Case report

Aggressive angiomyxoma mimicking cervical polyp

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Abstract: PURPOSE: The purpose of this paper is the presentation of a case of an aggressive angiomyxoma of the uterine cervix in a 29–year-old woman. METHOD: The patient was presented with dysuria, and mild suprapubic pain. The preoperative diagnosis after the physical examination was uterine cervical polyp. RESULTS: Histological examination performed after surgical excision however, showed a densely vascular, poorly circumscribed neoplasm, composed of spindle-shaped cells widely spaced from each other in myxoid stroma. These findings were compatible with the diagnosis of an aggressive angiomyxoma. CONCLUSION: Aggressive angiomyxoma is a rare soft tissue tumor of the pelvis and perineum. Pathologic and clinical characteristics of the tumor are discussed.

Keywords: Dysuria; Aggressive angiomyxoma; Cervical polyp.

INTRODUCTION

Aggressive angiomyxoma is a mesenchymal tumor initially first described in 1983.[1] It typically appears as a soft tissue mass of the pelvis and perineum in women of reproductive age.[2] Its differential diagnosis includes myxoma, myxoid liposarcoma, sarcoma botryoides, and other soft tissue tumors with secondary myxoid changes.[1] We present a rare case of aggressive angiomyxoma of the uterine cervix, clinically simulating a pedunculated cervical polyp.

CASE REPORT

The patient, 29 years old, presented to the outpatient department for a routine health care visit. She was nulliparous, reported to be sexually active. She complained of mild suprapubic pain and dysuria. Upon the speculum examination, the presence of a large, polypoid, pedunculated mass originating from the uterine cervix was noticed. The presumptive diagnosis was cervical polyp and the treatment recommended was elective surgical excision.

The tumor was removed with an electrocautery blade electrode under general anesthesia. It originated from the external cervical os and its base of about 1cm in diameter was fulgurated. The patient was discharged the next day.

The surgical specimen measured 5 x 4 x 2 cm. It was glistening white, soft and solid (Fig. 1). Histologically, the lesion was poorly circumscribed, partially covered by mature squamous epithelium (Fig. 2) and contained rare endocervical glands. The intermediate stroma composed of mesenchymal spindle-shaped cells widely separated by myxoid stroma which also contained many small- and medium-sized thick-walled vessels. Rare bundles of smooth muscle cells and few mast cells were also identified Immunohistochemical examination showed positive reaction of the spindle cells to desmin and smooth muscle actin (SMA), while they were negative to S-100 protein and CD34 antigen. The latest revealed the rich vascular network of the neoplasm. Proliferation index antigen Ki67 was practically negative. These findings were consistent with aggressive angiomyxoma.

Two years after surgery the patient remains in good general condition with no signs of relapse.

DISCUSSION

The term dysuria refers to a condition during which a patient has difficulty in voiding. It is described as painful or uncomfortable urination and it is an extremely common symptom regarding urinary tract pathological cases. The most common cause of dysuria in women is urinary tract infections. It can be observed in any age and in both sexes. Urinary tract infections are one of the most common infections in women. However when a female patient is presented with dysuria a detailed medical and lifestyle history is obligatory. Bicycle riding, horse riding, depression or use of specific drugs such as anticholinergics for Parkinson's disease can be causes of dysuria. Physical examination including speculation of the vagina and the urethra may reveal causes of dysuria that are not related to urinary tract infections such as urethral

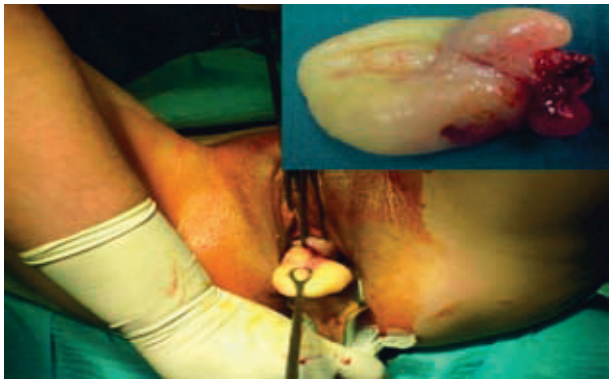

Fig. 1 - Glistening white, polypoid cervical mass, of soft and solid texture.

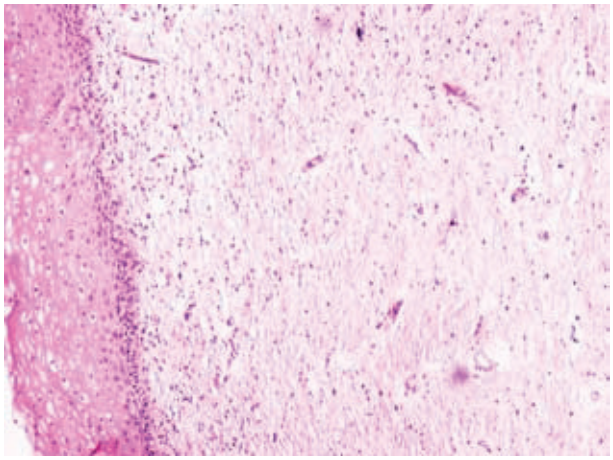

Fig. 2 - Poorly circumscribed bulky mass, partially covered by cervical epithelium. Histochemical stain H&E, X200.

damage during sexual intercourse, vaginitis, vulvovaginitis or tumors.

Aggressive angiomyxoma is an uncommon, slow growing mesenchymal tumor that usually affects the pelvis and perineum of adult women. It may be clinically misdiagnosed as an inguinal hernia or a Bartholin cyst, while polypoid configuration has not so far been reported.

Its pathogenesis is not completely understood. Recent cytogenetic studies revealed chromosomal translocations in specimens of aggressive angiomyxoma. Theese include HMGA2 protein rearrangement at chromosome 12,[3-4] and a t(5;8)(p15;q22) translocation.[5]

The tumor is poorly circumscribed; it consists of spindle and stellate cells separated by myxoid stroma, focally rich in collagen fibrils and prominent vessels. Vascular spaces vary in size and include large thick-walled vessels without an arborizing pattern. Stromal fibrils and spindle cells immunohistochemically exhibit myofibroblastic differentiation (smooth muscle and fibrous tissue),[1-6] a fact that histologically differentiates the mass from common cervical polyps. Mitotic activity is usually exceedingly low.

The term "aggressive angiomyxoma" was designated to this neoplasm to emphasize the neoplastic nature of the blood vessels and its tendency to be locally aggressive and recur after treatment. Recurrence rates up to 70 % have been reported, thus long term follow-up of the patient is necessary.[1-7]

It is typically a benign, non-metastasizing neoplasm, in two cases however[8-9] multiple metastases have been reported, including pulmonary involvement, and both women died.

Surgical excision with free margins is the treatment of choice in most of the times. In several cases, removal of the tumor may be difficult due to local infiltration and adjuvant therapy may be used. Treatment options include use of hormonal manipulation, such as tamoxifen, raloxifen or GnRH analogs, radiotherapy and arterial embolisation.[10-11-12] Nevertheless, their role in the definite treatment or treatment of recurrences of aggressive angiomyxoma needs further investigation.

CONCLUSION

Although symptoms as dysuria or suprapubic pain refer to common situations as urinary tract infections each patient which is presented with these symptoms should be carefully evaluated. Medical history and physical examination can help in the diagnosis of rare conditions that may require special treatment or any invasive therapy. Usually dysuria is treated with the use of antibiotics. But in cases in which the administration of antibiotics is not helpful and the symptoms persist, the presence of situations other than urinary tract infections should be considered and the patient should be closely revaluated.

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