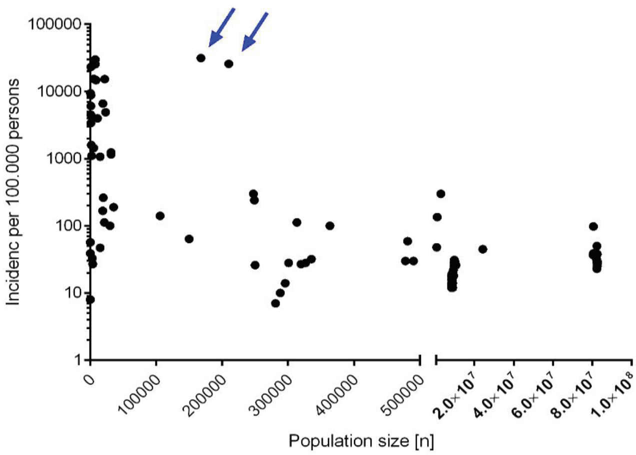

Figure 1: PSD-Incidence per 100,000 persons in the study population [n]. Please note the log. Scale on the Y-axis and separate the x-axis. Blue arrows indicate Karydakis′ studies (1974 and 1992, published in 1992) from Greece.

A study population below n=200,000 individuals was associated with a wide variation in incidence rates, ranging from 8 per 100,000 to 30,000 per 100,000, with an average of 7,460 per 100,000. When the study population exceeds 200,000 persons, the incidence rates tend to center around 74 per 100,000, with a narrower range of 7 to 300 per 100,000.

Two incidence rates, indicated by blue arrows in Figure 1, stand out as outliers and deviate from the expected pattern. These outliers originate from military studies conducted by Karydakis in Greece and exhibit a much higher incidence rate than anticipated[16].

In 1974, an unusually high incidence rate of 25,800 per 100,000 was reported among 210,000 Greek soldiers. In 1992, the incidence rate among 168,006 Greek soldiers rose to 31,500 per 100,000 (army size numbers were not given by Karydakis but by Government sources). However, since Karydakis did not provide more detailed data, it is not easy to provide further commentary on these outliers. Therefore, it is advised to interpret these two numbers (blue arrows in Figure 1) with caution.

Discussion

The present study emphasizes the significance of the study population size when estimating PSD incidence rates. It demonstrates that larger study populations yield more reliable and consistent estimates, while smaller populations may lead to wider variations in incidence rates. This finding underscores the importance of conducting studies with larger sample sizes to obtain more accurate and representative results.

The outliers observed in the military studies conducted by Karydakis in Greece raise intriguing questions about the factors contributing to the unusually high incidence rates reported in these populations.

This study represents the largest body of collective evidence on authors publishing about PSD incidence rates using their original data. It serves as a valuable resource for future analyses of PSD incidence data. As a minimum requirement, future research endeavors should focus on studies encompassing a population base exceeding 200,000 individuals.

Considering the scarcity of PSD data from Asian and African populations, calculating incidence rates, if possible, may require an even larger population base. Relevant limitations of our work are different definitions of incidence rates. It has been shown that period prevalence was higher than point prevalence and contact prevalence.[55]

In addition, different denominators of the incidence rates also affected the results.[56]

Two studies from Turkey might exemplify this issue: Sekmen and co-workers[46] used the contact prevalence and calculated an incidence rate of 6,100 per 100,000 persons. At the same time, Asians and colleagues [50] preferred the period prevalence and subsequently calculated an incidence rate of 9,400 per 100,000 persons. We could not systematically consider this aspect, as the older studies needed more relevant methodological detail, as was common at their time, to account for potential differences.

Future directions

In terms of future directions, it would be valuable to investigate population- and sex-based factors that may contribute to the development of PSD. Understanding potential disparities in incidence rates among different populations and genders can provide insights into the underlying risk factors and help guide targeted prevention strategies.

Furthermore, exploring the military's role and the Mediterranean region as potential risk factors for PSD could yield essential findings. Investigating whether specific occupational or environmental factors associated with military service or regional characteristics contribute to the higher incidence rates observed in these contexts can provide valuable insights for prevention and management efforts.

In conclusion, this study highlights the importance of considering the study population size, suggests future research directions, and emphasizes the need for further exploration of population-based and regional factors that may influence the incidence of PSD. By addressing these aspects, we may advance our understanding of PSD and work towards more effective strategies for its prevention and management.

Ethics: The analysis done in this study did not contain any interventions that could potentially cause harm to human participants. All proceedings were done according to established international studies and regulations.[1]

Conflicts of interest: All authors declare no conflicts of interest. No relevant or minor financial relationships exist between relatives or next of kin and external companies.

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Data availability: The datasets generated and analyzed are private because they have yet to be fully published. They will be available from the corresponding author in due time upon reasonable request

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