The Journal of Obstetrics and Gynecology of India (July-August 2020) 70(4):314–316
https://doi.org/10.1007/s13224-020-01315-7

CASE REPORT

Inflammatory Myofibroblastic Tumour at Episiotomy Site: A Rare Case Report with Review of Literature

Meenal Bhati[1] · Meenakshi Gothwal[2] · Pratibha Singh[3] · Garima Yadav[2]

Received: 29 November 2019 / Accepted: 13 April 2020 / Published online: 27 May 2020

Introduction

An inflammatory myofibroblastic tumour (IMT) was considered under the broad category of inflammatory pseudotumour, but the designation of IMT was adopted by the 2013 World Health Organization (WHO) Classification of Tumours of Soft Tissue and Bone and classified it as the tumour of intermediate biological potential [1]. It is most commonly seen in children and young adults. Common sites of involvement are the lungs but it can arise from the mesentery, omentum, retroperitoneum, pelvis, and abdominal soft tissues. It rarely involves female genital tract, and among its most common sites is uterus [2]. Anaplastic lymphoma kinase (ALK) gene rearrangement is noted in 50–60% of the cases, and it can be identified by ALK immunohistochemistry. IMT has the recurrence rate of 25%, and rarely, metastasis can be seen in 2% of the cases [1]. However, female genital tract IMTs are rare; to the best of our knowledge, there are no reported cases of IMT involving episiotomy site.

Case Presentation

A 27-year-old female, Para one live one, presented to our OPD with complaints of a mass protruding from the episiotomy scar site and spotting from the growth site from past 10 days. The patient had undergone term vaginal delivery with right mediolateral episiotomy 3 months back. There was no history of any antepartum, intrapartum, or postpartum complication. She had lactational amenorrhea, and her previous menstrual cycles were regular. Her past medical and surgical history was unremarkable. On physical examination, built and nourishment was average and her vitals were stable. On examination, the abdomen was soft, no organomegaly. On local examination, a mass of 4 × 5 cm size protruding from introitus was seen, arising from previous episiotomy site that bleeds on touch, non-tender, and no abnormal discharge seen. Her routine investigations were within normal limit. Excision of granulation tissue was done under local anaesthesia. The histopathology report was suggestive of inflamed polypoidal spindle cell neoplasm. After 1 month, she reported back with a complaint of a recurrent mass at the same site of size 2 cm × 1 cm that bleeds on touch and non-tender. No significant abnormality was detected on contrast-enhanced computed tomography of the chest and abdomen from thoracic inlet to pubic symphysis. Magnetic resonance imaging of pelvis was done that was suggestive of mildly enhancing soft tissue lesion involving the right lateral wall of lower 1/3 of the vagina. The lesion was partially bulging into the vaginal canal, well separated from levator ani and puborectalis muscles (Fig. 1 a, b, c, d). Size of the lesion is 15 × 11 mm. Superior margin of the lesion is just below the level of the perineal membrane. Immunohistochemistry test was run on the previously operated tissue blocks, which suggested that the tumour is immunoreactive for smooth muscle actin (SMA), anaplastic lymphoma kinase (ALK) and is focally immunoreactive for BCL2 and desmin, negative for CK, CD 34, S 100 protein, suggestive of an inflammatory myofibroblastic tumour. Wide excision of the tumour was planned under general anaesthesia. The intraoperative finding was suggestive of 1 cm × 2 cm polypoidal growth over the anterolateral vaginal wall on the right side that was bleeding on touch (Fig. 2a–c). During surgery, at least 1 cm margin of normal tissue removed along with the tumour, multiple bleeding vessels present on

✉ Meenal Bhati
drmeenalbhati@gmail.com

[1] Department of Obstetrics & Gynaecology, All India Institute of Medical Sciences, Jodhpur, Rajasthan, India

[2] Associate Professor, Department of Obstetrics & Gynaecology, All India Institute of Medical Sciences, Jodhpur, Rajasthan, India

[3] Professor & HOD, Department of Obstetrics & Gynaecology, All India Institute of Medical Sciences, Jodhpur, Rajasthan, India

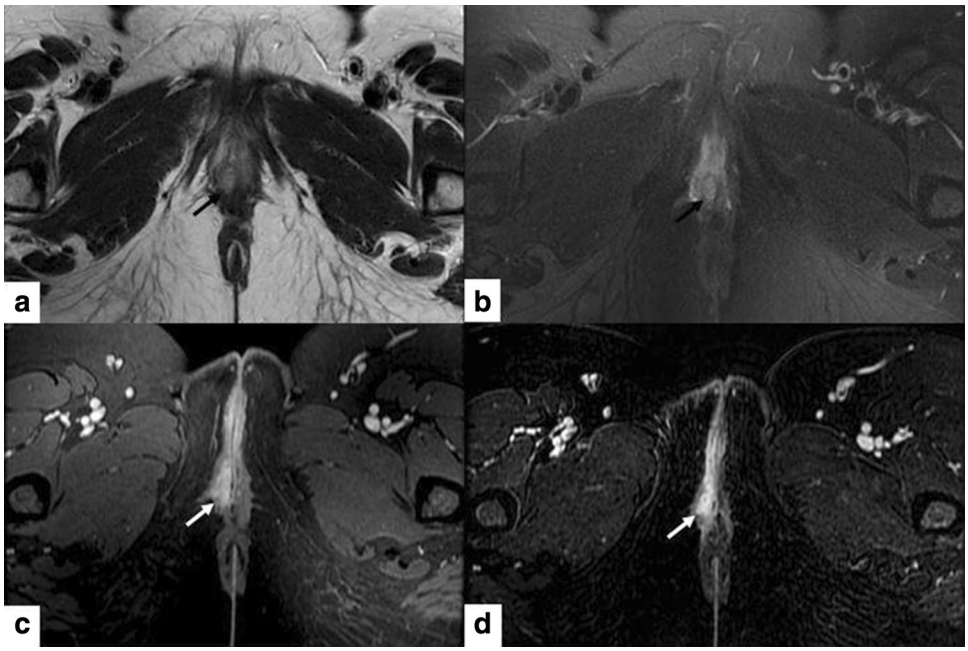

Fig. 1 MRI axial T1W (**a**) and T1W fat suppressed (**b**) images showing small hyperintense soft tissue lesion (black arrow) in the right lateral wall of lower 1/3 of vagina, bulging into vaginal canal. Post-gadolinium-enhanced T1W (**c**) and (**d**) axial images showing mild homogenous enhancement of the lesion with different parameters (white arrow)

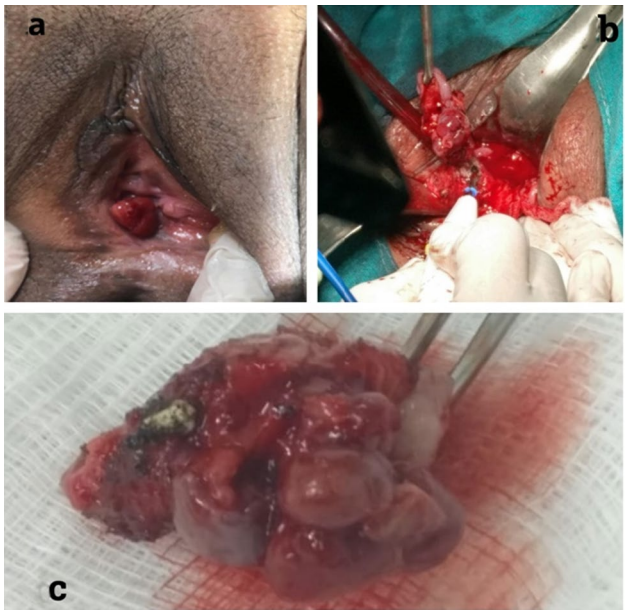

Fig. 2 **a** Pre-operative image of tumour arising from episiotomy site. **b** Intraoperative image showing resection of tumour. **c** Resected tumour specimen

tumour bed indicating feeding vessels, mild fibrotic planes were encountered during dissection, and haemostasis was achieved. The histopathology report of the excised specimen was suggestive of an inflammatory myofibroblastic tumour. Immunohistochemistry was done. The tumour was immunoreactive for smooth muscle actin (SMA), focally immunoreactive for BCL2, desmin and negative for CK, CD 34, S 100 protein. Based on the above finding, the final diagnosis of the inflammatory myofibroblastic tumour was made. Resection margins and the base were uninvolved by the tumour. The patient was kept for follow-up. At the most recent follow-up (7 months after surgery), the patient was symptom-free and there was no evidence of tumour recurrence on imaging.

Discussion

The inflammatory myofibroblastic tumour is the rare benign mesenchymal tumour with intermediate biological potential. The commonest site of involvement is lungs, liver, and orbit. They are rarely seen in the female genital tract, and among them, it has been reported in the uterus, cervix, ovaries, fallopian tubes, broad ligament, pelvic cavity, and placenta [4]. No case till date has been reported on IMT arising from the episiotomy scar site in the vagina. The aetiology of this tumour is still unclear. Infection, abnormal immunological reactions, or trauma can be one of the pathogenic factors for IMT [3, 4]. Rearrangements of the ALK gene are seen in almost 50% of cases [1]. Organizing mucoceles, myoepitheliomas, foreign body reactions, and reactions to certain infectious agents, nodular fasciitis, amyloidosis, myofibromas, solitary fibrous tumour, low-grade myofibroblastic sarcomas, and malignant sarcoma are some of the differential diagnosis for IMT [4]. As in our case, the 27-year-old female has a painless mass originating from the site of episiotomy and presented with spotting per vaginum. The first histopathological report was suggestive of inflamed polypoidal

spindle cell neoplasm, but after the recurrence wide excision of the mass was done and histopathology and immunohistochemistry reports were suggestive of IMT. It is quite common for patients with IMT to undergo multiple biopsy procedures to establish a diagnosis. On biopsy evaluation, IMT appears to be a diagnosis of exclusion. IMT can be firm, fleshy, or gelatinous, with a white or tan cut surface on gross examination and rarely calcification, haemorrhage and necrosis. Microscopic findings are suggestive of spindle cell proliferation in a myxoid to collagenous stroma with a prominent inflammatory infiltrate of plasma cells and lymphocytes, with occasional eosinophils and neutrophils. Clinical and histological features of IMT can mimic like ``inflammatory fibrosarcoma''. IMT can have various histological forms depending upon the dominant pattern: myxoid/vascular, compact spindle cell, or fibromatosis-like [4]. It is positive for vimentin (99%), SMA (92%), muscle-specific actin (89%), desmin (69%), and anaplastic lymphoma kinase (ALK) and negative for human melanoma black (HMB45), neurofilament (NF), CD68, CD34, and CD30, pan-keratin AE1/AE3, cluster of differentiation (CD) 117 (C-kit), S100-protein [3]. Not all IMTs are ALK-positive, as they are positive only in 50% of the cases [4]. Surgery is the primary treatment of choice, and favourable outcomes are seen after radical excision of IMT. Chemotherapy and radiotherapy are not having very promising results, but previous studies have shown good results of anti-TNF alpha-binding antibody (infliximab) infusion treatment and anti-inflammatory agents [3]. Long-term follow-up is mandatory in IMT.

Conclusion

The IMT is low-grade benign that tends to mimic malignancy on account of its aggressive clinical and radiographic features. Histopathology immunohistochemical analysis can help in making the diagnosis. To the best of our knowledge, we present the first case of IMT presenting at the episiotomy site in the vagina. This case report is unique because of the rarest site of presentation of the inflammatory myofibroblastic tumour. MRI and CT imaging with contrast assist in characterizing the lesion for surgical planning. The complete resection of the tumour is the treatment of choice. Fortunately, the tumour generally follows a benign clinical course; however, surveillance for recurrence and metastasis must be done. In our case, a follow-up of 7 months was done and there was no evidence of recurrence.

Funding NIL.

Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

References

1. Coffin CM, Fletcher JA. Inflammatory myofibroblastic tumour. In: Fletcher CDM, Bridge JA, Hogendoorn PCW, Mertens F, editors. World Health Organization classification of tumours of soft tissue and bone. 4th ed. Lyon: International Agency for Research on Cancer; 2013. p. 83–4.
2. Sinha R, Smita SK, Jamal I, et al. Inflammatory myofibroblastic tumour of ovary simulating malignancy with review of literature. Pac Group e-J. 2017. https://doi.org/10.21276/apalm.1285.
3. Abrantes C, Silva M, Oliveira R, et al. Inflammatory myofibroblastic tumour arising incidentally as a polypoid lesion in the gallbladder. J Bras Patol Med Lab. 2015;51(6):422–6.
4. Caporalini C, Moscardi S, Tamburini A, et al. Inflammatory myofibroblastic tumor of the tongue. Report of a pediatric case and review of the literature. Fetal Pediatr Pathol. 2018;37(2):117–25.

About the Author

Dr. Meenal Bhati is Third Year Post Graduate, in the Department of Obstetrics & Gynaecology, All India Institute of Medical Sciences, Jodhpur, Rajasthan, India. I am actively participating in the institutional co-curricular and extra co-curricular activities. I had attended various conferences and had presented many posters, paper and had also participated in various quiz at both international level conference and at state level conference. I have keen interest in laparoscopic surgeries and in gyneacologic oncology.