CASE REPORT

A rare case of vulval pilonidal sinus: incidental diagnosis

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SUMMARY

A 63-year-old woman presented with a 4-week history of vulval bleeding. On examination, an 8 mm fleshy irregular vascular lesion was present on the vulva in the periclitoral area. This vulval lesion was treated by surgical excision. Histological analysis showed irregular pieces of skin partly covered by hyperplastic squamous epithelium. There were areas of fistulous-like endophytic proliferations lined by hyperplastic squamous epithelial cells. The intervening stroma showed granulation tissue with severe active chronic inflammation. At least five hair follicle shafts surrounded by foreign body type giant cells were also identified within the inflamed area. There was no evidence of dysplasia or malignancy. This chronically inflamed fistulous tract together with hair shafts within the wall of the tract were diagnostic of a pilonidal sinus of the vulva. This case report summarises the importance of diagnosing pilonidal sinus at an unusual location.

BACKGROUND

Pilonidal sinus disease is an important cause of morbidity in young adults. Pilonidal disease of the genital tract is very rare and disease involving the vulva extremely rare with only five cases described in the literature. A delay in diagnosis and treatment of this relatively harmless condition can lead to complications from abscess formation. Early diagnosis and complete excision are followed by an excellent prognosis. By writing up this case, we aim to raise awareness of and present further evidence on diagnosing pilonidal sinus at unusual sites.

CASE PRESENTATION

A 63-year-old woman presented with a 4-week history of vulval bleeding. She had a medical history including a compound naevus of the abdomen, a superficial spreading malignant melanoma of the thigh and a 3-year history of Castleman's disease. In addition, the patient had been diagnosed with multiple myeloma 2 years previously, treated by bone marrow transplantation. On examination, an 8 mm irregular fleshy vascular lesion was noted on the vulva in the periclitoral area. This vulval lesion was treated by surgical excision.

Histological analysis showed irregular pieces of skin partly covered by hyperplastic squamous epithelium (figure 1). There were areas of fistulous-like endophytic proliferations lined by hyperplastic squamous epithelial cells. The intervening stroma showed granulation tissue with severe active chronic inflammation. At least five hair follicle shafts surrounded by foreign body type giant cells

were also identified within the inflamed area. There was no evidence of dysplasia or malignancy. This chronically inflamed fistulous tract together with hair shafts within the wall of the tract were diagnostic of a pilonidal sinus of the vulva.

INVESTIGATIONS

The vulval lesion was excised and sent for histopathological examination, which confirmed the presence of pilonidal sinus.

OUTCOME AND FOLLOW-UP

The patient was followed for 1 year and did not have any complications related to this disease.

DISCUSSION

Pilonidal disease was first described by Mayo in 1833, and was named from the Latin word meaning hair nest.¹

In the common sacrococcygeal form, pilonidal sinuses appear as small openings in the intergluteal fold about 3.5–5 cm posterior to the anal orifice. This opening then continues into a sinus tract, which is directed upwards in 93% of the cases.

These lesions are thought to result from excessive and repetitive trauma in these regions such as from friction or shaving.²

Pilonidal sinus has also been described in other anatomical areas where skin folds are prominent, such as the umbilicus, clitoris and axilla.

Pilonidal disease of the genital tract is very rare and disease involving the vulva extremely uncommon with only five cases described in the

Figure 1 H&E stained (×4 magnification) showing inflamed fistulous tract (thin arrow) lined by hyperplastic squamous epithelium (double arrows) with hair shaft (inset: hair shafts highlighted by thick arrow, ×10 magnification).

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literature.^{3–7} The differential diagnosis in these areas include hidradenitis suppurativa (HS) or dissecting cellulitis.²

Two theories of the pathophysiology of pilonidal sinus formation have been proposed: congenital and acquired. The first is an embryonic origin whereby pilonidal cysts and sinuses were thought to be embryological remnants resulting from failed involution of neural tube structures. It is now believed, however, that the majority of pilonidal sinuses have an acquired pathogenesis whereby trauma caused by friction and/or shaving allows hairs to puncture the skin, initiating cyst formation.

Pilonidal disease of genitalia has very rarely been described in the literature, resulting in a paucity of information on how to diagnose, investigate and treat these conditions.

Knowledge regarding pilonidal disease in the vulva is important to allow timely diagnosis to direct optimal management and help prevent complications of recurrent abscess formation

resulting in morbidity from pain, disfigurement and rare cases of associated squamous cell and other carcinomas.⁸

Further studies on the origin and causes of pilonidal sinus are required to prevent delay in the diagnosis and treatment of this relatively harmless condition, which can lead to complications if left untreated.⁹

Contributors AM drafted and revised the paper; MBA proposed the case for submission and revised the drafted paper; PK revised the drafted paper.

Competing interests None declared.

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REFERENCES

- 1 Doll D, Friederichs J, Dettmann H, *et al*. Time and rate of sinus formation in pilonidal sinus disease. *Int J Colorectal Dis* 2008;23:359–64.
- 2 Sion-Vardy N, Osyntsov L, Cagnano E, *et al*. Unexpected location of pilonidal sinuses. *Clin Exp Dermatol* 2009;34:599–601.
- 3 Baker T, Barclay D, Ballard C. Pilonidal cyst involving the clitoris: a case report. *J Low Genit Tract Dis* 2008;12:127–9.
- 4 Werker PM, Kon M. A pilonidal sinus of the clitoris? *Ann Plast Surg* 1990;2:63–4.
- 5 Betson JR, Chiffelle TL. Pilonidal sinus involving the clitoris. A case report. *Am J Obstet Gynecol* 1962;84:543–5.
- 6 Palmer E. Pilonidal cyst of the clitoris. *Am J Surg* 1957;93:133–6.
- 7 Latunde-Dada AO, Lee-Elliott CE, Carpenter TT. A rare case of vulval pilonidal sinus diagnosed on ultrasound. *J Obstet Gynaecol* 2013;33:211–12.
- 8 Pilipshen SJ, Gray G, Goldsmith E, *et al*. Carcinoma arising in pilonidal sinuses. *Ann Surg* 1981;193:506–12.
- 9 Kafali H, Duvan CI, Bayrak R, *et al*. Vulvar pilonidal sinuses: treatment of a rare cause of vulvar pain. *Arch Gynecol Obstet* 2008;278:487–8.

Learning points

- Pilonidal disease is a relatively common condition, but awareness of its presence at unusual locations is important to prevent the complications associated with it.
- Careful histological examination for the presence of hair shafts is important in making the diagnosis.

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