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Spontaneous abdominal wall endometrioma: A case report

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ABSTRACT

INTRODUCTION: The abdominal wall is the commonest site of extra pelvic endometriosis (endometrioma), defined as the presence of functional endometrial gland and stroma outside the uterine cavity. Spontaneous abdominal wall endometriosis (AWE) represents an ectopic functional endometrial tissue situated superficial to peritoneum in a scar less abdomen. Spontaneous AWE is rare, accounting for 20% of all abdominal wall endometriosis. It is unfamiliar to general surgeons because of a large number of potential pitfalls in its diagnosis.

CLINICAL PRESENTATION AND INTERVENTION: We report a case of spontaneous abdominal wall endometriosis (endometrioma) presenting to our general surgery clinic with a painful nodular mass in hypogastric area below umbilicus. The initial diagnosis was a desmoid tumour of the anterior abdominal wall. Pain was a remarkable complaint in our patient. Abdominal wall endometrioma was diagnosed by histopathology postoperatively. Excision planned, during operation, one mass was spotted and excised within healthy limits. Histopathology revealed: endometrial glands surrounded by mantle of endometrial stroma, few scattered hemosiderin laden macrophages and fibrous scar tissues. Our patient had no previous scar. She was discharged from hospital on 5th postoperative day uneventfully. (On account of uncontrolled DM and Hypertension). One year of follow up after the surgery, she is free from disease and no recurrence has been observed.

DISCUSSION: Endometriosis is characterised by the presence of endometrial glands and stroma outside the uterine cavity with the maximum prevalence reported in the 4th decade of life. Most cases of spontaneous endometriosis occur in a scar less abdomen. Multiple imaging techniques have been used and described for its diagnosis. The alleged aetiopathogenesis of spontaneous endometrioma is still debatable. It is usually diagnosed by the histopathology and the preferential treatment in cases of endometrioma is total excision of the mass. The hormonal therapy can be added to surgical excision if there is proven pelvic endometriosis. The surgical excision should be wide enough to prevent its recurrence.

CONCLUSION: Spontaneous abdominal wall endometriosis is an extremely rare gynaecological entity, accounts for 18–20% of all abdominal wall endometriosis. The diagnosis of abdominal wall endometrioma is hardly established prior to surgery. The triad: mass, pain and cyclic symptom aids in diagnosis, unfortunately cyclic symptom is not present in all cases (as in our case). Spontaneous abdominal wall endometrioma is usually diagnosed by high index of clinical suspicion and histopathology. The results of imaging techniques are nonspecific. It may pose a diagnostic dilemma due to its rarity and atypical presentation. The preferential treatment of choice is wide excision.

1. Introduction

Abdominal wall endometriosis (AWE) represents a non-neoplastic, ectopic or heterotopic growth of endometrial gland and stroma outside the uterine cavity [1]. Endometriosis is a common gynaecologic entity, with incidence of 8–18% of all menstruating women. In most cases it is located within the pelvis. Endometriosis is found in up to 24% of all gynaecological laparotomies [2] and presents clinically with dysmenorrhea, dyspareunia, infertility and occasionally symptoms and signs of acute and chronic abdomen. The endometrial implants may be categorised as cystic, mixed or solid, with the cystic implants being most common. Although,in some cases endometrial implants may occur spontaneously. Endometriosis can be intra or extra pelvic in location. Most cases are intra pelvic, usually involving ovary, pouch of Douglas, pelvic peritoneum, uterosacral ligament, urinary bladder, rectum, broad and round ligament. Extra pelvic implantation of endometrium has been reported in every organ of body, includes skin, spleen, liver, lung, pleura, brain, extremities, anterior abdominal wall and umbilicus. Abdominal wall is a common site of extra pelvic endometriosis, usually develops in an abdominal surgical scar. This ectopic presence of endometrial tissue occurs in the abdominal wall in 0.03–1.08% of women with history of obstetric or gynaecological procedures. Spontaneous abdominal wall endometriosis (AWE) occurs in a scarless abdomen accounting for

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20% of all AWE. The diagnosis can be very challenging in patients with nonspecific symptoms. Endometriosis is rarely seen by general surgeons and is often diagnosed on histological examination postoperatively [3]. Such cases are poorly reported in the literatures. The aim of this case report is to remind that some very rare site may be involved in endometriosis. The case report is in line with SCARE 2018 criteria [23].

2. Historical background

Daneil Schroen, a German physician, first reported endometriosis in 1690. During the mid-part of the 19th century, Rokitansky had a great intuition: endometrial glands and stroma can be present in ovarian and uterine neoplasias. However, using histological parameters of endometrial structure and activity, the first scientist to delineate peritoneal endometriosis under the name 'adenomyoma' was Cullen. On the other hand, Rokitansky was the first to describe a form of adenomyosis (adenomatous polyp). Early description of ovarian endometrioma as 'haematomas of ovary' or 'chocolate cysts' date back to the end of the 19th century. The first mention of an 'ovary containing uterine mucosa' was published in 1899 by Russel, but Sampson was the first to demonstrate specific endometrial activities, such as desquamation at the time of menstruation and decidualization in pregnancy, subsequently, he presented a theory on its pathogenesis.

3. Case history

A 42 year-old Hindu women of Indian nationality without any previous operative history, presented to surgery outpatient clinic with 5-month history of focal non-cyclic pain and a palpable nodular mass, located in hypogastric area below the umbilicus in the midline. She stated that she had continuous dull ache pain in swelling, not related to onset of her menstrual period. Pain was a remarkable complaint in our patient. She has also been suffering from diabetes mellitus, hypertension and hypothyroidism, and on regular medications for her comorbidities. At physical examination, a nodular tender mass of 3 × 2 cm was identified and fixed to rectus sheath. A clinical diagnosis of a desmoid tumour was made preliminarily. Biochemical and complete blood counts were within normal Limits. X-Rays abdomen showed no features of intestinal obstruction. The exploration (under general anaesthesia) with transverse incision over the swelling was performed, a firm nodular mass was spotted and excised (Fig. 1). Patient was given injection Ceftriaxone 1gm IV at the time of induction of anaesthesia and one dose postoperatively and IV Paracetamol 1gm-BD for one day. The operating surgeon was the main author (Postgraduate Specialist, Master of Surgery) and patient was managed postoperatively in DSP Hospital, Durgapur.

The cut surface of specimen consists of dense tan-white rubbery tissue with focal haemorrhage and multiple cystic spaces (Fig. 2). The histopathology revealed endometrioma (Figs. 3–5). The patient was discharged on 5th Postoperative day uneventfully on account of her comorbid conditions. (Uncontrolled Diabetes Mellitus & Hypertension) and stitches were removed on 9th postoperative day. One year of follow up after the excision, patient is free of disease and no recurrence has been observed. The patient was followed up at 3-month interval by serial Ultrasonography (abdomen and pelvis) at DSP Hospital.

4. Discussion

Endometriosis is usually located in the pelvic organs and is a relatively common gynaecological problem in women of reproductive age. It can be found at any organs of our body like

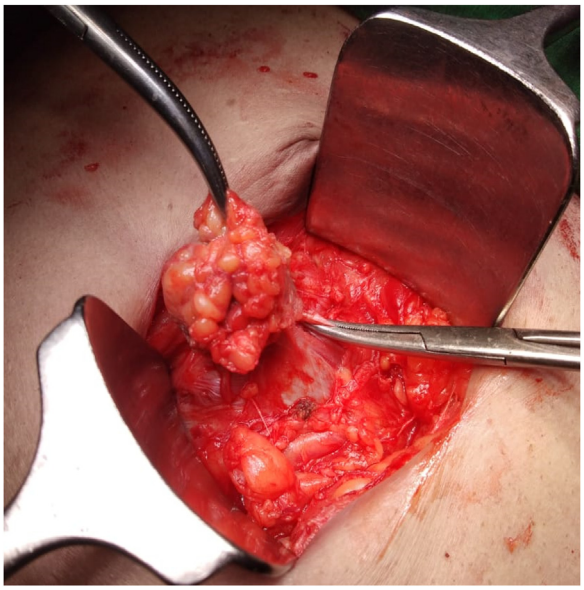

Fig. 1. Endometrioma in parietal wall of abdomen.

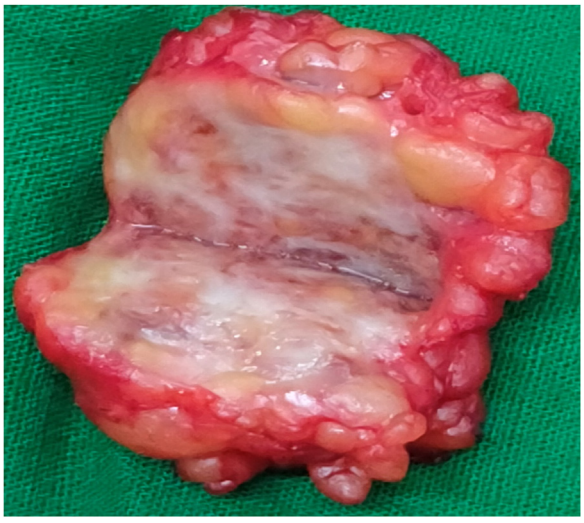

Fig. 2. Resected endometrioma: cut surface consists of dense tan-white, rubbery tissue with focal haemorrhage and multiple cystic spaces.

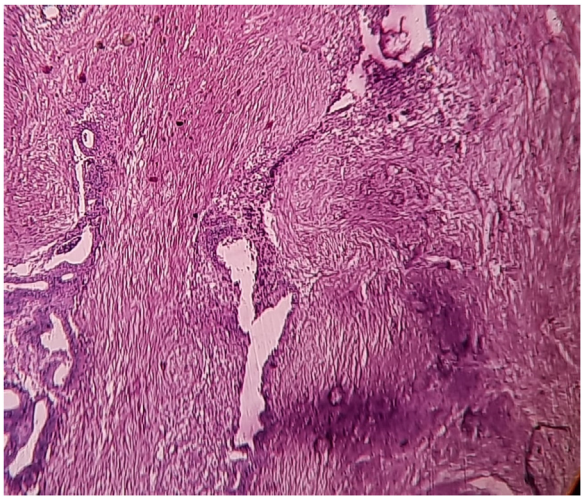

Fig. 3. Haematoxylin and Eosin Stain 10× (Low Power): Endometrial glands surrounded by endometrial stroma embedded in dense abdominal fibrous tissues.

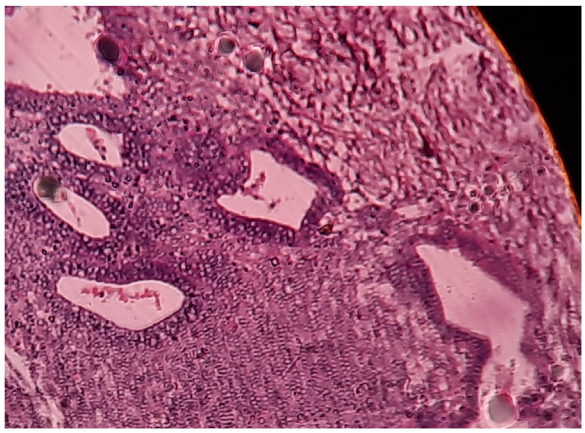

Fig. 4. Endometrial stroma and endometrial glands with signs of intra-glandular haemorrhage. Occasional haemosiderophages seen. Haematoxylin Eosin stain. Magnification: 40× (High Power).

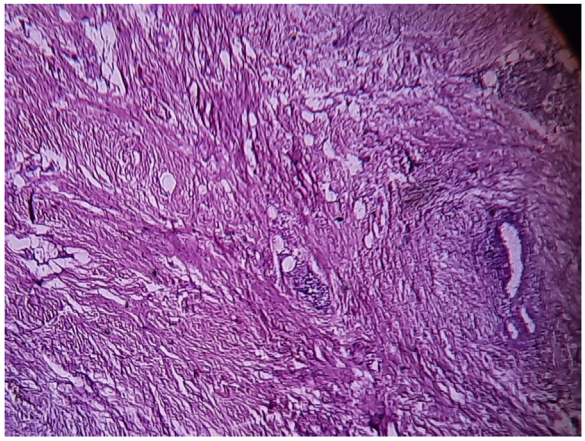

Fig. 5. Scanner view: Endometrial glands and stroma embdded in dense fibrous tissue (Haematoxylin and Eosin Stain).

bowel, urinary bladder, lungs, pleura, ureter, brain, and anterior abdominal wall [4,5]. Abdominal wall endometriosis is most commonly associated with surgical scars, especially caesarean section, laparoscopic surgery and any pelvic surgery [5]. The incidence of abdominal wall endometriosis after caesarean section is 0.03–1.7% of all cases [7]. Spontaneous abdominal wall endometriosis (AWE) is defined as presence of ectopic endometrial glands superficial to the peritoneum in a scar less abdomen [22]. Spontaneous abdominal wall endometriosis (SAWE)) is a rare clinical entity, accounting for 20% of all abdominal wall endometriosis. [5]. Umbilicus is the most common site for developing spontaneous abdominal wall endometriosis, followed by inguinal area and anterior abdominal wall [3,5,10]. Steck and Helwig [6] reported 82 patients with ectopic endometriosis, of which 77 endometriomas appeared in pre-existing scars at the umbilicus. Only five were considered to be spontaneous. Several alleged pathophysiological theories concerning the origin of endometriosis have been postulated, including direct extension, coelomic metaplasia, implantation or reflex, induction theory, lymphatic and vascular metastasis. The Dissemination theory developed by "Halban," proposes that migration of endometrial cells through 'lymph vascular route to different location outside the pelvis [11,12]. The precise pathogenesis of spontaneous abdominal wall endometriosis(SAWE) remains an enigma. In our case spontaneous primary endometrioma was due to vascular or lymphatic metastasis. Two leading theories exist for its pathogenesis. One theory claims that the mesenchymal cells with retained multipotential may under favourable circumstances undergo metaplasia into an endometrioma. The other hypothesis states that endometrial cells may be transported to ectopic sites forming an endometrioma. The extra pelvic endometriosis has been described in nearly all body cavities and organs. But its most frequent locations are umbilicus, inguinal areas and anterior abdominal wall. [3,5,10]. In general, the characteristic clinical symptoms of endometriosis are cyclic pain associated with menstruation. Our patient with abdominal wall endometriosis presented with abdominal pain, not associated with menstrual cycle. The noncyclic nature of pain in endometriosis of abdominal wall has occasionally been reported by other authors. However, it has generally been regarded as atypical presentation, explaining why it is clinically misdiagnosed, as was the case in our patient [8,9,14]. The main symptom is only a palpable mass lesion at the site of maximum tenderness. This palpable mass varies in size following menstrual cycle. Concomitant pelvic endometriosis is the cause of endometriosis in a scar, was reported in 26% of cases [15]. In our case there was no associated pelvic endometriosis. Imaging techniques (Ultrasonography, Computerised Tomography, Magnetic resonance imaging), characteristics of abdominal wall endometriosis are nonspecific, showing a solid enhancing mass in the abdominal wall [9,16–18]. The major role of CT Scan and MRI may be only to depict the extent of disease preoperatively, henceforth a biopsy is essential to reach a definitive diagnosis. FNAC is not diagnostic but have some aid to diagnose scar endometriosis [19]. Two histological features out of three diagnostic triads are required to diagnose endometriosis: endometrial glands, endometrial stroma or hemosiderin pigments (Figs. 3–5). Differential diagnosis of anterior abdominal wall endometriosis especially when symptoms are noncyclic are: abscess, lipoma, suture granuloma, desmoid tumour, sarcoma, lymphoma, incisional hernia or primary and metastatic cancer [7]. The preferential treatment of endometrioma is surgery and diagnosis is confirmed by histopathology. Most of literatures agree that surgical management with wide excision is a key to avoid recurrence and exclude malignancy. Hormonal therapy like oral progesterone, Danazol, gonadotropin-releasing hormone analogue can be added to surgical treatment with proven pelvic endometriosis [20]. Poor results have been observed with Danazol [13], Leuprolide [21] and progesterone [15]. Surgical treatment is preferable and medical management cannot be recommended except in the premenopausal patients. Local recurrence after adequate excision is rare. We suggest that abdominal wall wound should be cleaned thoroughly and irrigated vigorously with normal saline to prevent abdominal wall endometriosis. Diagnostic failure was challenges encountered during case study due to atypical clinical presentation and nonspecific results of imaging techniques, which was overcome by high index of clinical suspicion and histopathology.

5. Conclusion

Spontaneous abdominal wall endometriosis is extremely rare, accounts for only 20% of all abdominal wall endometrioma. The characteristic clinical Triad: mass, pain and cyclic symptoms help in diagnosis, but it is not present in all cases. Spontaneous abdominal wall endometriosis is diagnosed by histopathological examination and the treatment of choice is wide excision. The success rate of medical therapy has been reported to be low, offering only temporary alleviation of symptoms, often followed by recurrence after cessation of the drug. This clinical entity must be included in the differential diagnosis of any abdominal mass in fertile female patients with or without surgical history. The diagnostic and therapeutic delay must be avoided in such an uncommon pathological condition, as endometriosis is a progressive disease, delay in diagnosis

and treatment would increase risk of severe pain, obliteration of pelvic anatomy and infertility. Recurrence of abdominal wall endometriosis is high, especially when first surgery is not appropriate and leave compromised surgical margins.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

As it is a case report, ethical approval is exempted by our institution.

Consent

Written informed consent was obtained from patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in- Chief of this journal on request.

Author's contribution

RAJ RANJAN KUMAR is the chief author of this case report. No other author contributed.

Registration of research studies

Not applicable.

Guarantor

Dr Runu Mukherjee, Joint Director, Durgapur Steel Plant Hospital is the Guarantor for this case report.

Provenance and peer review

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