Intermammary pilonidal sinus – A case report

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ABSTRACT

Pilonidal sinus is a blind-end tract lined with granulation tissue, which leads to a cystic cavity lined with epithelial tissue. This case report aims to present a case of pilonidal sinus presenting in an unusual location – the intermammary region, in a 16-year-old female patient who presented with complaints of serous discharge mixed with pus intermittently from the intermammary region for 5 months. Although a pilonidal sinus is most commonly seen at the sacrococcygeal region, the presence of one such entity in the intermammary region of a female represents a rare occurrence with very few cases reported worldwide – more so in young females with large, heavy, and pendulous breasts. A proper surgical repair in terms of its excision and closure remains the mainstay in management.

Key words: *Case report, Intermammary, Pilonidal sinus, Primary excision, Young female*

The name "pilonidal" was coined by Hodge in 1880 from the Latin word pilus, which means hair, and nidus, which means nest [1,2]. The pilonidal disease consists of a spectrum of entities ranging from asymptomatic hair containing cysts and sinuses to a large abscess. Pilonidal sinus is a blind-end tract lined with granulation tissue, which leads to a cystic cavity lined with epithelial tissue. These are usually found in the sacrococcygeal region. However, they may also occasionally occur in the axilla, groin, interdigital web, umbilicus, nose, intermammary areas, suprapubic area, clitoris, prepuce, penis, occiput, or on the feet [3,4].

Intermammary pilonidal sinus disease is seen in fatty females with increased distribution of hairs [5]. It is particularly common in Arab females and in females with heavy pendulous breasts. After the onset of puberty, sex hormones affect the pilosebaceous glands, and, subsequently, the hair follicle becomes distended with keratin. As a result, a folliculitis is created, which produces edema and follicle occlusion. The infected follicle extends and ruptures into the subcutaneous tissue, forming a pilonidal abscess. This results in a sinus tract that leads to a deep subcutaneous cavity [1]. The sinus is caused by the friction of the skin, leading to the embedding of the hair beneath the surface. The hair forms small cavities or pits, which go on to become sinuses. Bacteria and debris enter this sterile area, producing local inflammation and the formation of pus-filled abscesses. In chronic conditions, the sinus becomes an open cavity, constantly draining small amounts of fluid [6,7].

CASE REPORT

A 16-year-old unmarried female presented to us in the outpatient department with a complaint of serous discharge mixed with pus intermittently from the intermammary region for 5 months. There was associated complaint of pain for 1 month at a local site which was dull aching type and intermittent. The patient had no complaint of fever/history of trauma. The patient had attained menarche at the age of 13 years, was non-alcoholic and a non-smoker, and had no other remarkable history.

The patient had normal body temperature, with a pulse rate of 78/min, and a blood pressure of 110/74 mmHg. The patient was conscious, cooperative and had no other remarkable abnormalities in the general examination. Local examination revealed discharging sinus tracts with surrounding induration in the intermammary region. Two sinus tract openings were found; one, approximately 2 cm below the sternal notch in the midline, and the other, about 5 cm below the first opening. Examination of the surrounding region revealed large, pendulous, and heavy breasts. The overlying, as well as surrounding skin, the nipple areola complex, and the axillary region, was found to be normal.

Basic routine blood investigations including the total white blood cell count, hemoglobin, serum creatinine, and bilirubin were done and found to be normal. X-ray chest was found to be normal. Ultrasonography of the local region showed inflamed subcutaneous tissue with no evidence of any underlying collection and no intrathoracic communication. An X-ray sinogram was done, which revealed intercommunicating sinus tracts in the subcutaneous plane over the sternal region, anterior chest wall in the midline (Fig. 1).

Excision with primary closure was done under general anesthesia. Careful dissection of the area was done after cutting of the skin and subcutaneous tissue. Methylene blue dye mixed with hydrogen peroxide was inserted through an infant feeding tube

to identify the tract. Complete excision of the sinus tract *en bloc* was done. Hemostasis was achieved using electrocautery. The subcutaneous layer was closed using vicryl (2-0) in an interrupted manner. Skin closure was done using polyamide black (3-0) in a vertical mattress manner and a sterile dressing applied. The excised tissue was sent for histopathological examination.

The patient was discharged on the 2nd post-operative day after doing the first dressing. Follow-up dressing was then done on every 3rd day, and suture removal was done on the 14th day. The patient was advised to wear appropriate innerwear. The patient was given an oral antibiotic amoxicillin plus clavulanic acid 625 mg BD for 7 days post-discharge along with analgesics diclofenac 50 mg BD. No wound discharge/dehiscence/seroma formation or any other immediate post-operative complications were noted. The histopathology showed sinus tracts with chronic inflammatory tissue, confirming the diagnosis. After suture removal, the patient was kept on a monthly follow-up for 6 months (Fig. 2). No other complications or recurrence was noted.

DISCUSSION

Although a pilonidal sinus is most commonly seen in the sacrococcygeal region, it can occur occasionally at various other locations in the body. The presence of one such entity in the intermammary region of a female represents a very rare occurrence.

Treatment for symptomatic intermammary pilonidal sinus involves surgery to incise and drain the abscess. The surgery can be either wide excision and healing by secondary intention (longer healing time and low chance of recurrence), excision and primary closure by sutures (quicker healing and risk of recurrence), or plastic surgery technique using flaps (for recurring and/ or extensive sinus) [8]. The bother procedures evolving are with topical natural polyphenols/laser epilation [9,10]. Shareef *et al.* in their case series of 12 patients from the Kurdistan region of Iraq [11] reported that up until 2017 only six cases of intermammary pilonidal sinus had been reported in the world [3,5,12-15].

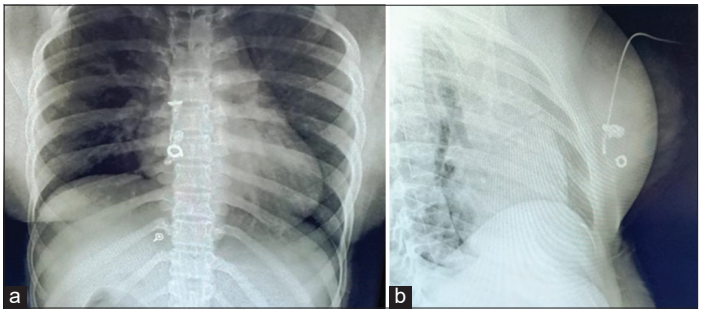

Figure 1: X-ray sinogram (a) anterior view; (b) lateral view

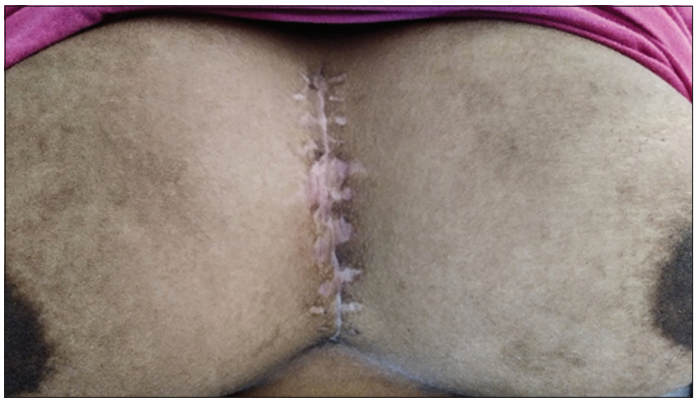

Figure 2: Clinical image at the 6th month of follow-up

The patients presenting with complaints of discharge from an opening on the skin surface require thorough clinical examination along with other imaging modalities or interventional procedures to find the underlying cause and any other systemic disease. Confirmation of the diagnosis can be achieved by the dye study to gauge the extent of underlying tracts. In the case of intermammary pilonidal sinus, the presenting age of the female along with breast architecture can aid in supporting the diagnosis of pilonidal sinus. Pilonidal sinus is a disease of young patients with age around 15–30 years [13,16]. Regarding the age of affection, intermammary pilonidal sinus is consistent with other types of pilonidal sinus.

Sunkara *et al.* likse Salih *et al*. reported the intermammary sinus occurring in 16-year-old female [13,15]. It was reported that hairiness is among the most important risk factor for developing pilonidal sinus. However, this risk factor was not mentioned in the case of the intermammary variety in the literature [2,5,12-15]. Shareef *et al.* in their study confirmed that being hairy is not necessary to develop intermammary pilonidal sinus as all cases did not have hairs in the area of the disease [11].

Hence, for the intermammary region particularly, the term "pilonidal" is perhaps a misnomer as it does not involve the presence of hairs *per se*. Another well-known risk factor for being affected by pilonidal sinus is obesity [17]. This is consistent with the findings of this study, where the body mass index of the patient was 33.3. Thus, the presenting age as well as the built of our patient apart from the investigations contributed in leading us to our diagnosis.

Other differential diagnoses of such a presentation include hidradenitis, chronic osteomyelitis involving sternum, and tubercular abscess, among others. However, the chest X-ray, along with sinogram, helps in achieving the diagnosis of an intermammary pilonidal sinus, along with confirmation by post-operative histopathological confirmation. Some studies including that of Kakamad *et al.* [15] have mentioned the use of a drain to be kept for a few days after surgery. Other surgical options for closure include healing by secondary intention, or as are seen in cases of sacrococcygeal pilonidal sinus, closure with help of flaps – with the Limberg flap being used with quite favorable results [18]. Similar results can be achieved in intermammary cases by mobilization of both breasts. In this case, surgical excision with primary closure was done without the use of the drain. There were no intraoperative or immediate post-operative complications. There was no incidence of any seroma/hematoma formation noted. Suture removal was done on the 14th post-operative day. No recurrence was noted in the 6 months of follow-up.

CONCLUSION

An intermammary pilonidal sinus is an extremely rare condition seen in young, obese females with large, pendulous, and heavy breasts. The presence and ultimately the diagnosis of such an entity are difficult, particularly due to the part of the body that is involved. Hence, clinical features, along with the presenting age and the body morphology of the patient combined with investigation modalities like the sinogram, can aid in the diagnosis.

REFERENCES

1. Chintapatla S, Safarani N, Kumar S. Sacrococcygeal pilonidal sinus: Historical review, pathological insight and surgical options. Tech Coloproctol 2003;7:3-8.
2. Lion-Cachet J. Inter-mammary pilonidal sinus. S Afr J Surg 1971;9:141-2.
3. Murshid KR. Pilonidal sinus: Primary closure with low recurrence. Emirates Med J 1996;14:96-100.
4. Al-Jaberi TM. Excision and simple primary closure of chronic pilonidal sinus. Eur J Surg 2001;167:133-5.
5. Richardson HC. Intermammary pilonidal sinus. Br J Clin Pract 1994;48:221-2.
6. Hull TL, Wu J. Pilonidal disease. Surg Clin North Am 2002;82:1169-85.
7. Surrell JA. Pilonidal disease. Surg Clin North Am 1994;74:1309-15.
8. Ertan T, Koc M, Gocmen E. Does technique alter quality of life after pilonidal sinus surgery? Am J Surg 2005;190:388-92.
9. Aksoy HM, Aksoy B, Egemen D. Effectiveness of topical use of natural polyphenols for the treatment of sacrococcygeal pilonidal sinus disease: A retrospective study including 192 patients. Eur J Dermatol 2010;20:476-81.
10. Abbas O, Sidani M, Rubeiz N, Ghosn S, Kibbi AG. Letter: 755-nm alexandrite laser epilation as an adjuvant and primary treatment for pilonidal sinus disease. Dermatol Surg 2010;36:430-2.
11. Shareef SH, Hawrami TA, Salih AM, Kakamad FA, Rahim HM, Hassan HA, *et al*. Intermammary pilonidal sinus: The first case series. Int J Surg Case Rep 2017;41:265-8.
12. Demiralay E, Höbek A, Altaca G. Inter-mammary pilonidal sinus; an extremely rare location: Case report. BTDMJB 2009;5:78-9.
13. Sunkara A, Wagh D, Harode S. Intermammary pilonidal sinus. Int J Trichology 2010;2:116-8.
14. Yavalkar P, Shelke R. Intermammary pilonidal sinus: A case report. Indian J Med Case Rep 2014;3:20-2.
15. Salih AM, Kakamad FH, Abdulqadr MH. Intermammary pilonidal sinus: A rare presentation. Int J Case Rep Images 2016;7:48-50.
16. Salih A, Kakamad F, Essa R, Aziz MS, Salih RQ, Mohammed SH. Pilonidal sinus of atypical areas: Presentation and management. Pilonidal Sinus J 2017;3:8-14.
17. Sengul I, Sengul D, Mocan G. Axillary pilonidal sinus: Fig. 1: Histological appearance of pilonidal sinus (H and E stain), a case report. N Am J Med Sci 2009;1:316-8.
18. Aithal SK, Rajan CS, Reddy N. Limberg flap for sacrococcygeal pilonidal sinus a safe and sound procedure. Indian J Surg 2013;75:298-301.

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