



Pilonidal Disease of the Intermammary Cleft: Report of Two Cases and Literature Review

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Abstract

Although pilonidal disease (PD) characterized by chronic inflammation of the skin and subcutaneous tissue most commonly seen in the intergluteal cleft, it rarely affects the umbilicus, axilla, nose, neck, interphalangeal and toe webs, and breast and intermammary cleft. Herein, we aimed to present two patients with intermammary cleft PD and to provide a literature review on this subject. The first case was a 16-year-old woman who presented to our clinic with discharge from the intermammary cleft that had first started 6 months earlier. Her physical examination revealed two sinus orifices consistent with PD. Several days after drainage and antibiotherapy, the patient was operated and all involved tissue was excised. The second case was an 18-year-old woman referred to our hospital after having undergone several drainage procedures for a pilonidal sinus detected in the intermammary cleft at an outside center. After completing preoperative tests, the patient was taken into operation and all tissues until the anterior aspect of the sternum were excised. The defects of both patients were primarily closed and no recurrent PD developed in any of them. Hidradenitis suppurativa and breast abscesses should be considered in the differential diagnoses of PD localized to the intermammary cleft or breast tissue. Drainage and antibiotherapy are the most appropriate approaches among cases presenting with abscess formation. As cosmetic issues are more of a concern with intermammary cleft PD, excision and primary closure technique may be prioritized instead of flap techniques in chronic PD.

Keywords Pilonidal disease · Unusual location · Breast · Intermammary cleft

Introduction

Pilonidal disease (PD) is a chronic recurrent disease characterized by inflammation of the skin and subcutaneous tissue triggered by hair in folded parts of the human body [1–3]. Despite most commonly seen in intergluteal cleft also known as sacrococcygeal region, PD rarely involves umbilicus, axilla, nose rim, interphalangeal and toe webs, and breast and intermammary cleft [2, 4, 5]. PD localized to the intermammary cleft or breast tissue is extremely rare. To our knowledge, a limited number of case report have been published so far. Therefore, a general consensus on the management of intermammary cleft PD has not yet been established. Hidradenitis suppurativa and breast abscesses should be considered in the differential diagnoses of PD localized to the

intermammary cleft or breast tissue. Its treatment varies by its localization and extension of the diseased tissue. Herein, we aimed to report how two female patients with PD in the intermammary cleft were managed. We also attempted to make a medical literature analysis on breast and intermammary PD by reviewing some studied on this subject using some databases.

Material and Methods

The primary objective of this study is to share two patients who underwent surgical treatment due to pilonidal disease located at the intermammary cleft. The secondary aim is to analyze the previous published articles related with pilonidal disease located at the breast or intermammary cleft. We searched for previously published articles using different combinations of key words, including breast, mammary, intermammary sulcus, intermammary cleft, and sternum, in PubMed, Medline, Google Scholar, and Google databases.

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The publication language was not an exclusion criterion, and studies published before 1 March 2019 were included.

Results

Case-1

A 16-year-old female patient presented to our clinic with fluid discharge from the intermammary cleft. She stated that that complaint had started 6 months earlier, for which she had been prescribed antibiotics and had undergone abscess drainage for a single time. On physical examination, there was a PD presenting with two separate orifices in the intermammary cleft (Fig. 1). No PD was present in the intergluteal cleft or sacrococcygeal region. During the preoperative preparation period, the orifices of the sinuses were dilated and antibiotic therapy was administered. The patient was operated a few days later. After injecting methylene blue through both orifices, an elliptic incision was made to excise all soft tissues until the anterior aspect of the sternum (Fig. 2). Then, the resulting defect was primarily closed (Fig. 3). No complication or PD recurrence was observed during a two-month follow-up postoperatively. Histopathological findings were compatible with PD.

Case-2

An 18-year-old female patient had presented to a primary care center with redness and swelling of the intermammary sulcus. She had undergone several drainage procedures and antibiotic therapy. She was referred to our center for a definitive therapy. On physical examination, there was an appearance consistent with a pilonidal abscess (Fig. 4). First, pilonidal abscess drainage



Fig. 1 Two separate sinus orifices appear in the intermammary sulcus



Fig. 2 After methylene blue injection, an elliptic incision was made to excise all soft tissue

was performed and antibiotherapy was initiated. After completion of preoperative tests and healing of pilonidal abscess, the patient was operated. Following the administration of methylene blue from the sinus orifice, an elliptic incision was made to make an excision that involved the whole granulation tissue. Then the resulting defect was primarily closed. No PD recurrence was observed during the follow-up. Histopathological findings were compatible with PD.

Discussion

Background and Etiopathogenesis

PD was first described in 1833 by Mayo as a hair-containing sinus in the intergluteal cleft [6]. In 1880 Hodges combined the Latin term pilus for hair and nidus for nest to form the currently



Fig. 3 Intraoperative view of primary closure technique



Fig. 4 Pilonidal abscess appear in the intermammary sulcus

used term of pilonidal sinus [3, 6]. After the first description of PD, some congenital factors have been implicated in its etiology for years [3]. Although some theories regarding congenital causes have been put forth, it is recently recognized that acquired factors such as increased prevalence in males, young people, and some professionals, and the absence of hair follicles on sinus walls plays a much more important role in the development of the disorder [3]. The main predisposing factors for sacrococcygeal PD include male sex, obesity, local trauma, sedentary lifestyle, professions requiring prolonged sitting, deep intergluteal cleft, increased hair density in folded body parts, excessive sweating, young age, poor hygiene, and familial predisposition. PD is usually seen in post-pubertal young adults [1]. It affects men one to four times more commonly than women [1, 3, 6, 7]. The highest incidence of the disease being at 15–30 years and its rarity after 40 years are explained by increased sex hormones after puberty affecting pilosebaceous glands to increase their secretions [1, 6].

A body region does not necessarily need to be hairy in nature in order to be involved by PD. PD may develop from hair particles falling off the neck or head region onto women's intermammary region or customers' hair particles falling onto barbers' interphalangeal regions [6–8]. Although PD mostly involves the intergluteal cleft located in the sacrococcygeal region where exposure to microtrauma is common, it may involve almost all regions where hair particles can penetrate the skin [5]. Rare locations for PD include umbilicus, subungual region, perialar region, biparietal region, interphalangeal region, penis, clitoris, anal canal, neck, suprapubic region, axilla, breasts, and intermammary cleft [1, 4, 6].

Differential Diagnosis

The most notable disorders in the differential diagnosis of breast and intermammary cleft PD are hidradenitis suppurativa and

breast abscesses [9]. Histopathologically, PD is characterized by a sinus, a sinus tract opening to outside, granulation tissue inside the sinus tract, one or more hair particle(s), and inflammatory cell infiltration inside the tract. Hidradenitis suppurativa is a chronic and recurrent inflammatory disease of the apocrine sweat glands, mostly located in the intertriginous regions where the apocrine sweat glands are located. Although the disease is typically seen in the axilla, inguinal, and perineal regions, it may rarely involve the perianal region, inner aspects of thighs, genital region, breasts, and intermammary region [9]. Hidradenitis suppurativa is histopathologically characterized by follicular hyperkeratosis, active folliculitis or abscess formation, sinus tract formation, fibrosis, granuloma formation, apocrine and eccrine glands' inflammation, fat necrosis, and subcutaneous tissue inflammation [9]. Hence, when the differential diagnosis of discharging lesions and abscesses involving these regions are concerned, a combined assessment of clinical signs and histopathological features is of paramount importance for both treatment planning and preventing disease recurrences [4].

Literature Review

There is a limited number of published case reports about PD located in the breast or intermammary cleft. A total of 21 articles/poster titles in which 53 cases were presented up to date [1–8, 10–22]. The full-text versions of 18 of these articles/posters were accessed whereas full-text versions of three other articles were not available. The disease was localized to the intermammary cleft in 40 patients; breast tissue in nine (one of them was TRAM); and anterior sternum in the remainder four patients; anterior sternum actually corresponds to the intermammary cleft. Only one of the patients was male who was working as a barber. The details of the literature analysis were provided in Table 1.

Possible Risk Factors for Breast and Intermammary PD

Obesity, large breasts, wearing tight bras, and having some risky professions like hairdressing are reported to be possible risk factors for intermammary cleft PD [1, 2, 7, 13, 20]. Shareef and colleagues [1] reported that 12 of their patients had a BMI ≥ 30 kg/m², and most wore tight bras to make their breasts look small. Since tight bras bring both breasts together, they form a deep sulcus between the breasts, just like the one in the intergluteal area, thereby forming a suitable region for hair particles falling off higher body regions to penetrate the skin [1, 20]. Both patients in our report had large breasts and wore tight bras. Although many studies have suggested a relationship between hirsutism and sacrococcygeal PD, no such relationship has been demonstrated between perimammary PD and hirsutism [1, 6, 12]. Neither of the patients reported in this paper had hirsutism. Although it is a well-known fact for a long time that barbers

Table 1 Summary of 21 published articles related to Intermammary or breast pilonidal disease in the medical literature

References	Years	Article source	Country	Case	Age/sex	Initial complaints	Duration of symptoms	Location	Surgery	Recurrence
Polat	2018	Fulltext	Turkey	1	18/F	Abscess	NA	Intermammary	Semi-open intraflexion	No
Shareef	2018	Fulltext	Iraq	12	13–29/F	Discharge	4 wk–2 years	Intermammary	Excision+Primary repair(<i>n</i> = 9) Excision+Secondary healing(<i>n</i> = 3)	Yes (<i>n</i> = 2) Yes (<i>n</i> = 1)
Hughes	2017	Fulltext	UK	1	38/M	Discharge	20 years	Breast	Excision	NA
Caman	2017	Poster	Turkey	1	16/F	Discharge	1 years	Intermammary	Excision+Karydakias flap	No
Arun	2017	Fulltext	India	10	14–18/F	Discharge	NA	Intermammary	Excision	No
Yakan	2016	Poster	Turkey	2	18/F	Discharge	NA	Intermammary	Excision	NA
					20/F	Discharge	NA	Intermammary	Excision	NA
Ozkan	2016	Poster	Turkey	8	15–28/F	Discharge	3–12 months	Intermammary	Excision	No
Salih	2016	Fulltext	Iraq	1	16/F	Discharge	1 year	Intermammary	Excision	No
Ciftci	2015	Fulltext	Turkey	3	19/F	NA	NA	Sternum	Excision	Yes
					17/F	NA	NA	Sternum	Excision	No
					37/F	Abscess	NA	Breast	Excision	NA
Yavalkar	2014	Fulltext	India	1	20/F	Discharge	6 months	Intermammary	Excision	No
Lahiri	2014	Fulltext	UK	1	28/F	Abscess	4 weeks	Breast	Excision	No
Sunkara	2010	Fulltext	India	1	16/F	Discharge	2 years	Intermammary	Excision	No
Demiralay	2009	Fulltext	Turkey	1	17/F	Discharge	1 year	Intermammary	Excision	No
Ahmed	2002	Fulltext	Ireland	1	14/F	Abscess	6 weeks	Sternum	Excision	NA
Rafi	2002	Fulltext	Pakistan	1	22/F	Discharge	10 years	Sternum	Excision	NA
Ferdinand	1997	Fulltext	UK	1	NA/F	Lumps	8 months	Breast (TRAM)	Excision	NA
Gannon	1988	Fulltext	UK	1	21/F	Abscess (Bilateral)	3 months	Breast	Drainage	NA
Bowers	1982	Fulltext	UK	3	18/F	Lump	NA	Breast	Biopsy+Wool fibers removal	NA
					30/F	Lump	NA	Breast	Hair removal with tweezers	NA
					25/F	Lump	NA	Breast	NA	NA
Lion-Cachet	1971	NA	S. Africa	1	NA	NA	NA	Intermammary	NA	NA
Saggar	1967	NA	NA	1	NA	NA	NA	Intermammary	NA	NA
Kaufman	1961	NA	USA	1	NA	NA	NA	Breast	NA	NA

develop PD of the interphalangeal area, only a few reports of PD cases involving breast tissue and intermammary cleft has been reported so far [8, 13, 15, 18]. It has been demonstrated that short hair particles entering through shirts initiated PD in perimammarian tissues among barbers trimming short-haired customers [8, 15, 18]. A similar risk has been reported for veterinarians, sheep shearers, and groomers [2]. Hence, among patients with risky occupations, PD development may be prevented by using a protective cover [15, 18].

Management

Irrespective of PD's localization, drainage and antibiotherapy are the most appropriate approaches among cases presenting with abscess formation. As for chronic or recurrent PD, excision after

making a differential diagnosis is the most suitable approach. Wound management after excision may be performed using wound closure, delayed wound closure, or one of the various flap-based wound closure techniques (Limberg flap, Karydakias flap, Bascom's cleft-lift procedure, V-Y advancement flap, Z-plasty). As cosmetic issues are more of a concern with mammarian and intermammary cleft PD, excision and primary closure technique may be prioritized for closure instead of flap techniques [6].

Conclusion

PD is a chronic recurrent disease that may involve many body folds, with the sacrococcygeal region being most commonly

involved. The intermammary and breast tissue are regions where PD very rarely involves, and such conditions mostly occur in women with large breasts wearing tight bras. Its differential diagnosis should definitely include disorders characterized by breast abscesses as well as hidradenitis. The most appropriate treatment of acute disease is drainage and antibiotic therapy whereas chronic disease is best treated by excision and primary closure.

Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no conflict of interest.

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