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Incidental Finding of Angiomyxoma Cervix in Uterovaginal Prolapse: A Case Report

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Abstract **Case Report**

Angiomyxoma is a rare, slow-growing mesenchymal tumor that usually arises in the pelvis and perineal regions of women in reproductive age, with a marked tendency of local recurrence. Most of the times it presents as asymptomatic perineal mass. The tumor is composed of scattered spindle cells and abundant medium-sized vessels embedded in a myxoid matrix. Surgical resection is the main treatment modality of angiomyxoma. Here, we are describing a rare case of angiomyxoma cervix, in a 35-year-old woman in which the diagnosis was made after histological examination. She presented with uterovaginal prolapse with infravaginal elongation of cervix and exceesive bleeding during surgery. In such cases follow up is important as cases of metastasis have been reported in literature.

Keywords: Angiomyxoma, mesenchymal, recurrence.

INTRODUCTION

Angiomyxoma is a rare locally invasive soft tissue tumour arising from the connective tissue of lower pelvis or perineum. It usually presents as an asymptomatic mass in the perineum. If tumour is large it can present with regional pain, dyspareunia, or a pressure-like sensation. (Steeper TA *et al*., 1983). It was first reported by Steeper and Rosai in 1983. Angiomyxoma is thought to be estrogen progesterone sensitive which is supported by the fact that it occurs usually in the reproductive age group with peak incidence during 3rd decade of life (Chan YM *et al*., 2000). In women most common site of involvement is vulva (Abdul Karim *et al*., 2001). Here, we are describing a case of angiomyxoma cervix associated with uterovaginal prolapse and infravaginal elongation of cervix.

CASE REPORT

A 35year old woman, P3L3 presented to the outpatient clinic with complaints of something coming out of introitus since 2 years. She used to reduce it by herself. It was associated with backache since 2-3 months. Menstrual cycles were normal and regular lasting for 4-5 days and occurring at interval of 28-30 days but associated with dysmenorrhoea. She had normal deliveries and tubectomy was done 10 years back. General physical examination was unremarkable. Local examination revealed third degree UV prolapse with infravaginal elongation of cervix. Pelvic examination revealed bulky, mobile uterus with clear adnexa. Her blood group was AB negative. USG showed bulky uterus suggestive of adenomyosis. Endometrial thickness was 7.4mm. Bilateral ovaries were normal. Patient was posted for vaginal hysterectomy after covid-19 RTPCR test. During surgery she had excessive bleeding and blood was transfused intraoperatively. Postoperative haemoglobin was 7.8g/dl and one more blood was transfused. Patient was discharged in a satisfactory condition on 5th postoperative day. Sample was sent to pathology department.

Gross pathology: Grossly uterus with cervix measured 15x8x6cm. Cervix was elongated and measured 6.5x5x4.5 cm. On cut section cervix was tan yellow, soft with gelatinous areas as shown in Figure 1& 2. Endometrial canal was patent and endomyometrium measured 2.8cm in maximal thickness. Vaginal flap measured 4x2x0.8 cm.

Histopathology of the sample showed chronic cervicitis with angiomyxoma of cervix. Endometrium showed chronic endometritis.

Microscopic examination of cervix revealed presence of benign, hypocellular tumour composed of spindled stromal cells with a prominent vascular component set within copius myxoid matrix. The vessels were medium sized to large, with few of them showing showing thick hyalinized walls, and were evenly distributed throughout the tumour. Lesional spindle cells had delicate eosinophilic bipolar cell processes, and the nuclei were bland and ovoid or rounded.Also collections of eosinophilic smooth muscle cells (myoid bundles) were seen. As shown in Figure 3 & 4.

Fig 1&2: Cut section of uterus and cervix: Shows elongated cervix which on cut section is tan yellow, soft with gelatinous areas

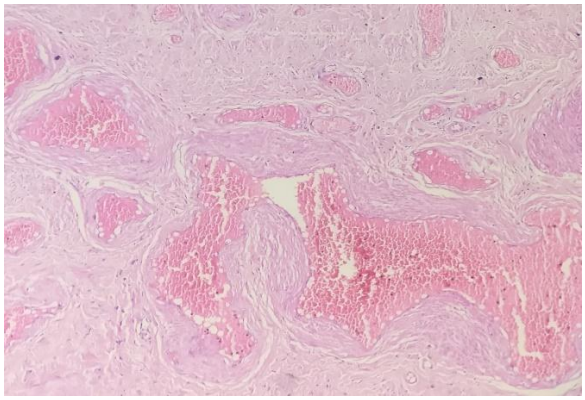

Fig 3: Haematoxylin and eosin stained section (low power): medium size to large blood vessels, few of them showing thick hyalinized walls evenly distributed throughout the tumour

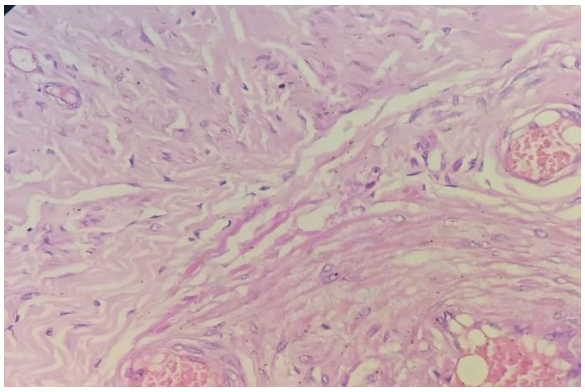

Fig 4: H & E(High power): Lesional spindle cells having delicate eosinophilic bipolar cell processes & bland ovoid nuclei

DISCUSSION

Angiomyxoma is an uncommon mesenchymal tumor which is mostly derived from the pelvic and perineal regions including vulva, vagina, bladder, and rectum. However, uncommon localizations such as lung, liver, larynx, and orbit have been reported (I. Dierickx *et al*., 2008). We have found angiomyxoma in cervix which is also an uncommon site. Patients are often asymptomatic at the time of diagnosis, with perineal or vulvar masses discovered incidentally during physical examination or radiologic imaging. Angiomyxoma presents in the reproductive age group with few cases reported during pregnancy also. While reviewing the literature, it was found that angiomyxoma has presented with uterine and cervical masses in most of the cases.

Grossly these tumours are homogenous in consistency with a glistening gelatinous appearance on cut section (Steeper TA *et al*., 1983). Microscopy shows a hypocellular mesenchymal lesion with scattered bland spindled and stellate cells in a myxoid stroma. The tumour lacks cytologic atypia, increased/ atypical mitoses and coagulative necrosis. Numerous blood vessels of varying calibre is the characteristic feature of the tumour. Same findings have been reported on histopathological examination of our sample. The tumour is usually positive for vimentin, desmin and negative for S-100 on imomunohistochemistry (Zhang P *et al*., 2011).

Angiomyxoma usually presents as a slow growing mass due to which it may be mistaken for a benign growth. It should be differentiated from angiomyofibroblastoma (AMF), myxoid leiomyoma,

cellular angiofibroma, superficial myofibroblastoma, myxoid liposarcoma, as they differ in the management (Layfield LJ *et al*., 1997). The diagnosis of angiomyxoma may be difficult to establish. The distinctively striking vascular component in angiomyxoma helps to rule out the others. Angiomyofibroblastoma occurs later in life (3rd to 9th decades) and involves vulva and vagina. It presents as a submucous mass and shows high cellularity, large number of blood vessels and plump stromal cells with perivascular accentuation on histopathology. They do not recur. Cellular angiofibroma/spindle cell lipomas are small in size and are mixed with fat with mural hyalinization of blood vessels.

Angiomyxoma is thought to displace rather than invade surrounding tissue. In the usual case presenting as a perineal mass, careful anatomic dissection should be done to avoid injury to anal sphincters and lower urinary system (Chan YM *et al*., 2000). In case of a cervical or uterine polyp, complete removal of the polyp and pedicle should be done to ensure disease free margins.

Complete surgical excision is the gold standard, because of its tendency to recur locally. The recurrence rate varies from 36-70%. There is no correlation between tumour size and recurrence rate (Chan YM *et al*., 2000). Incomplete or partial resection is acceptable when high operative morbidity is anticipated and fertility is an issue. Treatment with GnRH agonists and tamoxifen can be tried. These agents may help in reducing the extent of surgery by reducing the size of the tumour (Haldar K *et al*., 2010). Though radiotherapy and chemotherapy may seem to be less useful due to the low mitotic activity and low cellularity of the tumour, various reports have documented reduced recurrence with their use (Rhomberg W *et al*., 2000). The presence of multiple feeding vessels also limits the use of embolization (Layfield LJ *et al*., 1997). Individualization of the treatment options and multimodal treatment seem to be the most appropriate (Coppola S *et al*., 2013).

It is typically a benign, nonmetastatizing neoplasm. However, multiple metastasis to lungs have been reported (Bai HM *et al*., 2013). Hence, long-term follow-up of patient is necessary. Our patient is followed after surgery. There is no evidence of recurrence till date.

CONCLUSION

Despite its rarity, Angiomyxoma should be considered in the differential diagnosis of any painless swelling located in the pelvic and perineal region, particularly in women of reproductive age. Histopathology reveals pathology in asymptomatic cases which otherwise is masked. The principle treatment should be complete surgical excision with tumor-free margins. Long-term follow-up and careful monitoring are essential due to its high local recurrence rate in spite of wide excision of the tumor.

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