CASE REPORT

Perineal scar endometriosis: a comparison of two cases

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SUMMARY

Perineal scar endometriosis is a devastating illness with grave consequences. An early diagnosis is imperative as a delayed diagnosis results in extension of the disease process causing increasing damage to adjoining structures like the anal sphincter and rectum. A late recognition of the disease also prolongs the agony of the patient owing to its being a very painful condition. Two cases are presented of parous women in their early thirties who had episiotomy scars preceding the development of endometriosis. The cases were treated by a perineal incision and removal of suspicious structures. However, a recurrent endometrioma was formed in one case, which had to be excised again. In the other case, symptoms of recurrence had begun, which subsided after a dose of GnRh analogue (leuprolide acetate 3.75 mg). Both patients are on follow-up and are fine until now.

BACKGROUND

Perineal endometriosis is a rare disease. In a study in the Peking Union Medical College & Hospital, about 17 263 women were treated for endometriosis between January 1992 and April 2011. They reported an incidence of only 3.7% for perineal endometriosis of the total number of women with endometriosis.¹ Perineal scar endometriosis is an extremely painful condition, but still a diagnostic dilemma. Diagnosis is often delayed, resulting in prolonged agony to the patient and increased morbidity. The case is being presented to familiarise my colleagues with the presentation of this disease. The ultrasound pictures of these two cases are so lucid that once seen they cannot be forgotten.

CASE PRESENTATION

Case A

A 30-year-old woman, para 2 abortion 0, presented in the outpatient department on 6 February 2012 with a history of perineal pain over the past

4 years. She delivered 6 years ago and remained well for 2 years before she started having pain at the earlier site of episiotomy. The pain became very severe around the time of menses. On examination, the episiotomy scar was seen on the left side of the perineum. On palpation with two fingers, including one inside the vagina, a hard subcutaneous nodule was felt at the episiotomy scar site. A clinical diagnosis of scar endometriosis was considered.

On transperineal ultrasonography, the report stated that the perineal region of the left side showed a hypoechoic lesion measuring 11×7 mm with peripheral vascularity probably a subcutaneous abscess (figure 1).

The patient was taken up for surgical exploration on 11 February, 2012 under sedation with pentazocine and promethazine and local anaesthesia. A transperineal incision was made. A cystic fluctuant swelling was seen in the deep subcutaneous tissue. The anterior wall of the cyst was removed and a dirty black coloured fluid oozed out. The remaining bits of the cyst wall were removed in bits, the cavity packed and left to granulate.

On histopathology, our pathologist reported that the sections showed normal epidermis in the subcutaneous area. Besides, the muscle is a haemorrhagic necrotic area bordered by chronic granulation tissue with no granulomas and no endometriosis. She proposed a diagnosis of a non specific abscess in subcutaneous region. Conclusion—non specific abscess.

After about 7 months, the woman reported on 13 September, 2012 with severe pain at the site of excision. No local signs of redness or swelling were seen, and a hard nodule was felt on the dimple formed by earlier excision on the perineum.

On ultrasonography,—focal thickening and a hypoechoic texture of the skin on the left side of the perineal region were seen. Also, there was a focal lesion with small cystic spaces in the subcutaneous

Figure 1 Case A – Scan showing a hypoechoic area in the left perineum.

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Figure 2 Case A – Scan showing a focal lesion in the left perineum, recurrent endometrioma.

plane of the perineal region, measuring 16×14 mm, which most likely was of granulation tissue (figure 2).

A course of anti-inflammatory drugs was prescribed. However, the pain became more intense and excision was planned under short duration ketamine anaesthesia. A transperineal incision was made and a well-circumscribed mass, approximately 3 cm×3 cm, was removed. The dissection was so deep that we were afraid we would end up opening the rectum.

On histopathology of the mass, the sections revealed muscle bundles and fibroconnective tissue with many foci of endometrial glands with stromal elements. There was no atypia.

Conclusion—Endometriosis (figures 3 and 4).

On receiving the histopathology report, the patient was given a dose of 3.75 mg leuprolide acetate on 24 December 2012.

Case B

A 31-year-old woman presented on 12 September, 2012 with agonising pain at the site of the earlier episiotomy. She was para 1, and her last delivery had occurred 7 years previously. She reported pain for the last 2 years, which worsened during menstruation.

Figure 3 Case A – Histopathology slide of tissue from recurrent endometrioma from left perineum.

On inspection of the perineum, there was no redness or obvious local swelling. On deep palpation, a thickened indurated nodule was felt at the scar site.

Transperineal and transvaginal ultrasonography was performed.

The report stated: uterus, ovaries normal. No adnexal lesion seen.

A heterogeneous cystic and soft tissue area was seen in the subcutaneous plane in the perineum and posterior vulva measuring 13×15 mm, a probable diagnosis of scar granuloma was suggested (figure 5).

Exploration was performed on 19 September, 2012. There was a cystic mass in the subcutaneous plane measuring around 2 cm×1 cm. Some black coloured fluid oozed out. The mass was excised and a rim of adjoining healthy tissue was removed.

The histopathology report read as follows:—sections show a cyst in the subcutaneous plane lined by a transitional epithelium with apocrine differentiation at places. No atypia was seen.

Conclusion—Benign cyst consistent with apocrine hydrocystoma.

Our patient reported again on 28 December 2012 with recurrence of severe pain. Examination revealed tenderness and thickening in the middle zone of the perineum, just medial to

Figure 4 Case A – Histopathology slide of tissue from recurrent endometrioma from left perineum.

Figure 5 Case B – Scan showing heterogenous area in subcutaneous plane in right perineum.

the earlier site of excision. A histopathology review was asked for and the revised report was given.

The tissue in the subcutaneous plane includes skeletal muscle and fibrous tissue, among which a few endometrial glands along with scanty stromal cells are identified. Also, there is a dilated apocrine gland. No atypia was seen.

Conclusion—Endometriosis (perineum) (figure 6).

Considering the possibility of an impending recurrence, the patient was given a single dose of leuprolide acetate 3.75 mg. The effect was dramatic. Within 5 days, the pain and underlying palpable induration subsided.

OUTCOME AND FOLLOW-UP

Cases A and B are being followed up. After excision and medical therapy with gonadotropin releasing hormone analogue therapy, the women are symptom free. More than 4 months have elapsed and both women are doing fine.

DISCUSSION

Perineal endometriosis is often diagnosed late. These two patients had been to various clinics before coming to us. The chief reason for the delay in recognition of the illness was that very few visible signs were present on local inspection of the perineum in both our patients. There was no discolouration of

the perineal skin, no local swelling or periodic leakage of dark coloured fluid. In some cases, bloody discharge oozing out from the scar has been reported,² while a visible swelling was present in one case.³

The tender nodule and cystic swelling in both women could be felt only on deep palpation and many a time a hurried examination in the outpatient can miss it.

Various ultrasonic modalities like transperineal, transvaginal and endovaginal sonography have been used to aid the diagnosis. Our ultrasonologists were not familiar with pictures of perineal endometriosis and hence described the lesion in several ways. A hypoechoic cystic lesion was reported in case A, whereas a heterogeneous mass with cystic and soft tissue areas was reported in case B. These ultrasonic findings matched with those in another study.¹

Three typical characteristics of perineal scar endometriosis have been described, which are elucidated below.

1. Past perineal tear of episiotomy during vaginal delivery.
2. A tender nodule or mass at the perineal lesion.
3. Progressive and cyclic perineal pain. If these criteria were met, the predictive value of perineal endometriosis was 100%.⁴

All these criteria were met in our patients.

MRI and CT have also been used for the diagnosis of perineal endometriosis.

Ca 125 has also been measured in cases of perineal endometriosis. However, it was raised only in 6.7% of patients.⁴

Even on histopathology, there was delayed recognition of the disease. In case A, a diagnosis of pyogenic abscess was considered in the beginning. An endometrioma was reported when recurrence occurred; the mass was excised again and sent for reexamination. In case B, an apocrine cystoma was reported and the endometrial glands were searched and reported when a review of the sections was asked for, because of the recurrence of signs and symptoms.

It is because of the rarity of these cases that perineal endometriosis was not suspected initially.

In perineal endometriosis, there is a very high frequency of recurrence and also a propensity for malignant change, though rare. Therefore, exploration and excision of the endometriotic mass should be performed under general anaesthesia. Wide excision with a 0.5–1 cm cuff of surrounding healthy tissue should be removed.

To prevent recurrence, various authors have reported using the gonadotropin-releasing hormone analogue with benefit.¹ In both our cases, a single dose of 3.75 mg leuprolide acetate was helpful. Preoperative gonadotropin-releasing hormone analogue has also been used to shrink a large mass prior to excision to conserve neighbouring structures like the anal sphincter.¹

Figure 6 Case B – Histopathology slide of a tissue from the right perineum episiotomy site.

In some studies, a coexistence of perineal and pelvic endometriosis has been reported.⁵ In both our patients, there was no clinical or ultrasonic evidence of associated pelvic endometriosis. These two cases of perineal endometriosis are being presented to highlight this agonising but enigmatic disease, and to be considered for insertion in postgraduate books by my senior colleagues to acquaint the budding gynaecologists.

Learning points

- ▶ Perineal endometriosis should be suspected whenever a woman with prior episiotomy complains of periodic perineal pain coinciding with her menstrual cycle.
- ▶ Transperineal ultrasonography is the foremost investigation of choice.
- ▶ Excision of the endometrioma should be performed under general anaesthesia.
- ▶ The patient should be kept on follow-up and warned about the chance of recurrence and the remote possibility of a malignant transformation.

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Competing interests None.

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