



Turkey is leading in the 21st century pilonidal sinus disease research

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ABSTRACT

Objective: Pilonidal sinus disease (PSD) has been a recognized pathology for the past 188 years. We studied whether scientific interest in this common disease has grown or declined over time. Our investigation included analysis of the world literature between 1833 and 2018.

Material and Methods: A PubMed search was conducted to identify all publications on pilonidal sinus disease, broken down by country, year of publication and number of patients included or described.

Results: The number of patients studied has been increasing, with date of more than 10,000 patients published per decade since 1970, and the total number of affected patients exceeding n= 40.000 in 2010 and Turkey leads the research, contributing 39% of the Mediterranean patients and 18% of the patients globally, while Italy provides 26% of the Mediterranean patients and 12% of the global total. Flap studies have increased, whereas primary open treatment studies have decreased from 40% in 1940 to less than 10% at present. Twenty percent of the studies performed today report primary median approaches, and the number of randomized controlled trials has increased.

Conclusion: Surgeons in Turkey currently publish the lion's share of the pilonidal sinus literature.

Keywords: Hair, pilonidal sinus, surgery, recurrence rate, publications, study size

INTRODUCTION

Pilonidal sinus disease research began in 1847 with Dr. AW Anderson, who removed tufts of hair from a non-healing wound on the "back" of a young man. Shortly thereafter the wound healed, and the cure was published (1). Since then, there have been thousands of publications, ranging from case reports to studies containing several thousand patients. At the time, open wound treatment was considered the standard of care as this was a septic wound. This became a problem during World War II, when more than 70.000 soldiers were temporarily decommissioned due to open wound treatment for PSD, amassing 47.000 sick leave days in 1941 alone (2). Primary closure was attempted as early as 1933 (3), but wound complications were high in the pre-antibiotic era. Several subsequent publications focused on surgical methods, including H-like (4) and Star-like (5-7) incisions, partial closure, obliteration of dead space using metal wires (8-10) or cotton rolls (11-13), and even perceived advantages and disadvantages of surgical drains (14,15). Roentgen irradiation, radioactive substances and obliterating substances were tried. When antibiotics finally became available to the surgical community (16) and asymmetric flap procedures increased, results improved-and research took off.

Contemporary work still mentions a number of therapeutic options, possibly either due to lack of comprehensive knowledge or to therapeutic negligence. In order to evaluate historical and current research efforts, we analyzed all publications addressing pilonidal sinus disease, from the first description up to the present literature, focusing on the number of patients treated and the number of articles published.

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MATERIAL and METHODS

We built a database with all of the literature found in a systematic search for the NCBI Medical Subject Heading (MeSH) term "pilonid*", as well as "dermoid" AND "cyst" in MEDLINE, PubMed, PubMed Central, Scopus, Ovid, Embase, and the Cochrane Central Register of Controlled Trials (CENTRAL), as described previously by Stauffer (17) and Doll (18). In brief, all randomized, non-randomized, prospective, retrospective, and observational studies such as cohort, case-control, and cross-sectional studies, and case reports published between 1833 and 2017 were

included. Figure 1 displays the results of the systematic search for evidence regarding recurrence and long-term follow-up data associated with common surgical procedures in PSD, based on the preferred reporting items for systematic reviews and meta-analyses (PRISMA).

Specific details of data collection, data extraction and quality assessment, as well as grouping of therapeutic procedures and statistical analyses, can be found in the papers by Stauffer (17) and Doll (18).

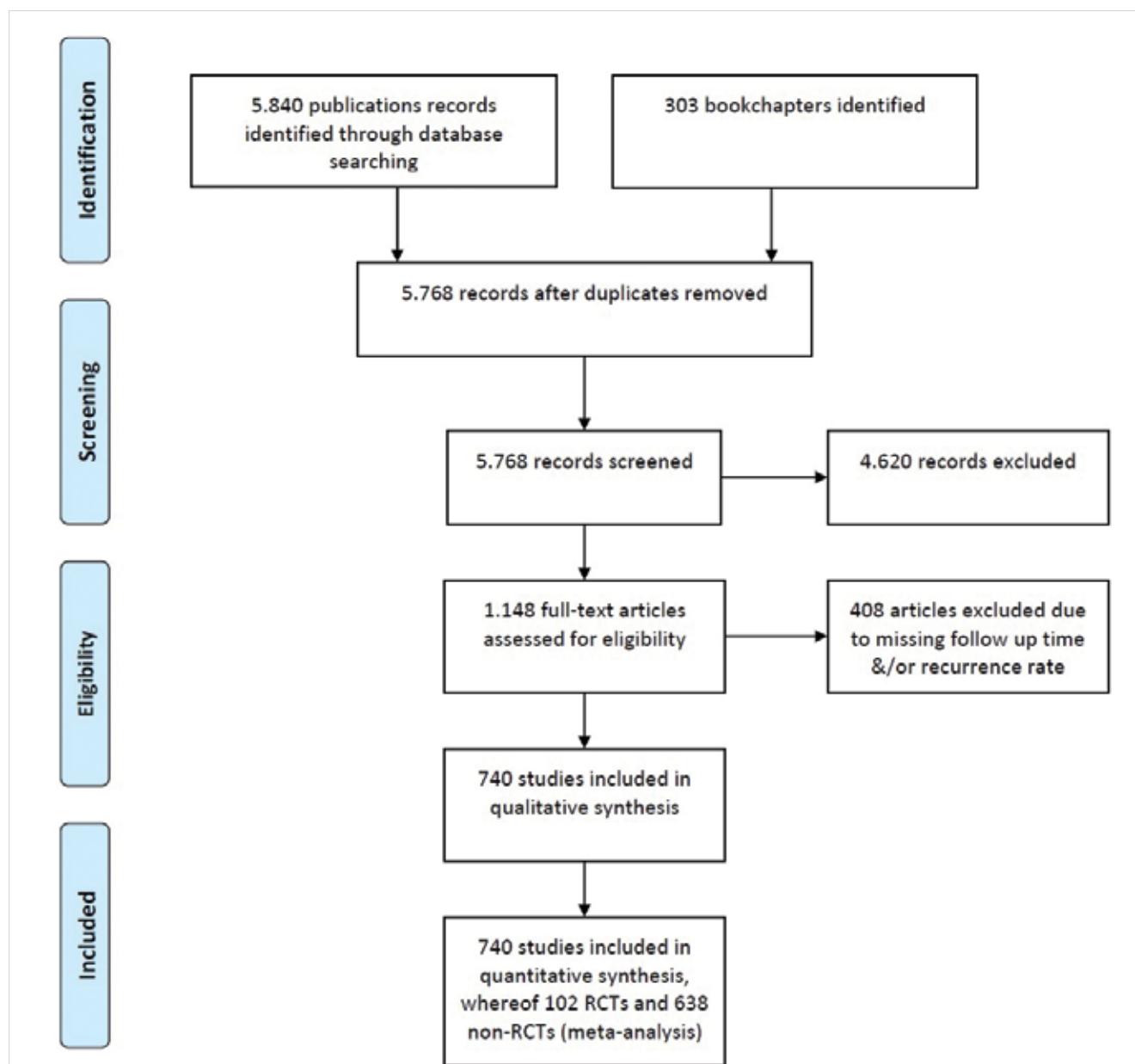


Figure 1. Flow diagram based on preferred reporting items for systematic reviews and meta-analyses (PRISMA) illustrating the systematic search for evidence regarding recurrence and long-term follow-up data associated with common surgical procedures in PSD.

RESULTS

The average sample size was $n = 73$ patients (mean $n = 33 \pm 8$ patients), with 60% of all studies published between 1833 and 2018 containing fewer than 50 patients (Figure 2). Thirty percent of the publications contained between 50 and 149 patients. Ultimately, 90% of all studies reported fewer than 150 patients on average. Sample size did not increase over time between 1940 and 2010, as illustrated by Figure 3. Here, the sample size over time was depicted from 1930 to 2010 (earlier publications are too scant to calculate a meaningful average). Please note that the values are given in mean \pm SEM (standard error of the mean).

Interestingly, the sample size per publication did not increase over time, and has remained between $n = 50$ patients and $n =$

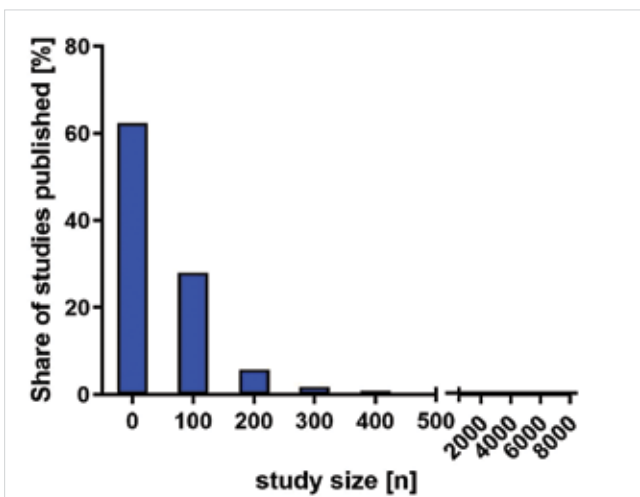


Figure 2. Histogram of pilonidal sinus disease (PSD) study sizes from 1833 to 2018.

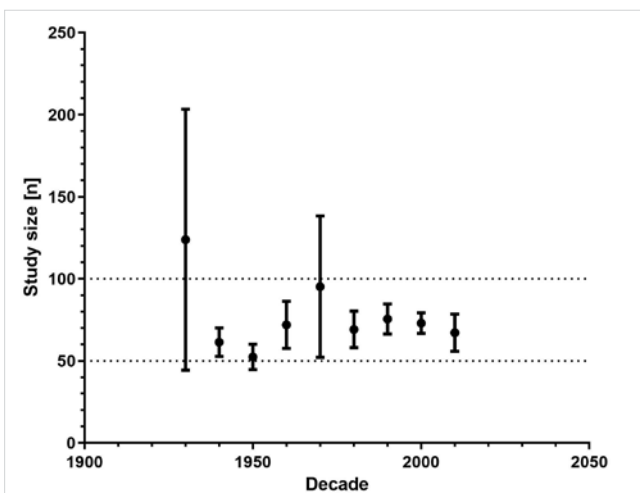


Figure 3. Size of published pilonidal sinus studies over time between 1930 and 2010, given as mean \pm SEM. Data from 1920 and earlier are too few to calculate average values \pm SEM.

100 patients over the past 8 decades. In order to analyze whether interest in PSD has grown over time, we investigated the total study population over time. Adding up all publications over time in decades, another picture arose (Figure 4). As Figure 4 illustrates, the total number of PSD patients mentioned in pilonidal sinus publications has been increasing significantly since the 1940's. Significant interest in the disease first arose during World War II due to the temporary decommissioning of more than 78.000 soldiers with PSD between 1941 and 1945 (19). Soldiers were absent for weeks and sometimes even months following surgery and primary open treatment. A more than fourfold increase could be seen during the first post-millennial decade.

While publications from North America (Canada and the USA) initially dominated between 1940 and 1950, interest seems to have decreased there, with cumulative numbers plateauing in the past 30 years (Figure 5). Conversely, interest from Northern Europe and the Mediterranean has shown a persistent increase over the

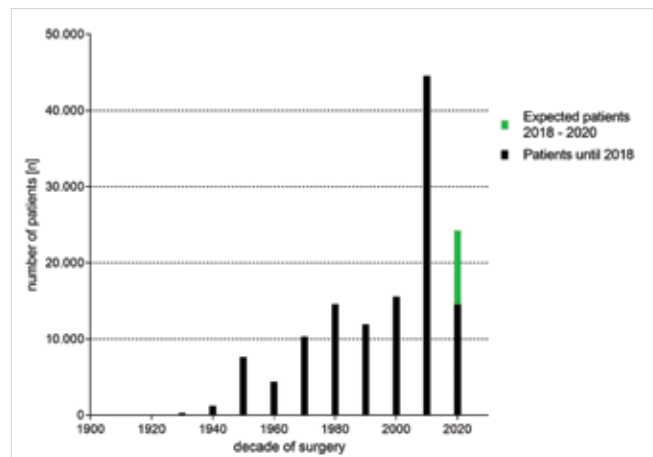


Figure 4. Number of published PSD patients according to decade of surgery. Please note that the 2020 value is estimated.

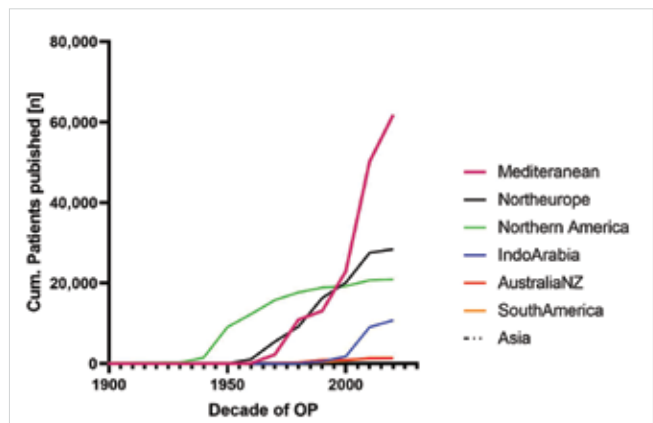
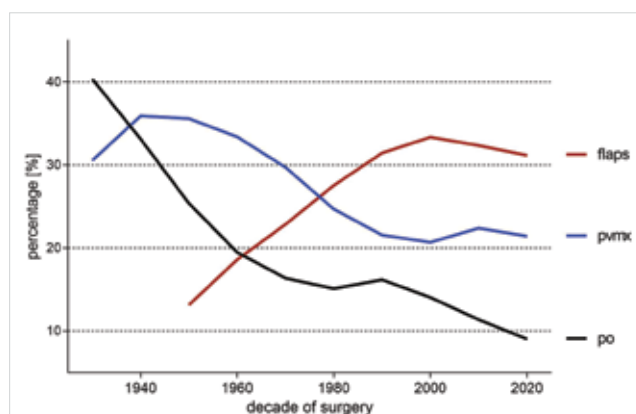


Figure 5. Number of pilonidal sinus disease (PSD) patients in research published between 1833 and 2018, by decade of surgery in different land groups (smoothed over 4 decades).

Table 1. Distribution of Mediterranean pilonidal sinus disease (PSD) patients in research published between 1833 and 2018

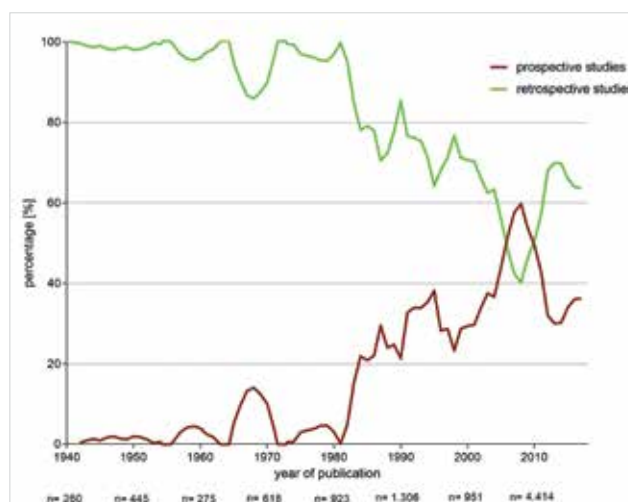
Country	Number of patients [n]	% Mediterranean	% worldwide
Turkey	23,998	38.56%	18.00%
Italy	16,088	25.74%	12.07%
Egypt	3,995	6.39%	3.00%
Spain	1,391	2.23%	1.04%
Israel	6,496	10.39%	4.87%
Greece	9,667	15.47%	7.25%
Lebanon	257	0.41%	0.19%
Croatia	100	0.16%	0.08%
Bosnia-Herzegovina	90	0.14%	0.07%
Morocco	14	0.02%	0.01%
Serbia	127	0.20%	0.10%
Portugal	8	0.01%	0.01%
Total	62,231	100%	46.67%

**Figure 6.** Percentage use of therapeutic methods by decade of surgery (Percentage depicts percentage of patients with certain therapy; smoothed over 4 decades).

past 6 decades. The Indo-Arab region started publishing study results from 2000 onwards. Asian patients are scant, and to our knowledge, there are still no studies from Africa. With the exception of isolated case reports, the disease seems to be practically non-existent among Africans.

In the Mediterranean region, Turkey and Italy are clearly the largest contributors to knowledge of PSD (Table 1). Italy contributed 25% of the Mediterranean patients, and 12% of the global total published between 1833 and 2018. Turkey heads the field, with 39% of Mediterranean and 18% of world PSD patients studied and published within this time frame.

Interest in primary open treatment seems to be decreasing (Figure 6). Flap surgeries are at the forefront, having been used in

**Figure 7.** Percentage of prospective vs. retrospective PSD studies by year of publication, smoothed over 4 decades.

more than 30% of the studies. Twenty percent of the studies still appear to include primary midline closure, which is characterized by prohibitive long-term recurrence rates (17).

Recent literature reveals a 30:60 ratio of retrospective to prospective studies, with a clear decrease in the number of retrospective studies since the 1980's. Despite being more laborious in terms of effort and resources, the perceived benefits and superiority of prospective studies are possible explanations (Figure 7).

DISCUSSION

Our study shows that a large body of knowledge on pilonidal disease has accumulated in the past decades, and the numbers of both publications and patients recruited are still increasing.

Evidence for new surgical techniques has emerged over the past two decades, with flap techniques being studied much more often compared to primary open or midline technique. It is mindboggling that 20% of the patients studied were still treated with midline closure in 2010 despite prohibitively high recurrence rates. This was impressively shown by Stauffer et al., who analyzed the same patient data as we did (17). With a 32% recurrence rate at 10 years, midline closure is only surpassed by limited excision, with 34% recurrence at 10 years of follow-up (17). These techniques both exceed by far the currently accepted standard of care, delivering acceptable recurrence rates of 1% (good result) to 2% per year of follow up (20).

Most of the contemporary PSD research derives from the Mediterranean. Turkish surgeons have contributed a large body of evidence over the past 40 years, and the share is still rising. There are many potential reasons, including the large case load in the civilian and military populations (both overt and silent disease) (21-23), greater awareness of the disease among surgeons, and scientific interest.

There is increasing evidence that geographical location may contribute to recurrence rate and therapy efficacy (18), as well as social and familial factors (24-27). The large burden of patients has encouraged Turkish surgeons to publish research on the factors contributing to greater PSD incidence (23), such as cut hair removal (28-30) as well as regular showering (31,32). New methods have been proposed (33-35), classifications defined, systematic reviews compiled (36) and rarities published (37-41). It is therefore understandable that-of all the general surgical articles in Turkey-15.4% cover pilonidal sinus disease and its treatment (42). More and more surgeons understand that the recurrence rate is a function of time, and while a rate of 2% per year of follow-up (i.e., 4% RR at 2 years) is acceptable, it can be lower (i.e., 5% at 3 years of follow-up) (20,43,44).

This goes hand in hand with an increased willingness to strive for more precise results through prospective randomized studies, which are shown to be increasing. Randomized controlled trials are generally conducted by large private and university-affiliated hospitals with specialized staff and research facilities. Nevertheless, small to midsize hospitals are the main location for pilonidal sinus surgeries, and the expertise of all surgeons involved in the treatment of pilonidal sinus disease is most welcome for inclusion in studies and publications.

We are aware of the limitations of our study, inherent in this type of epidemiological research. First, not all treated patients are included in published studies, and it would in fact be interesting to investigate this relation. Second, texts written in languages other than English, Spanish, French and German are lacking. Third, articles not indexed in PubMed, not published in larger journals, and not accessible via the Internet were not identifiable using our research algorithm. Fourth, there is a cer-

tain lag period between therapy, research, publication and the condensation of extended therapy results in a body of scientifically recognized knowledge. Analyses and recommendations for new therapies therefore often emerge following delays of several years.

It is therefore expected that analysis of additional publications in the coming years will shed more light on emerging and re-emerging techniques such as pit picking, endoscopic surgery and the use of lasers. More precise recurrence rates that incorporate longer follow-ups and new RCTs are to be expected.

The quality of studies has improved remarkably within the last decades, and is expected to increase further. Prospective randomized studies currently constitute 30% of the body of published articles.

CONCLUSION

In conclusion, both surgical interest and scientific activity are increasing in pilonidal sinus disease, and the Mediterranean region-especially Turkey-is contributing the lion's share to this body of evidence. As there is an increased interest in new treatment methods, new surgical approaches and respective evidence are eagerly awaited.

Ethics Committee Approval: This article does not contain any studies with human participants, human samples or live vertebrates. Therefore, no informed consent was needed or obtained prior to preparation of the current manuscript.

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REFERENCES

1. Anderson AW. Hair extracted from an ulcer. *Boston Med Surg J* 1847; 36: 74.
2. Granet E, Ferguson LK. Pilonidal Disease. *Am J Surg* 1945; 70: 14.
3. Weinstein M. Pilonidal sinus. *Ann Surg* 1933; 97: 80-4.
4. McCutchen GT. Pilonidal sinus: application of plastic surgical principles in a new surgical approach. *Ann Surg* 1943; 118: 430-7.
5. Gage M. Pilonidal sinus: sacrococcygeal ectodermal cysts and sinuses. *Ann Surg* 1939; 109: 291-303.
6. Berman JK. A method of treatment for pilonidal sinus. *Am J Surg* 1945; 70: 360-3.
7. Cantor AJ. Pilonidal dimple, sinus, cyst and abscess. *J Int Coll Surg* 1945; 8: 514-9.

8. Bowers WF, Williamson WS. Pilonidal sinus; preliminary report on a fresh approach. *AMA Arch Surg* 1956; 73: 931-5.
9. Dwight RW. Pilonidal sinus; an evaluation of plastic closure using flaps of gluteal muscle. *AMA Arch Surg* 1952; 64: 438-42.
10. Finestone EO. Technic of excision and primary closure of pilonidal sinus. *NY State J Med* 1952; 52: 1291-4.
11. Turner FP. Pilonidal sinus: primary closure with equal musculofascial flaps and removable far-and-near sutures; analysis of fifty-nine consecutive cases. *Ann Surg* 1954; 140: 687-94.
12. Turner FP, O'Neil JW. Treatment of pilonidal sinus by primary closure; a seven-year study. *AMA Arch Surg* 1959; 78: 398-405.
13. Laforet EG. Pilonidal sinus; a method of treatment by primary closure. *Am J Surg* 1957; 93: 873-5.
14. Kline LB. Pilonidal Cyst. *Medical Bulletin of the Veteran's Administration* 1935; 11: 241-6.
15. Macfee WF. Pilonidal cysts and sinuses: a method of wound closure: review of 230 cases. *Ann Surg* 1942; 116: 687-99.
16. Holman E. Pilonidal sinus-treatment by primary closure. *Surg Gynecol and Obstet* 1946; 83: 94-100.
17. 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.
18. Doll D, Orlik A, Maier K, Kauf P, Schmid M, Diekmann M, et al. Impact of geography and surgical approach on recurrence in global pilonidal sinus disease. *Sci Rep* 2019; 9: 15111.
19. Clark RL, Jr. The evolution of the treatment of pilonidal cysts and sinuses. *J Indiana State Med Assoc* 1946; 39: 387-92.
20. Doll D. 5- and 10-year recurrence rate is the new gold standard in pilonidal sinus surgery benchmarking. *Med Princ Pract* 2010; 19: 216-7.
21. Akinci OF, Bozer M, Uzunkoy A, Duzgun SA, Coskun A. Incidence and aetiological factors in pilonidal sinus among Turkish soldiers. *Eur J Surg* 1999; 165: 339-42.
22. Aysan E, Ilhan M, Bektas H, Kaya EA, Sam B, Buyukpinarbasili N, et al. Prevalence of sacrococcygeal pilonidal sinus as a silent disease. *Surg Today* 2013; 43: 1286-9.
23. Yücesan S, Dindar H, Olcay I, Okur H, Kilicaslan S, Ergoren Y, et al. Prevalence of congenital abnormalities in Turkish school children. *Eur J of Epidemiol* 1993; 9: 373-80.
24. 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.
25. Kueper J, Evers T, Wietelmann K, Doll D, Roffeis J, Schwabe P, et al. Sinus pilonidalis in patients of German military hospitals: a review. *GMS Interdiscip Plast Reconstr Surg DGPW* 2015; 4: 8.
26. Yildiz T, Elmas B, Yucak A, Turgut HT, Ilce Z. Risk factors for pilonidal sinus disease in teenagers. *Indian J Pediatr* 2017; 84: 134-8.
27. Yildiz T, Elmas B, Yucak A, Turgut HT, Ilce Z. Risk factors for Pilonidal Sinus Disease in teenagers. *Indian J Pediatr* 2016; 82: 134-8.
28. Bosche F, Luedi MM, van der Zypen D, Moersdorf P, Krapohl B, Doll D. The hair in the sinus: sharp-ended rootless head hair fragments can be found in large amounts in pilonidal sinus nests. *World J Surg* 2018; 42: 567-73.
29. Doll D, Bosche F, Hauser A, Moersdorf P, Sinicina I, Grunwald J, et al. The presence of occipital hair in the pilonidal sinus cavity-a triple approach to proof. *Int J Colorectal Dis* 2018; 33: 567-76.
30. Doll D, Wilhelm D, Ommer A, Albers K, Mordhorst H, Iesalnieks I, et al. Immediate cut hair translocation to the intergluteal fold in the hairdressers shop – another link to pilonidal sinus disease. *PSJ* 2019; 5: 23-32.
31. Bolandparvas S, Moghadam Dizaj P, Salahi R, Paydar S, Bananzadeh M, Abbasi HR, et al. Evaluation of the risk factors of pilonidal sinus: a single center experience. *Turk J Gastroenterol* 2012; 23: 535-7.
32. Sekmen Ü, Kara VM, Altintoprak F, Şenol Z. Pilonidal sinus in the army: Its incidence and risk factors. *Turk J Surg Article* 2010; 26: 95-8.
33. Dandin O, Tihan D, Karakas DO, Hazer B, Balta AZ, Aydin OU. A new surgical approach for pilonidal sinus disease: "de-epithelialization technique". *Turk J Surg* 2018; 34: 43-8.
34. Zorlu M, Sahiner IT, Zobaci E, Kocak C, Yasti AC, Dolapci M. Early results with the Mutaf technique: a novel off-midline approach in pilonidal sinus surgery. *Ann Surg Treat Res* 2016; 90: 265-71.
35. Sabuncuoglu MZ, Sabuncuoglu A, Dandin O, Benzin MF, Celik G, Sozen I, et al. Eyedrop-shaped, modified Limberg transposition flap in the treatment of pilonidal sinus disease. *Asian J Surg* 2015; 38: 161-7.
36. Emiroglu M, Karaali C, Esin H, Akpinar G, Aydin C. Treatment of pilonidal disease by phenol application. *Turk J Surg* 2017; 33: 5-9.
37. Agir H, Sen C, Cek D. Squamous cell carcinoma arising adjacent to a recurrent pilonidal disease. *Dermatol Surg* 2006; 32: 1174-5.
38. Eryilmaz R, Bilecik T, Okan I, Ozkan OV, Coskun A, Sahin M. Recurrent squamous cell carcinoma arising in a neglected pilonidal sinus: report of a case and literature review. *Int J Clin Exp Med* 2014; 7: 446-50.
39. Güner A, Barlas D, Çelik F. A very rare presentation of pilomatrixoma: Pilonoidal sinus [Pilomatriksoma'nin çok nadir bir prezentasyonu: Pilonoidal sinus]. *Erciyes Tip Derg Article* 2007; 29: 467-9.
40. Irkoren S, Sivrioglu N, Karaca H. Rare complication after VAC therapy in the treatment of tissue defect due to surgical excision of pilonidal sinus. *J Wound Ostomy Continence Nurs* 2013; 40: 641-3.
41. Oruc M, Kankaya Y, Colak Aslan O, Ozer K, Kocer U. Squamous cell carcinoma on the basis of pilonidal disease: A complication of chronicity. *EUR J PLAST SURG Letter* 2013; 36: 605-6.
42. 4Mayir B, Bilecik T, Dogan U, Koç U, Ensari CO, Oruc MT. Most cited articles in general surgery from Turkey. *Ulus Cerrahi Derg* 2015; 31: 85-9.
43. Doll D, Friederichs J, Dettmann H, Boulesteix AL, Duesel W, Petersen S. Time and rate of sinus formation in pilonidal sinus disease. *Int J Colorectal Dis* 2008; 23: 359-64.
44. Doll D, Krueger CM, Schrank S, Dettmann H, Petersen S, Duesel W. Timeline of recurrence after primary and secondary pilonidal sinus surgery. *Dis Colon Rectum* 2007; 50: 1928-34.

**ORİJİNAL ÇALIŞMA-ÖZET**

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Türkiye 21. yüzyıl pilonidal sinüs hastalığı araştırmalarında liderdirDietrich Doll¹, Verena Stauffer², Maja Diekann¹, Pieter Van Wyk³, Markus M Luedi⁴¹ St. Marienhospital, Prokto Cerrahi Anabilim Dalı, Vechta, Almanya² Lindenhofgruppe, Acil Tıp Anabilim Dalı, Bern, İsviçre³ Karolinska Üniversitesi Hastanesi, Travma ve Akut Bakım Bölümü, Stockholm, İsveç⁴ Bern Üniversitesi Hastanesi, Bern Üniversitesi Anesteziyoloji Anabilim Dalı, Bern, İsviçre**ÖZET**

Giriş ve Amaç: Pilonidal sinüs hastalığı (PSD), son 188 yıldır tanınan bir patolojidir. Bu yaygın hastalığa bilimsel ilginin zaman içinde artmış mı yoksa azalmış mı olduğunu araştırdık. Araştırmamız, 1833-2018 yılları arası dünya literatürünün analizini içeriyordu.

Gereç ve Yöntem: Pilonidal sinüs hastalığı ile ilgili, ülkeye, yayın yılına ve dahil edilen veya açıklanan hasta sayısına göre değerlendirilmiş tüm yayınları tanımlamak için bir PubMed araştırması yapılmıştır.

Bulgular: İncelenen hasta sayısı artmakta olup, 1970 yılından bu yana her dekada 10.000'den fazla hasta yayınlanmıştır ve 2010 yılında toplam hasta sayısı n= 40.000'i aşmıştır. Türkiye, Akdeniz bölgesindeki hastaların %39'unu, dünya genelindeyse hastaların %18'ini bildirerek bilimsel araştırmalara öncülük etmektedir. İtalya Akdeniz hastalarının %26'sını ve küresel toplamın %12'sini bildirmektedir. Flep çalışmaları artarken, birincil açık tedavi çalışmaları ise 1940'taki %40'lık değerden günümüzdeki %10'ların altındaki düzeylere düşmüştür. Bugün yapılan çalışmaların yüzde yirmisi birincil medyan yaklaşımları bildirmekte ve randomize kontrollü çalışmaların sayısı artmaktadır.

Sonuç: Türk cerrahları şu anda pilonidal sinüs literatüründeki aslan payını yayınlamaktadır.

Anahtar Kelimeler: Kıl, pilonidal sinüs, cerrahi, nüks oranı, yayınlar, çalışma büyüklüğü

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