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Case Report

Management of challenges with large cervical masses

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ABSTRACT

Cervical adenomyomas are extremely rare benign tumors and are often overlooked as a differential diagnosis of large cervical masses. Herein, we report a case of a 23-year-old nulliparous woman with polypoid adenomyoma arising from the endocervix. The mass was filling up the vagina and stretching the anterior fornix up into the lower abdomen, hence the origin of the mass could not be identified on clinical examination or imaging. Therefore, management of this mass was challenging. Excision of this mass was done using an abdominoperineal approach. Histologically, the polypoid lesion was composed of endocervical epithelium with interlacing fascicles and bundles of tumor cells intermixed with endocervical glands. Clinicians should be aware of such lesions so that they can decide upon the appropriate course of management.

Keywords: Benign tumors, Cervical adenomyomas, Large cervical masses

INTRODUCTION

Adenomyomas are benign biphasic tumors mostly arising from the uterus and less commonly from the cervix and hence are often overlooked due to their rare presentation. These are well-circumscribed tumors with an endocervical glandular component and a mesenchymal myometrial component. They are detected mostly incidentally in women of reproductive or post-menopausal age (mean age 40 years). They may present as a mass or a polyp protruding through the external os associated with complaints of abnormal bleeding or discharge per vaginum. The biological behaviour of adenomyomas is benign with an excellent prognosis if completely removed.

However, when the tumor is large and has a broad base, it may be difficult to ascertain its origin and therefore complete resection can become a challenging proposition. A case of large polypoid adenomyoma of endocervical type is described here.^{1,2}

CASE REPORT

A 23-year-old nulliparous woman presented with complaints of foul-smelling discharge for 6 months, dull aching pain in the lower abdomen for 2 months, and a gradual increase in distension of the lower abdomen over one month. Upon admission, the per abdomen examination revealed a regular mass arising from within the pelvis of approximately 14-16 weeks in size. On retracting the labia minora, a huge mass was seen filling the vagina just 2 cm short of the introitus with an infected and necrosed surface that bled on touch. The same mass precluded adequate per vaginal examination. Therefore, the origin of the mass from the uterus or cervix could not be ascertained. On perrectal examination, the mucosa was free, and the upper end of the mass was not appreciable. The patient underwent an abdominal ultrasound in which the uterus gave a "lantern on St Paul's dome" appearance over a mass of 14×15 cm size. Magnetic resonance imaging (MRI) confirmed the presence of this large mass distending the cervical canal and the vagina with a stalk of 17 mm at the uterocervical

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junction. The bladder was advanced and pushed anteriorly and the rectum and sigmoid were displaced posteriorly.

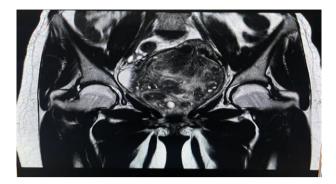


Figure 1: The coronal section of the MRI film shows a large mass that is heterogeneous with hyperintense to isointense areas in between. The mass is distending the vagina.

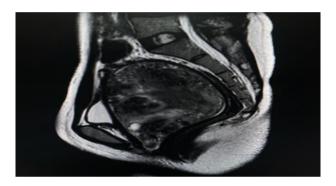


Figure 2: MRI film showing the sagittal section in which the upper end of the mass is reaching the sacral promontory.



Figure 3: Macroscopically, the mass was approximately 15×14×12 cm with pockets of degeneration.

After ensuring fitness for general anaesthesia, the patient was planned for excision of the mass. Considering the nulliparous status of the patient and the difficult surgical approach, an abdominoperineal approach was planned. Pre-operatively cystoscopy with bilateral ureteric stenting was done to avoid any injury to the ureter in case of excessive bleeding during dissection. The abdomen was opened with a small infra-umbilical incision. A small

unicornuate uterus was seen sitting atop a large mass that was bulging from the pelvis into the abdomen. The bladder was seen to be pushed up and stretched over the mass. The ureterovesical fold was opened and the bladder was pushed down to expose the anterior vaginal fornix in preparation for intervention in case of massive bleeding during vaginal dissection of the mass. With this preparation, we began excising the mass through a vaginal approach by a coring technique using cutting and coagulating current through a monopolar cautery. As we reached the base of the stalk, there was no identifiable pedicle and the mass appeared to be arising from deep within the cervical canal. The anterior lip of the cervix was identifiable from the 12 o'clock position to the 3 o'clock position. Since the patient was nulliparous, the mass was excised at the level of external os leaving behind a slightly thick cervix. The base was secured with haemostatic sutures. Complete haemostasis was achieved. Vagina was checked for any trauma. The per-urethral catheter was draining clear urine. The abdomen was closed in layers after closing the uterovesical fold. Total blood loss was around 400 millilitres. Ureteric stents were removed. The patient tolerated the procedure well.

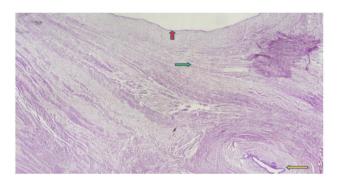


Figure 4: On microscopy: the red arrow represents the endocervical epithelium which is mucus-secreting columnar epithelium, yellow arrow represents endocervical glands surrounded by stroma rich in smooth muscle that is represented by the green arrow.

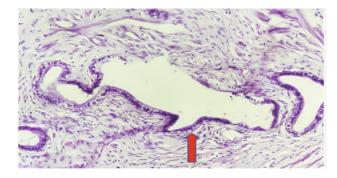


Figure 5: On microscopy: magnified view showing endocervical glands.

Histopathological examination of the specimen showed that the tumor was lined by endocervical endothelium composed of interlacing fascicles and bundles of oval to spindle-shaped tumor cells with intermixed endocervical glands. Histochemically, intracytoplasmic mucin of the glands was positive for PAS-positive neutral mucin. The endocervical epithelium was nuclear estrogen receptor (ER) positive, and nuclear progesterone receptor (PR) was non-contributory.

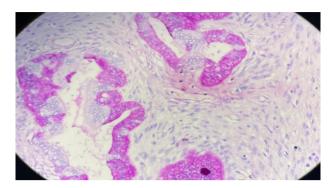


Figure 6: On immunohistochemistry, the mass showed PAS positivity.

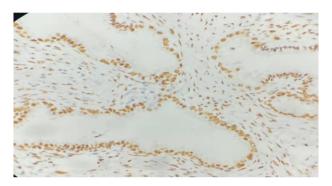


Figure 7: On immunohistochemistry, the mass showed estrogen nuclear receptor positivity.

DISCUSSION

Adenomyomas are rare benign tumors of the female genital tract that are composed of smooth muscle cells and endometrial glands separated from the normal myometrium by a pseudo capsule. The most common site is the uterine corpus and very rarely they may arise from the cervix. The cervical lesions are mostly polypoid. The patient may be asymptomatic or present with abnormal uterine bleeding or copious discharge.³ Adenomyomas are to be differentiated from a variety of benign lesions including lobular endocervical glandular hyperplasia, adenofibroma, endocervicosis, tunnel clusters, and malignant lesions like adenomyoma malignum.4 Microscopically, endocervical adenomyoma characterized by glands and cysts lined by a single layer of endocervical-type mucinous epithelium intermingled with smooth muscle.

The present case gains significance in the light of being a diagnostic dilemma in terms of the nature as well as the origin of the mass. The significant features of this case

included the young age and nulliparous status of the women, the large size of the tumor which on histopathology was shown to be a cervical adenomyoma, and the surgical challenge that it presented. Results of immunohistochemistry should be combined with gross and microscopic features, otherwise, this rare condition can be confused with adenoma malignum.

To the best of our knowledge, the abdominoperineal approach used for the management of this large mass has not been previously described in the literature. However, we deemed it best to use this approach and be prepared for any intervention in case of excessive haemorrhage. Ureteric stenting was done to avoid any inadvertent injuries while controlling anticipated bleeding and avoid the possibility of a hysterectomy. In retrospect, we believe that being prepared for a double setup procedure, abdominal as well as vaginal with two surgical teams could be another option. Thus, we could avoid opening the abdomen until such a need arose in case of excessive haemorrhage during vaginal excision.

CONCLUSION

Anticipation and adequate preparation for surgery is the key to avoid complications. The abdominoperineal approach could be an option in case of large vaginal masses to reduce the time to control excessive bleeding during excision of the mass through the vagina. Histopathology is mandatory to differentiate from adenomyoma malignum and adenosarcomas specifically in tumors of such large size. Another significance of this histopathology is its possible association with subsequent adenomyosis and endometriosis in the patient.

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