

Case Report

A different disease: extrasacroccygeal pilonidal sinuses etiopathogenesis

Fatih Çiftci¹, Ibrahim Abdurrahman²

¹Vocational School of Health Services, Istanbul Gelişim University, Avcılar, Istanbul, Turkey; ²Department of Internal Medicine, Safa Hospital, Bağcılar, Istanbul, Turkey

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Abstract: Pilonidal sinuses (PS) predominantly affect young male adults, usually occurring in the sacroccygeal region. However, PS occasionally occurs in other parts of the body, referred to as extrasacroccygeal pilonidal sinus (ESPS). We herein evaluate ESPS etiopathogenesis and treatment. Of a total of 949 PS cases treated between 2006 and 2011, 21 were of ESPS (2.2% of the total), which were evaluated retrospectively. The affected regions were the breast ($n = 1$), scalp ($n = 2$), sternum ($n = 2$), abdominal wall ($n = 5$), neck ($n = 2$), groin ($n = 4$), and axilla ($n = 5$). Lesions of the abdominal wall are rare, but less so than lesions in other regions. PS may mimic hidradenitis suppurativa histologically. To our knowledge, PS of the breast and groin have not previously been reported. Twelve of our patients reported shaving the affected region; we suggest this may have played a role in the disease pathogenesis.

Keywords: Pilonidal sinuses, extrasacroccygeal region, etiopathogenesis

Introduction

Pilonidal sinuses (PS) predominantly occur in sacroccygeal regions in young adult males; other regions that can be affected include the nose, umbilicus, ear, axilla, and the toe and finger web spaces. PS, which may result from excessive and repetitive trauma to the affected region, may predispose the sufferer to cellulitis, abscess formation, fistula, and infrequently, squamous cell carcinoma. PS can occur in other regions of the body, where it is often linked to repetitive trauma [1]. In this retrospective study we evaluated the medical records of PS patients treated between 2006 and 2011. We herein highlight cases involving atypical regions of the body and discuss the pathological findings and possible pathogenic factors.

Materials and methods

Between 2006 and 2011, we treated a total of 949 PS cases in our General Surgery Clinic. Following a retrospective review, we identified 21 ESPS cases (2.2%); medical records were evaluated by a pathology specialist, and diagnoses were confirmed histopathologically (**Figure 1**). For the statistical evaluation, the

SPSS 16.0 for Windows (SPSS Inc. Chicago, IL, USA) program was used to analyze the definitive statistics of the results. In the definitive statistics, continuous variables were shown as mean \pm standard deviations, and for the categorical variables, percentages and the number of cases were used.

A PS or cyst, which involves an infected cavity under the skin, may also be characterized by a hair protruding through a small orifice; hairs and skin debris from the dermis and epidermis can provoke a foreign body reaction within the tract due to the formation of inflamed granulation tissue. For economic reasons, all patients were treated with surgical excision at the lesion site under local anesthesia with lidocaine (3 mg/kg) infiltration, followed by total elliptical excision and primary defect closure. All patients were discharged and returned to work on the day of the operation; oral cefuroxime axetil (500 mg) was administered twice.

Results

ESPS cases represented 2.2% of all of the PS cases in our series. Lesion locations and patient characteristics are detailed in **Table 1**.

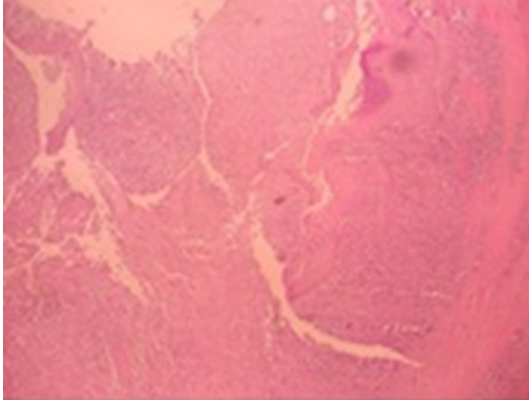


Figure 1. Photograph of a sinus tract with a fibrotic dermal stoma surrounded by intense inflammation and covered by multiple layers of squamous epithelia. The intergrity of the epithelia is partially altered.

Of the 21 ESPS patients, 11 were males, and 10 were females; we contacted all patients. The median follow-up period was 30 mo (range: 14-80 mo) and the patients' median age was 25 y (range: 17-45 y). Twelve patients reported habitual shaving of the affected region; a single patient used an electric razor, another waxed, and 10 used a straight razor. All patients were treated with surgical excision (**Figures 2-4**). Disease recurred in two patients, variously in the sternum and inguinal region.

Discussion

PS is rarely reported in extrasacroccocygeal regions. However, to the best of our knowledge the incidence rate of PS has not been defined, possibly due to the diversity of locations at which it can manifest. In our series ESPS, which can be confused with cellulitis and hidradenitis suppurativa, represented 2.2% of the total PS cases. Cases involving the axilla and inguinal region were diagnosed as hidradenitis suppurativa; these lesions were painful but not debilitating and did not lead to psychological discomfort. Recurrence was reported only in lesions of the axilla [2-4]. The lesions we evaluated all included hairs, which made us confident that they were PS. Apocrinitis exhibited histopathological features of folliculitis rather than hidradenitis suppurativa [5]. We suggest that in certain cases, PS may be a component of hidradenitis suppurativa. Cellulitis was rediagnosed in one case of scalp lesions. No alopecia scar developed, and there was no recurrence

[6, 7] indicating that cases involving the scalp also appeared to be related to PS.

One patient who presented with an abscess-containing mass in the medial lower quadrant of the left breast was initially provided with unsuccessful medical treatment (**Figure 6**). Therefore, surgical excision was performed, following which histopathological PS was diagnosed. This patient habitually wore a tight brassiere, which could have contributed to disease pathogenesis. PS of the periareolar region has also been characterized as an occupational hazard for hairdressers [8-11], which highlights the importance of breast surgeons' being aware of the occupation of non-lactational breast sepsis patients, particularly when surgery is required. Correct identification alone is not sufficient for surgical management, in this case entailing total duct excision; the patient must also receive advice to prevent recurrence.

PS of the scalp is rare and was observed herein in a male, Turkish patient whose sinus lesion was attributed to his habit of shaving with a straight razor. In a second patient, a cyst was detected attached to the temporal fascia, in which there was a granulomatous reaction. Pseudofolliculitis is frequently observed after shaving in curly-haired individuals who may subsequently develop PS. Two of our patients presented with inguinal lesions, in one of whom a prediagnosis of sebaceous cyst was rendered; hidradenitis suppurativa was diagnosed in the other case. However, histopathological diagnosis confirmed PS. Similar histological features have been reported in cases of hidradenitis suppurativa. Wax and epilation may also be predisposing factors for pseudofolliculitis [5]. One report of ESPS involving the axilla [13] included non-hirsute healthy females between 17 and 30 years of age, in whom friction due to arm movements and perspiration was responsible for the etiology. Other etiological factors included shaving, maceration and minor infections. Two of our patients habitually shaved the axilla (**Figure 5**). ESPS of the anterior abdominal wall, located at umbilical and periumbilical areas, has also been reported [14, 15] in young adult male patients, most of whom exhibited poor personal hygiene, deep navels, and excessive hair [14, 15]. In five of our patients, anterior abdominal wall ESPS lesions occurred external to the navel. Two female patients, aged 23

Extra-sacroccygeal pilonidal sinuses

Table 1. Extra-sacroccygeal location of pilonidal sinus: clinical features

Patient	Gender	Age (y)	Lesion site	Clinical diagnosis	Hair removed	Type of hair removal	Recurrence	Comments
1	F	19	Sternum	SC	-	-	Yes	-
2	M	27	Abdomen	SC	-	-	-	-
3	M	25	Abdomen		-	-	-	-
4	M	20	Abdomen	SC	No	-	No	Granuloma
5	M	29	Neck	SC	Yes	Electric razor	No	-
6	F	27	Groin	HS	Yes	Razor	-	-
7	M	19	Axilla	SC	Yes	Razor	-	-
8	F	26	Abdomen	Abscess	-	-	No	-
9	F	22	Axilla	LN	Yes	Razor	No	-
10	F	28	Groin	SC	Yes	Wax	No	-
11	F	37	Breast	Abscess	No	-	No	-
12	M	42	Axilla	LN	Yes	Razor	No	-
13	F	17	Groin	Abscess	No	Wax	Yes	-
14	F	17	Sternum	SC	Yes	-	-	-
15	M	39	Neck	Abscess	Yes	Razor	No	-
16	F	43	Abdomen	Abscess	No	-	-	-
17	M	27	Scalp	SC	-	-	-	-
18	M	20	Scalp	Cyst	Yes	Razor	-	-
19	M	45	Axilla	LN	Yes	Razor	No	-
20	F	25	Axilla	LN	Yes	Razor	-	-
21	M	27	Groin	SC	Yes	Razor	-	-

HS, hidradenitis suppurativa; LN, lymph node; SC, sebaceous cyst.



Figure 2. Inguinal region.



Figure 3. Neck region.

and 26 years, presented with abscesses in suprapubic areas. These patients also reported shaving the affected areas. Two young male adult patients presented with lesions in the hypogastric region, and a 43-year-old female patient displayed a lesion in the lower left quadrant of the abdomen (**Figure 7**).

We suggest that wearing tight trousers with a belt could also have played a role in the development of inguinal lesions in our patients.

Shaving-related trauma was prevalent among our ESPS patients. During the follow-up period recurrence was observed in only two cases (one of sternal and the other of inguinal lesions) subsequent to surgical excision. We attributed sternal lesions to a combination of large breasts and tight brassieres, leading to increased friction. Considering the similar pathogenic mechanisms underlying sacroccygeal PS and ESPS, recurrence of lesions subsequent to surgical excision is not unexpected.

Extra-sacroccocygeal pilonidal sinuses

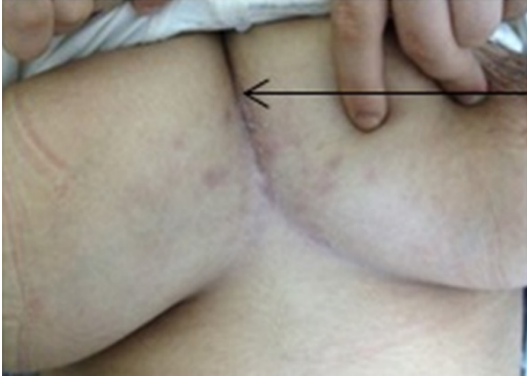


Figure 4. Sternal region.

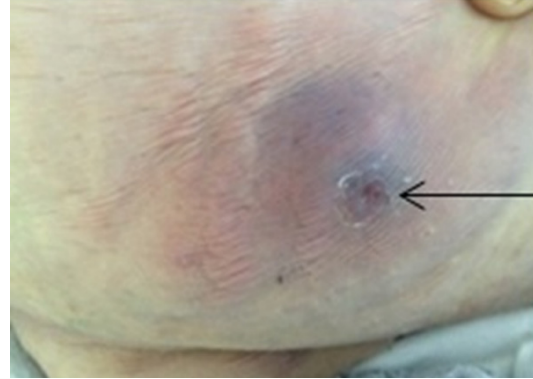


Figure 7. Abdominal region.

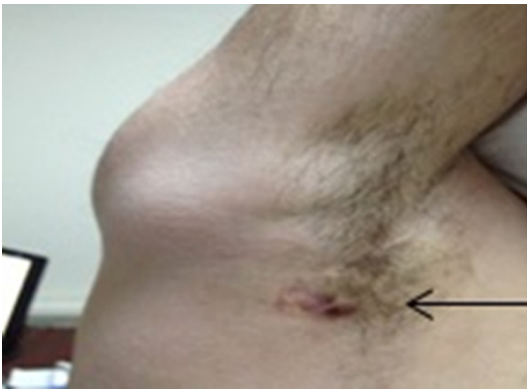


Figure 5. Axillary region.

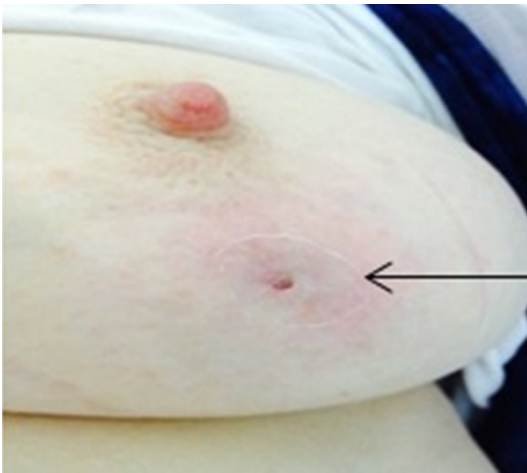


Figure 6. Lower quadrant of the left breast.

Conclusion

Histological features of PS may resemble those of cutaneous lesions, although cutaneous lesions cannot give rise to typical sacrococcy-

geal PS. PS should be considered in the differential diagnosis of extrasacroccocygeal cutaneous lesions. The major pathogenic factor for PS is local and repetitive minor trauma in hairy areas.

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Disclosure of conflict of Interest

None.

Address correspondence to: Dr. Fatih Çiftci, Basak mah, 2.etap D-35/24 Basaksehir/Istanbul-Turkey. Tel: 90 505 616 42 48; Fax: 90 212 462 70 56; E-mail: oprdrfatihciftci@gmail.cm

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