A rare case: Episiotomy scar endometriosis with anal sphincter involvement

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Abstract
Endometriosis is defined as the presence of functioning endometrial tissue outside uterine cavity. Scar endometriosis being a rare disease is difficult to diagnose. Incisional or scar endometriosis is an even rarer type with an incidence of less than 1%. The quoted episiotomy scar endometriosis incidence is about 0.06-0.07%. Present case is of a 38years old female, married since 15years with one para. She presented with cyclical pain and swelling at episiotomy site. She underwent surgical excision of mass which was suggestive of endometriosis in histopathology.

Keywords: Scar endometriosis, Episiotomy scar endometriosis, Anal sphincter reconstruction.

Introduction
Endometriosis was 1st described by Von Rokitansky 1860 as presence of the functional endometrial tissue outside uterine cavity.1) Various theories of endometriosis are Retrograde menstruation, Direct implantation, Coelomic metaplasia, Lymphatic dissemination, Hematogenous spread, Activation of embryonic cell rest, Metaplasia of urothelium and Hereditary and immunologic factor.2) Scar endometriosis is a rare disease and usually difficult to diagnose. Incisional or scar endometriosis is a rare type with an incidence of less than 1%. The quoted episiotomy scar endometriosis incidence is about 0.06-0.07%. The diagnosis and complete treatment of scar endometriosis can be challenging. Medical and surgical modalities both play an important role in managing cases of endometriosis depending on site of involvement.3)

Case History
A 38-year-old female married since 15yrs, belonging to low socioeconomic status came with complaints of pain and swelling at vulva right side since 3 years, with increased in intensity during menstruation. Pain was initially cyclical and later it was present throughout the menses with increased intensity. The swelling started to appear 2 years back and gradually increased in size to attain the present size of 3x3cms. There was no history of weight loss, loss of appetite, nausea and vomiting. No history suggestive of difficulty in defecation. Patient took treatment at other places but did not get relief. On menstrual history her cycles were regular with average flow. There was associated history of dysmenorrhea during the regular cycles.

Obstetric history, she was para one and had a full term vaginal delivery 12years back. Her past, family and personal histories were not significant. On examination her vitals were stable. There was no lymphadenopathy. Per abdomen examination was normal, inguinal lymph nodes were not palpable. Her local examination revealed a tender mass of 3 cm x 3 cm on right side of anus which was hard in consistency, with irregular margins. The skin over the mass was pigmented and puckered. On per speculum examination, cervix and vagina was healthy. Per vaginal examination, uterus was normal in size, freely mobile, no tenderness, both fornices were free. On per rectal examination was same and rectal mucosa was loculated. The differential diagnosis thought were scar endometriosis, abscess, granuloma or malignancy. Along with necessary routine investigations, the investigations like FNAC and MRI Pelvis and Perineum added in diagnosis.

On MRI, the lesion measured approximately 2cm x 3.1cm in transverse and antero-posterior dimensions and extended craniocaudally approximately 3.6cm (Fig.1). The lesion showed spiculations, irregular margins with peripheral strands. The lesion also involved the right external anal sphincter and puborectalis muscles (Fig. 2).

Fig. 1: On MRI, lesion measured approximately 2 cm × 3.1 cm in transverse and antero-posterior dimension

Fig. 2: Lesion showed spiculations, irregular margins with peripheral strands
A decision to excise the lesion and to reconstruct the sphincter was taken. Intra-operatively, a 3 cm fibrotic mass involving the external anal sphincter and puborectalis muscle was present (Fig. 3) which was excised along with 1cm of healthy margin and external anal sphincter was reconstructed. In the Post operative period patient was kept nil by mouth for 24 hours, followed by liquid diet and soft diet with laxatives. Adequate antibiotic cover and perineal care were given. She was relieved of her symptoms. Histopathology confirmed the diagnosis of scar endometriosis as it showed endometrial glands, stroma and hemosiderin laden macrophages (Fig. 3). She was followed up for six months and had total relief from her symptoms. There were no signs of recurrence of the disease.

Fig. 3: Intra-operative 3 cm fibrotic mass involving external anal sphincter and puborectalis muscle

Fig. 4: Histopathology showed endometrial glands, stroma and hemosiderin laden macrophages (confirmed diagnosis of scar endometriosis)

Discussion
Endometriosis is a clinical and pathological entity which is characterised by presence of tissue resembling functioning endometrial glands and stroma outside the uterine cavity. The quoted Incidence of Episiotomy scar endometriosis is 0.06-0.07% while that of Malignant Transformation is 0.3-1% of cases.\(^{(4)}\) The involvement of anal sphincter is infrequent in endometriosis. There is no role of conservative management, hormonal therapy is ineffective, thus the sphincter involvement should be diagnosed prior to surgery so as to avoid injury to external anal sphincter which can be reconstructed, so treatment of choice is surgical excision with margin of safety 1cm to prevent local recurrence.\(^{(5)}\)

Conclusion
Scal endometriosis with anal sphincter involvement is a rare type. Magnetic resonance imaging helps in diagnosing the extent of disease. The main modality of management is surgical excision with reconstruction of external anal sphincter.

References