https://doi.org/10.22416/1382-4376-2025-35-1-7-14
UDC 616.711.8-006

Pilonidal Sinus Carcinoma: Review of Literature and Own Data

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Aim: to analyse and estimate the incidence of pilonidal cyst cancer from 2023 to 2024.
Key points. In 2023, M. F. Safadi et al. published a review of the incidence of pilonidal cyst cancer, including 140 cases from 1900 to 2022. From 2023 to 2024, 14 observations of patients with malignant pilonidal cyst appeared in the literature. One case of malignant pilonidal transformation was reported in our practice. Patients' age varied from 19 to 86 years (54.5 ± 11.9 years). Among the patients, males predominated — 88.9 % (137/154), the male: female ratio was 8.1 : 1. The time from the first diagnosis of a pilonidal cyst to the development of cancer in the cyst ranged from 1 month to 62 years. The mean interval from detection of pilonidal cyst to malignancy was 21.1 ± 13.6 years (median — 20.0 years). One of the reasons for malignant degeneration of pilonidal cysts may be the presence of concomitant hidradenitis suppurativa, which is included in follicular occlusion syndrome. However, the direct association of follicular occlusion syndrome with pilonidal cyst malignancy has not been definitively confirmed.
Conclusion. Malignancy of pilonidal cysts is a rare complication of a long-term inflammatory process in the sacrococcygeal region. The authors concluded that it is necessary to emphasise the presence of a concomitant follicular occlusion syndrome in patients, which may alter the usual management tactics of pilonidal disease.
Keywords: pilonidal cyst, malignant transformation, follicular occlusion syndrome, epithelial coccygeal tract
Conflict of interest: the authors declare no conflict of interest.

For citation: Tsarkov P.V, Shlyk D.D., Safyanov L.A., Erdyneev D.V. Pilonidal Sinus Carcinoma: Review of Literature and Own Data. Russian Journal of Gastroenterology, Hepatology, Coloproctology. 2025;35(1):7–14. https://doi.org/10.22416/1382-4376-2025-35-1-7-14

Рак пилонидальной кисты: обзор литературы и собственные данные

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Цель обзора: анализ и оценка заболеваемости раком пилонидальной кисты с 2023 по 2024 г.
Основные положения. В 2023 г. M.F. Safadi et al. опубликовали обзор заболеваемости раком пилонидальной кисты, охвативший 140 случаев с 1900 по 2022 г. С 2023 по 2024 г. в литературе появилось 14 наблюдений за пациентами с малигнизированной пилонидальной кистой. В нашей практике был зафиксирован один случай злокачественной трансформации пилонидальной болезни. Возраст больных варьирует от 19 до 86 лет (54,5 ± 11,9 года). Среди пациентов преобладают мужчины — 88,9 % (137/154), соотношение мужчин : женщин — 8,1:1. Длительность от постановки первичного диагноза пилонидальной кисты до возникновения рака в ней варьирует в широких пределах — от 1 месяца до 62 лет. Средний промежуток от выявления пилонидальной кисты до малигнизации составил 21,1 ± 13,6 года (медиана — 20,0 года). Одной из причин злокачественного перерождения пилонидальной кисты может быть наличие сопутствующего гнойного гидраденита, который входит в синдром фолликулярной окклюзии. Однако прямая связь синдрома фолликулярной окклюзии с малигнизацией пилонидальной кисты окончательно не подтверждена.
Заключение. Малигнизация пилонидальной кисты является редким осложнением длительно текущего воспалительного процесса в крестцово-копчиковой области. Авторы пришли к выводу, что необходимо акцентировать внимание на наличии у пациентов сопутствующего синдрома фолликулярной окклюзии, который может изменить привычную тактику лечения пилонидальной болезни.
Ключевые слова: пилонидальная киста, злокачественная трансформация, синдром фолликулярной окклюзии, эпителиальный копчиковый ход
Конфликт интересов: авторы заявляют об отсутствии конфликта интересов.

Для цитирования: Царьков П.В., Шлык Д.Д., Сафьянов Л.А., Эрдынеев Д.В. Рак пилонидальной кисты: обзор литературы и собственные данные. Российский журнал гастроэнтерологии, гепатологии, колопроктологии. 2025;35(1):7–14. https://doi.org/10.22416/1382-4376-2025-35-1-7-14

Introduction

Pilonidal sinus disease (PSD) is one of the most common pathological conditions in surgical practice, occurring in 5 % of the population [1, 2]. Malignant transformation of PSD is a rare but highly significant complication. The first case of PSD carcinoma was described in 1900 by H. Wolff in a 21-year-old woman with recurrent disease after multiple surgical interventions [3]. In 2023, M.F. Safadi et al. published a review of the incidence of pilonidal sinus carcinoma, including 140 cases from 1900 to 2022 [4]. The risk of pilonidal sinus malignancy is approximately 0.1 % of the total number of cases [5].

Malignant degeneration is thought to be associated with prolonged inflammation in the sacrum and coccyx region caused by impaired hair growth. This leads to the formation of additional fistulas and damage to the protective epithelial barrier, favouring penetration of saprophytic microflora into the dermis, chronic inflammation and epithelial dysplasia [6]. In recent years, data have emerged on different variants of the course of PSD, including its 'clinical masks'. In particular, cases have been described in which PSD is a part of follicular occlusion syndrome or masks a postsacral cyst. Both of these conditions may increase the risk of malignancy over time [7]. However, the direct association of malignancy with follicular occlusion syndrome remains poorly understood.

Material and methods

A literature search was conducted to identify all cases of pilonidal sinus carcinoma using the keywords "pilonidal and carcinoma", "pilonidal and tumour", "pilonidal and sinus and carcinoma". The search was conducted using resources such as PubMed, Google Scholar, Medline, Cochrane, Scopus and Web of Science. All articles describing cases of malignant transformation of pilonidal cysts were selected, except for publications in which the tumour was not associated with an established diagnosis of PSD. Each case was analysed to exclude possible duplication of data.

The data was then organized and entered into tables using Microsoft Excel (version 2016, build 2409; Microsoft Corp., USA) in a systematic manner. The tables reflected the following parameters: author of the publication, year of publication, age and sex of the patient, time interval from the diagnosis of PSD to the development of malignisation, and histological type of the tumour. The data were analyzed using tables and graphs.

No additional information on the incidence of pilonidal sinus carcinoma was found in the published data from 1900 to 2022. However, 14 cases of patients with pilonidal sinus carcinoma have been described in the literature from 2022 to the present.

Furthermore, our practice has also documented a single case of malignant transformation of PSD.

Clinical case report

The patient is a 70-year-old male with a 50+ year history of pilonidal disease. During this time, four I&D procedures of PSD abscess have been performed. In addition, the patient had a long history of hidradenitis suppurativa in the axillary region requiring repeated opening and drainage of abscesses. MRI of the sacrococcygeal region in April 2024 revealed three sinuses in the subcutaneous tissue of the left buttock at intergluteal cleft, up to 2.5 mm in diameter, located medially at the level of the coccyx. The sinuses merged into a single cavity measuring 57 63 37 mm, perforating the external anal sphincter at the level of the subcutaneous part and opening into the anal canal at a distance of 10 mm from its outer edge. There were no additional spurs and no connection with the levator muscle had been found. The coccygeal vertebrae were involved, but the rectum was not. The lymph nodes of the small pelvis were unremarkable. Histological examination (No. 4196/2024 dated May 2024) confirmed a highly differentiated squamous cell carcinoma with signs of keratinization. At the multidisciplinary team (MDT) on 15 May 2024, it was decided to perform radiotherapy, which the patient is currently undergoing.

A total of 155 observations of patients with pilonidal sinus carcinoma were analyzed. The results of recent publications in addition to previously published data are shown in Table 1.

Country and language of the publications

From 1900 to 2022, M.F. Safadi et al. identified 140 published cases of pilonidal sinus cancer [4]. From 2023 to 2024, a further 14 cases of pilonidal cancer were identified (Table 1). Of the 154 described observations, the majority of publications were in English (75 %; 81 of 108). The majority of the articles reported single cases (82.4 %; 89 of 108), with the remainder reporting between 2 and 9 observations. Geographically, cases of malignancy were reported from the USA, Spain, Turkey, Russia, India and other countries.

Table 1. Reported cases of pilonidal sinus cancer in 2023–2024 (n = 15)
Таблица 1. Зарегистрированные случаи рака пилонидальной кисты за период 2023–2024 гг. (n = 15)

No. *№*	Year *Год*	Author *Автор*	Country *Страна*	Gender *Пол*	Age *Возраст*	Period *Период*	Histology *Гистология*
1	2023	Soria Rivas A. et al. [8]	Spain *Испания*	Male *Муж.*	63	30	SCC *ПКК*
2	2023	Soria Rivas A. et al. [8]	Spain *Испания*	Male *Муж.*	40	20	SCC *ПКК*
3	2023	Soria Rivas A. et al. [8]	Spain *Испания*	Male *Муж.*	53	20	SCC *ПКК*
4	2023	Soria Rivas A. et al. [8]	Spain *Испания*	Male *Муж.*	69	11	SCC *ПКК*
5	2023	Soria Rivas A. et al. [8]	Spain *Испания*	Male *Муж.*	70	20	SCC *ПКК*
6	2023	Soria Rivas A. et al. [8]	Spain *Испания*	Male *Муж.*	57	12	SCC *ПКК*
7	2023	Soria Rivas A. et al. [8]	Spain *Испания*	Male *Муж.*	68	51	SCC *ПКК*
8	2023	Soria Rivas A. et al. [8]	Spain *Испания*	Male *Муж.*	54	36	SCC *ПКК*
9	2023	Soria Rivas A. et al. [8]	Spain *Испания*	Male *Муж.*	64	19	SCC *ПКК*
10	2023	Sergatskiy K.I. et al. [9]	Russia *Россия*	Male *Муж.*	62	40	SCC *ПКК*
11	2023	Sergatskiy K.I. et al. [9]	Russia *Россия*	Male *Муж.*	59	30	SCC *ПКК*
12	2024	Sommovilla J. et al. [10]	USA *США*	Male *Муж.*	60	20	SCC *ПКК*
13	2024	Saddouki F. et al. [11]	Marocco *Марокко*	Male *Муж.*	32	15	SCC *ПКК*
14	2024	Khatri H. et al. [12]	Australia *Австралия*	Female *Жен.*	80	n/d *н/д*	BCC *БКК*
15	2024	Our own observation *Собственное наблюдение*	Russia *Россия*	Male *Муж.*	70	50	SCC *ПКК*

Note: *SCC — squamous cell carcinoma, BCC — basal cell carcinoma, n/d — no data.*
Примечание: *ПКК — плоскоклеточная карцинома, БКК — базальноклеточная карцинома, н/д — нет данных.*

Age and gender of patients

The age of patients ranged from 19 to 86 years (54.5 ± 11.9 years). Of the patients, 88.9 % (137/154) were male, and the male-to-female ratio was 8.1 : 1. The time from initial diagnosis of a PSD to the development of cancer in the cyst varied widely from 1 month [13] to 62 years [14]. The mean time from detection of PSD to malignancy was 21.1 ± 13.6 years (median — 20.0 years).

The histological type of the tumor

In terms of morphological structure, squamous cell carcinoma is the most common (91.6 %; 142/155). Other types include basal cell carcinoma, mixed squamous and basal cell carcinoma and adenocarcinoma. In 1989, a case of rhabdomyosarcoma was reported in Macedonia in a series of 5 observations [15].

None of the 154 previously described observations of patients with malignant pilonidal cyst were associated with manifestations of follicular occlusion syndrome or postsacral cysts.

Discussion

The incidence of pilonidal sinus cancer was first estimated by E.A. Gaston and W.L. Wilde in 1965. Over a period of 25 years, 891 people with PSD were operated on at Framingham Union Hospital & Boston City Hospital. Only one case of epidermoid carcinoma was reported. The incidence rate was 0.1 %. The authors compared the incidence of pilonidal cyst cancer with the incidence of chronic cutaneous fistulas and sinuses. A.K. McAnally and M.B. Dockerty found 13 patients at the Mayo Clinic and concluded that the percentage of malignancy was 0.23 % for chronic fistulous osteomyelitis, 0.1 % for rectal fistula and 0.07 % for sinus empyema. These data are comparable to the incidence

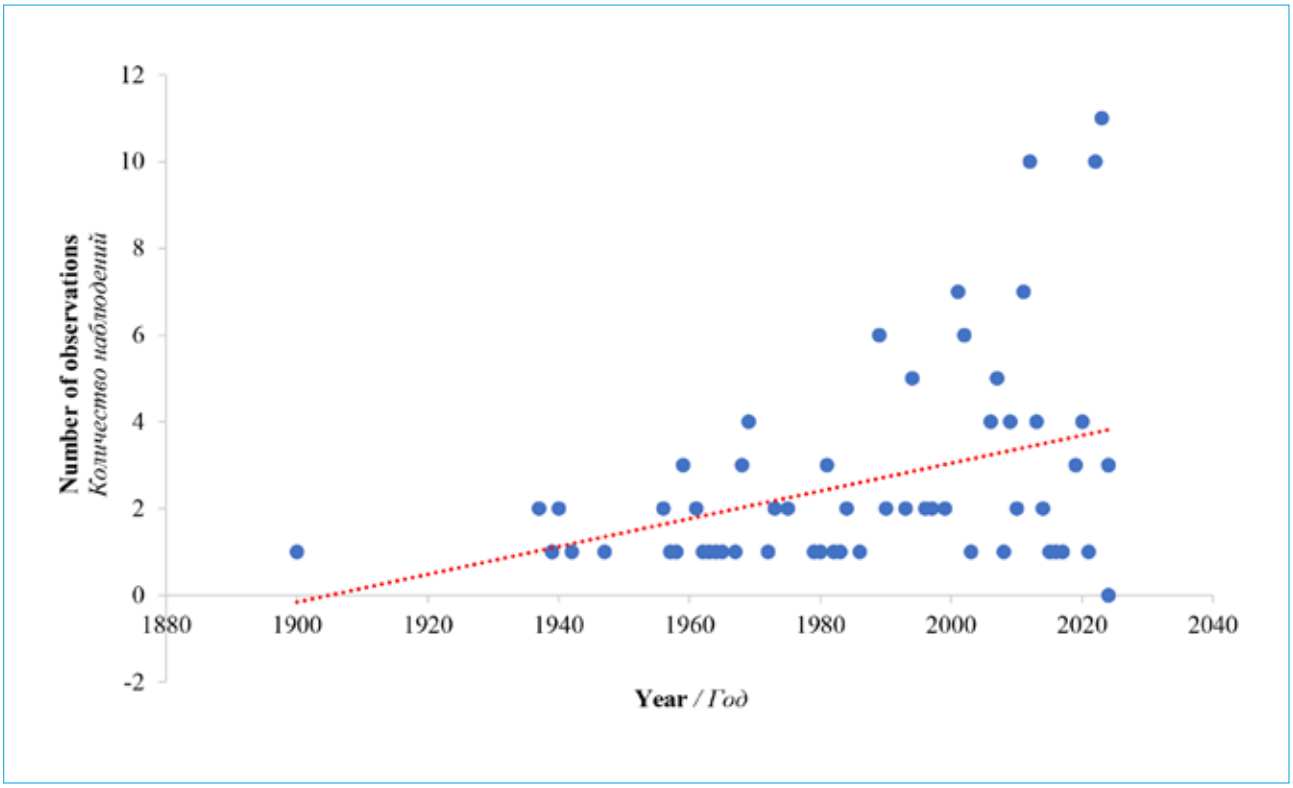

Figure 1. Distribution of registered cases of malignant transformation of pilonidal cyst from 1900 to 2024

Рисунок 1. Распределение зарегистрированных случаев злокачественной трансформации пилонидальной кисты в период с 1900 по 2024 г.

Figure 2. Distribution of patients with pilonidal cyst cancer depending on the morphological type of the tumor

Рисунок 2. Распределение больных раком пилонидальной кисты в зависимости от морфологического типа опухоли

of cancer in pilonidal cysts reported by E.A. Gaston and W.L. Wilde [5].

In addition, authors evaluating the incidence of pilonidal cyst cancer from 1965 to 2011 have approached the problem from different angles (Table 2). In 1981, S. Gupta et al. published a review in which the authors evaluated the incidence rate of pilonidal cyst cancer over 15 years at the Banaras University Hospital. There was one case of pilonidal cyst cancer in 18 patients with PSD. The incidence rate was 5.56 %. These results may be due to the fact that patients with a long history of the disease and advanced stage of the disease were more likely to seek treatment at this hospital [16].

On the other hand, an article published by M.A. Casberg in 1949 reports on 78,924 patients operated on for PSD during the Second World War, in which no cases of malignant transformation of PSD were reported. This is most likely due to the young age of the patients and the short follow-up of these patients [17].

In a 2011 review, I. Alarc n-Del Agua et al. estimated the incidence of pilonidal cyst cancer. Over a 13-year period, cancer was found in 4 patients out of 3,560 pilonidal cyst patients. The incidence rate was 0.11 % [18].

In the most recent review, published 12 years later, M.F. Safadi et al. in 2023 surveyed 1050 pathologists in 834 hospitals treating pilonidal sinus disease in Germany. The authors found 140 published cases of pilonidal sinus cancer and two unpublished cases from Germany. The male to female incidence ratio was 7.75 : 1. The mean age was 54.0 ± 11.8 years and the time from PSD to malignancy was 20.1 ± 14.1 years. The authors found an association between the high incidence of PSD and the development of cancer in PSD. They estimated that the incidence rate of pilonidal cyst cancer was 0.17 % of the total incidence. Taking into account race, level of medical care, geographical parameters and patient adherence to treatment, this rate may vary significantly from the reported rate in different countries and localities. If patients with pilonidal disease seek early medical attention, the incidence of pilonidal cyst cancer will decrease [4].

When comparing the data in the previously published analysis for 2023 and adding the new 15 cases with respect to patient age (54.0 ± 11.8 and 54.5 ± 11.9 years), male to female ratio (7.75 : 1 and 8.08 : 1), and time from diagnosis to malignancy (20.1 ± 14.1 and 21.1 ± 13.6 years),

Table 2. Publications that assessed the incidence of pilonidal cyst cancer
Таблица 2. Публикации, в которых проведена оценка заболеваемости раком пилонидальной кисты

Publication ***Исследование***	**Year** ***Год***	**Country** ***Страна***	**Period** ***Период***	**Number of PS cases** ***Кол-во случаев ПК***	**Number of PC cases** ***Кол-во случаев рака ПК***	**Percentage of morbidity** ***Процент***
Gaston E.A., Wilde W.L. [5]	1965	USA *США*	1940–1965	891	1	0.11 %
Mukhadze G.I. [20]	1975	Georgia (USSR) *Грузия*	–	245	2	0.82 %
Pilipshen S.J. et al. [21]		USA *США*	1932–1981	2457	2	0.08 %
Gupta S. et al. [16]	1981	India *Индия*	1965–1979	18	1	5.56 %
Mirceva D. et al. [15]	1989	Macedonia *Македония*	1967–1987	992	5	0.50 %
Adámek J. et al. [22]	1990	Czech Republic *Чехия*	1970–1990	123	2	1.63 %
Abboud B. et al. [23]	1999	Lebanon *Ливан*	–	500	1	0.20 %
Velitchklov N. et al. [24]	2001	Bulgaria *Болгария*	1975–1999	3734	1	0.03 %
Alecha Gil J. et al. [25]	2006	Spain *Испания*	1994–2004	367	3	0.82 %
Alarcón-Del Agua I. et al. [18]	2011	Spain *Испания*	1995–2008	3560	4	0.11 %
Total / *Всего*				12 887	22	0.17 %

Note: *PS — pilonidal sinus, PC — pilonidal carcinoma.*
Примечание: *ПК — пилонидальная киста.*

one can notice that the changes are insignificant and consistent with statistical error. However, in this review we focus on the association between pilonidal cyst malignancy and the risk factor of concomitant hidradenitis suppurativa.

In a previous publication on follicular occlusion syndrome, published in 2023, the authors analyzed 80 patients diagnosed with PSD between November 2018 and December 2019. In 7 observations (8.8 %), an association was found between the presence of PSD in the patient and different variants of the course of follicular occlusion syndrome, including hidradenitis suppurativa, acne conglobata and dissecting cellulitis of the scalp [19].

The low incidence of pilonidal cyst cancer reported by many researchers can be attributed, firstly, to the fact that surgeons do not always send resected preparations for pathological examinations. This is due to the non-mandatory nature of the preparation examination in some regions of the world and the desire to reduce treatment costs [26]. Secondly, young people with a short history of the disease, in whom the probability of malignisation is extremely low, are most often operated on. Even if the preparation has been sent for histological examination, the pathologist may make an insufficient number of sections, thus missing microfoci of malignancy [26, 27].

Currently, minimally invasive surgery is on the rise: phenol and fibrin glue obliteration, laser treatment of pilonidal sinus. These procedures destroy the epithelial lining without removing the cyst as a single block. As a result, microfoci suspicious for malignancy may be missed [28].

It should also be noted that many journals do not accept clinical case reports for publication. All of this may contribute to the inability to accurately assess the prevalence of malignant pilonidal sinus lesions. Finally, there are more articles in the non-English literature, which makes them difficult to find and consider in the global community [28, 29].

Some investigators have reported a lower rate of malignancy in PSD compared to other chronic skin conditions. The significantly low malignancy rate of PSD is mainly due to the high rate of early treatment in young patients in developed countries [26]. In 1954, O.N. Davage reported the presence of squamous epithelium in only 50 % of resected specimens after surgical treatment of pilonidal cysts. According to the study, the absence of this type of epithelium in the removed specimen halved the rate of malignancy [30].

One of the reasons for malignant transformation of pilonidal cysts may be the presence of concomitant suppurative hidradenitis. In 2021, M. Sachdeva et al. published a review examining the development of squamous cell carcinoma in the setting of hidradenitis suppurativa. The total number of observations in the publication was 95 (average age — 52.9 years). The most common sites of squamous cell carcinoma were the gluteal region (47.5 %; 58/122), the perianal region (18.9 %; 23/122), and the genitalia (13.9 %; 17/122). The median time from diagnosis of hidradenitis suppurativa to the development of squamous cell carcinoma was 25.5 years. The main causes of death were metastasis (34.1 %; 15/44) and sepsis (13.6 %; 6/44). The authors concluded that patients with a long history of hidradenitis suppurativa have a high risk of squamous cell carcinoma and metastases [31]. For the first time, we focus on the relationship between pilonidal cyst malignancy and the presence of concomitant hidradenitis suppurativa, which is confirmed by our clinical observation.

Conclusion

Malignancy of pilonidal cyst remains a rare but important complication of chronic suppurative processes in the sacrococcygeal region. The main risk factor is a long history of the disease, averaging about 20 years. We believe that increased attention should be paid to patients with a combination of PSD and follicular occlusion syndrome; such a combination may require not only surgical removal of the pilonidal cyst, but also specific systemic therapy to reduce the risk of transition of the disease to a long-term chronic form and thus the risk of malignancy. Further studies are needed to clarify the pathogenetic mechanisms and to develop optimal approaches for diagnosis and treatment of patients at high risk of malignant degeneration.

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Редактирование: Царьков П.В.
Проверка и согласование верстки с авторским коллективом: Эрдынеев Д.В.

Submitted: 13.11. Accepted: 28.01.2025 Published: 28.02.2025
Поступила: 13.11.2024 Принята: 28.01.2025 Опубликована: 28.02.2025

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