

Case report

Perianal endometriosis: An unusual cause of perianal pain

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ABSTRACT

Endometriosis is a common clinical condition where functional endometrial tissue is found outside the uterine cavity. Although most of this ectopic tissue tends to occur within the pelvis, on rare occasions, they can also be found in unusual locations, such as the thoracic cavity, perianal, episiotomy scar, and previous abdominal wound sites. This is a case report of a healthy 41-year-old Chinese female who presented with a painful perianal lump which was subsequently investigated and excised. The diagnosis of perianal endometriosis was made upon histological examination of the tissue. Many theories attempt to explain the phenomenon of endometriosis in the perianal region. In this case, the implantation theory is the likely explanation as the patient had a significant history of 3 normal vaginal deliveries which were followed by episiotomies. Advancements in endo-anal ultrasound (EAUS) have aided in the planning of surgery for this lesion.

1. Introduction

Endometriosis is a common clinical condition where functional endometrial tissue is found outside the uterine cavity. Conservative estimates worldwide seem to suggest that up to 3%–10% of all women have some form of ectopic endometrial tissue at some point of their lives.¹ Although majority of this ectopic tissue tends to occur within the pelvis, they have also been found in unusual locations such as the thoracic cavity, perianal region, episiotomy scar, and previous abdominal wound sites.²

In particular, perianal endometriosis is a rare form of endometriosis. Since the first reported case in 1923, fewer than 50 cases of perianal endometriosis have been recorded in literature.³ Here we present an unusual case report of perianal endometriosis in National University Hospital, Singapore.

2. Case report

We describe a healthy 41-year-old gravida 3 para 3 Chinese female

who presented to the Emergency Department with symptoms suggestive of haemorrhoids. The patient had an unremarkable medical, surgical and family history. The patient complained of a painful perianal lump present for the past 12 years since the birth of her third child. The lump became symptomatic over the past one year with worsening pain, enlargement, and bloody discharge. These symptoms were cyclical in nature and associated with the patient's menstrual periods.

The patient had a significant gynaecological history of chronic menorrhagia complicated by dysmenorrhea on follow up with a gynaecologist but did not have any prior imaging performed. The patient did not complain of dyspareunia, dyschezia, intermenstrual bleeding, or post-coital bleeding. There was no family history of endometriosis as well. The patient was pain free between menstrual periods and had 3 children born via normal vaginal delivery where a midline episiotomy was performed at each instance. There were no other perineal lacerations during each delivery.

Examination of the lump revealed a tender fluctuant mass at the 11 o'clock region that was 2cm × 2cm in size. Digital rectal examination

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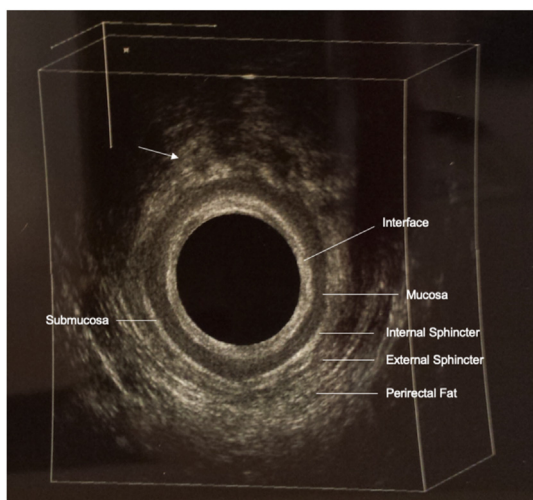


Fig. 1. Endoanal ultrasound (EAUS) image showing a perianal mass at 11 o'clock sparing the anal sphincters.

confirmed that the fluctuant mass was at the anal verge, but did not reveal anything else. Endoanal ultrasound (EAUS) was also performed, which showed a perineal mass at 11 o'clock, sparing the anal sphincters (Fig. 1).

The patient subsequently underwent an excision of the lump. Intra-operatively, perianal scar tissue with granulation was found at the 11 o'clock position, 3cm from the anal verge and superficial to the anal sphincters. There was minimal fluid under the mass and no pus was noted. There were no fistulas, sinus tracts or masses found on per-rectal examination. The lump was sent for subsequent histological examination, where endometrial-type stroma was found to be present together with anal mucous glandular tissue at the anal squamo-columnar junction, confirming the diagnosis of endometriosis.

The patient recovered uneventfully from the surgery. At follow up in our clinic one month after the surgery, the wound had healed well and there were no post-operative complications.

3. Discussion

The first reported perianal endometriosis was first reported by Schickele in 1923.⁴ Since then, there have been occasional case reports. However, the true incidence of this rare condition is unknown.^{3,5-7}

Perianal endometriosis is clinically hard to diagnose solely based on history and physical examination alone. Often, even experienced clinicians have failed to make this diagnosis in the presence of a classical history of cyclical pain, discharge and change in size of a perineal lesion.⁵ Often, diagnosis is only made retrospectively when histopathology of the tissue reveals typical endometrial-like stroma⁸ (Fig. 2).

Our patient displayed symptoms related to the menstrual cycle, which has been also seen in some newer case reports and series reported in China.⁷ A triad of symptoms which include a concordant history of past perineal injury (such as an episiotomy) during vaginal delivery, a palpable firm tender nodule or mass at area of previous injury, and progressive cyclical pain and swelling during menses has been proposed by Zhu.⁹ In the study, Zhu⁹ demonstrated a diagnostic accuracy of 100% for perineal endometriosis, although further studies would need to be done to validate this criteria.

Various theories have been proposed attempting to account for possible reasons why the phenomenon of endometriosis occurs. Common ones include the implantation theory (retrograde menstruation due to tubal reflux of menstrual tissue), coelomic theory (mesothelial metaplasia), transplantation theory (lymphatic, vascular, iatrogenic transplantation) and the altered immunological theory (altered recognition allowing for emboli acceptance¹⁰).

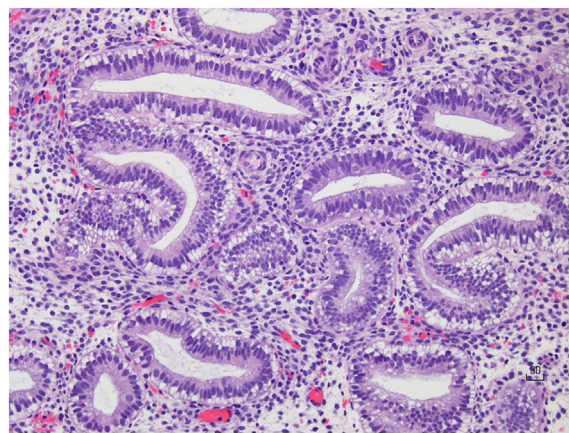


Fig. 2. Typical histopathological image of endometrial-type stroma.⁸

Given that our patient had a significant obstetric history of 3 previous normal vaginal deliveries with episiotomies, the transplantation theory would be a more favourable reason behind the peri-anal presence of the endometrial tissue. Since an episiotomy was performed at each delivery, this would have increased the patient's risk of developing perineal endometriosis as most reported cases seem to suggest a positive correlation.¹¹ However, it is interesting to note that the endometriosis was found at 11 o'clock in the lithotomy position, away from the episiotomy site which was in the midline. Despite much interest in the condition, there has yet to be conclusive evidence to suggest that one theory is more likely than the other as no single theory has been able to account for all the reported cases of extra-pelvic endometriosis.

Advancements in endo-anal ultrasound (EAUS) have aided in the planning of surgery for perianal and anal lesions. Its practical use lies in determining the nature of the lesion, assessing for sphincter involvement, and ruling out other possible differentials; such as a perianal abscess or haemorrhoids.

As shown in the case series from Peking Union Medical College, up to 40% of their cases of perineal endometriosis involve the anal sphincter. Proper imaging of the lesion is thus all the more essential in the pre-operative assessment of the patient to ensure good outcome with a sufficiently wide resection.¹² Incomplete or narrow resections would otherwise result in recurrence.⁶

In our case, the patient underwent a pre-operative EAUS performed at the bedside which enabled good visualisation of its location and for the presence of fistulas and connecting sinuses to be ruled out, allowing for a wide excision of the lesion to be planned. At two years of follow up, the lesion has not recurred.

4. Conclusion

Although rare, perianal endometriosis should be a differential diagnosis considered in the evaluation of a perianal mass with uncommon features. Peri-operative EAUS is an important assessment way that is essential in guiding operative management of this condition.

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Ethical statement

The subject gave informed consent and patient anonymity was preserved. An Ethics Committee approval is not required.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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