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A Rare Incidental Case of Lipoleiomyoma Cervix

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ABSTRACT

Lipoleiomyomas are uncommon benign mesenchymal neoplasms of uterus and are regarded as a rare variant of uterine leiomyomas. They constitute less than 0.2% of benign uterine tumors. These tumors usually occur in postmenopausal women and are seen mainly in uterus as intramural growths but can rarely arise in cervix, broad ligament, reteroperitoneum and ovary. We present a case of cervical lipoleiomyoma in a premenopausal woman because of its rarity, interesting histiogenesis, deceptive characteristics with emphasis on role of imaging and histopathology with immunohistochemistry in diagnosis of these tumors.

Keywords: Lipoleiomyoma, Cervix, Histogenesis, Immunohistochemistry

Introduction

Uterine tumors of adipose tissue are very unusual benign soft tissue tumors which constitute 0.03 - 0.2% of uterine leiomyomas. These tumors are composed of variable proportion of mature adipose tissue and smooth muscle cells [1]. They are predominantly located at the uterine corpus and rarely occur in cervix with only few case reports in the literature [2,4,5]. We report a case of lipoleiomyoma of cervix in a premenopausal women which was clinically diagnosed as malignant tumor.

Case report

A 39 year old, premenopausal women presented with chief complaints of continuous vaginal bleeding, pain lower abdomen and difficulty in micturation for past 4 months. Her past medical and family history was insignificant. The general and systemic examinations along with her haematological parameters were within normal limits. Biochemical analysis showed mild increase in triglycerides (160mg/dl) and cholesterol levels (230mg/dl). Per vaginal examination revealed a firm mass in the posterior lip of cervix. Bilateral fornices were filled with a firm, non tender mass.

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Speculum examination showed a cervical mass protruding through the external os. Pelvis ultrasonogram (USG) revealed a normal uterus with endometrial thickness of 4mm. Multiple small hyperechoic lesions were seen in anterior myometrial wall possibly suggestive of intramural fibroids. Right ovary showed a cyst measuring 4x3cms with thin septations and left ovary was not visualised. Pouch of Douglas was clear. The cervix showed a well defined hyperechoic lesion measuring 3x3cms. On computer tomography (CT), the uterus was normal in size, attenuation and enhancement pattern. Multiple hypodense lesions were noted in the uterine body suggesting intramural fibroids and also there was a well defined hypodense lesion with a small enhancing solid component measuring 3x3cms in the posterior lip of cervix. Right ovary showed a well defined round lesion measuring 5x5cms with small focal areas of fat and calcification. Bilateral fallopian tubes were normal. Both, USG and CT findings were suggestive of multiple intramural uterine fibroids and cervical fibroid with right ovarian dermoid cyst. The patient underwent total abdominal hysterectomy and bilateral salpingo-oopherectomy. Grossly, the uterus, cervix along with attached bilateral adenexas measured 7x5x4cms. Cut surface of uterus revealed numerous intramural fibroids which were solid and gray -white with whorled appearance. Cervix showed a well circumscribed grey yellow mass measuring 3x3cms located at the posterior

lip. Right ovary measured 6x3x3cms and on cut revealed a cyst measuring 5x3x2cms which was filled with haemorrhagic material Left ovary and bilateral fallopian tubes were grossly unremarkable.

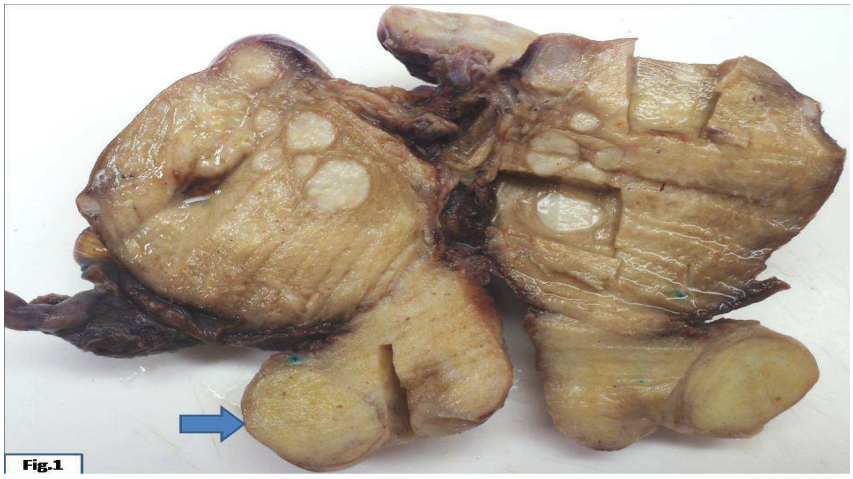

Fig. 1: Gross specimen showing a well circumscribed grey yellow tumor at cervix

On histopathological examination, sections from cervix showed a tumor comprising of mature adipose tissue along with bundles of smooth muscle fibers and fibrous connective tissue. Blood vessels in the stroma were not prominent and were thin walled. No atypia in smooth muscle cells or adipocytes was seen. No necrosis, mitosis, calcification was observed (Fig. 2A & 2B).

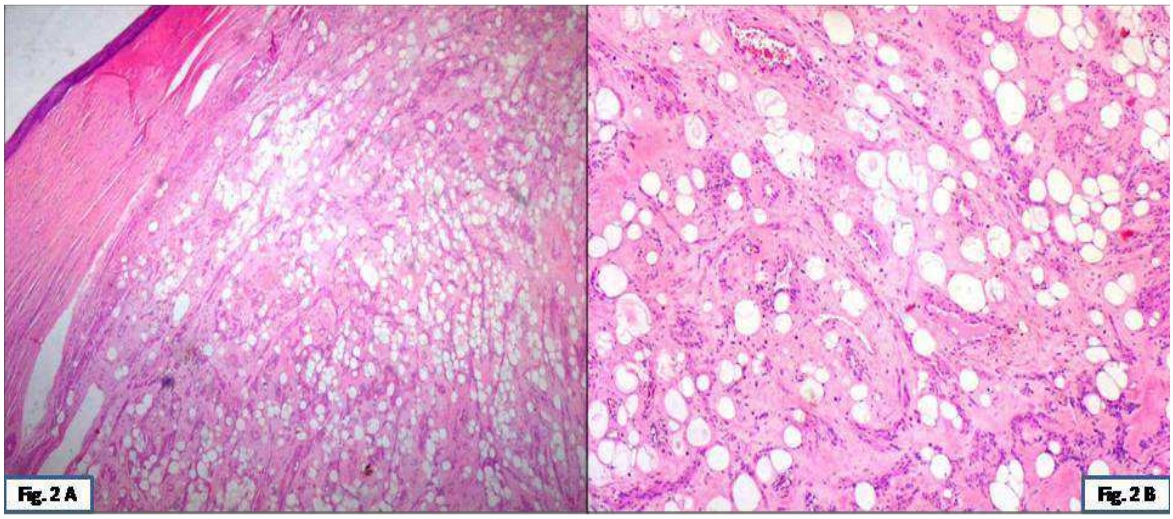

Fig. 2A - Photomicrograph showing stratified squamous epithelium of cervix with underlying tumor comprising of adipose tissue along with bundles of smooth muscle fibers (H & E, 10x)
Fig. 2B - H & E, 40x

Immunohistochemically (IHC), the smooth muscle fibers intervening the adipose tissue were positive for desmin, smooth muscle actin and vimentin whereas the adipocytes were positive for vimentin and negative for estrogen receptor (ER) and progesterone receptor (PR) (Fig. 3A,3B, 3C) , thus confirming the diagnosis of lipoleiomyoma . Sections from fibroids located in body of uterus showed features of typical leiomyoma.

Endometrium revealed endometrial glands in proliferative phase, right ovary showed features of endometriosis while left ovary and both the fallopian tubes were unremarkable.

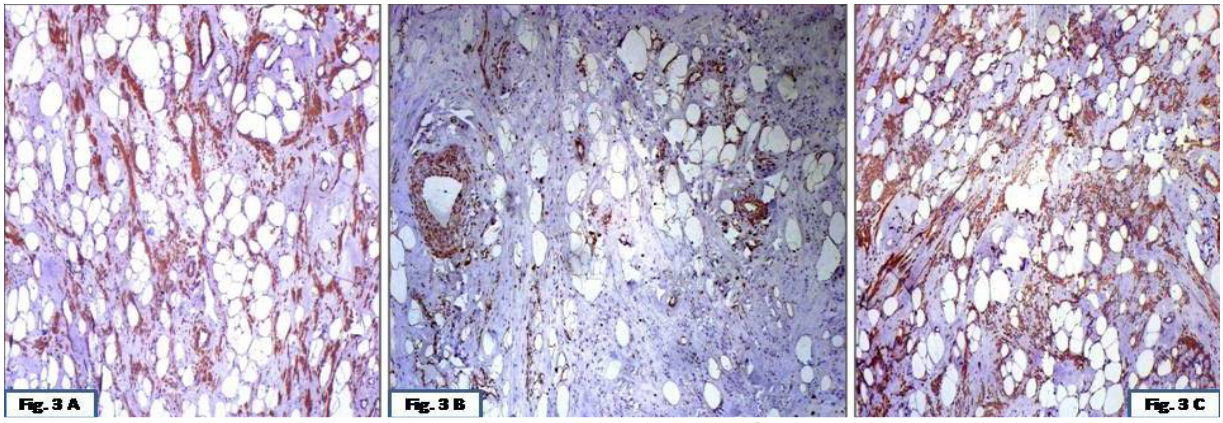

Fig.3A - Photomicrograph showing smooth muscle bundles positive for SMA (IHC, 10x)
Fig.3B - Adipocytes positive for vimentin ((IHC, 10x)
Fig.3C - Smooth muscle bundles positive for desmin (IHC, 10x)

Based upon the histological findings, the case was designated as benign lipoleiomyoma, cervix. The outcome was favourable. The patient was kept under follow up and till her last outpatient visit she was symptom free and had no complaints.

Discussion

Cervical leiomyoma is the most common cervical benign tumor arising in the cervical muscular tissue. It is usually small, typically measuring 0.5 to 1 cm, solitary, smooth, firm, similar to uterine myomas which in general, are multiple. Cervical fibroids constitute 1-2% of total fibroids and are of 3 types- interstitial, supravaginal and polypoidal, each presenting differently. Supravaginal fibroids can be central surrounding the entire cervical canal and can also displace the uterus superiorly. They can also be unilateral or bilateral, intramural or subserosal, and can be lying in the pelvis. When symptoms do occur, they are dependent on the direction in which the enlarging myoma expands. The expanding myoma produces symptoms secondary to mechanical pressure on adjacent organs causing dysuria, urgency, ureteral obstruction, dyspareunia, or obstruction of the cervix [2]. Occasionally a cervical myoma may become pedunculated and protrude through the external os of the cervix. These prolapsed myomas are often ulcerated and can masquerade as a malignant tumor. Spontaneous vaginal expulsion of the cervical fibroid have been described: the pathological analysis of the product must be systematic because it is necessary to eliminate the presence of malignancy, especially leiomyosarcoma [3].They can be also be asymptomatic and usually never present as any menstrual disorder.

The presence of fatty tissue in the myometrium is anomalous, interpreted as lipomatous degeneration, fatty metamorphosis, smooth muscle metaplasia or as a benign tumor called as lipoleiomyoma [1]. They occur in different locations predominantly being at uterine corpus and are rarely found in cervix as found in our case [2,4,5].

Its pathogenesis is controversial and immuno histochemical studies have helped in explaining its complex histiogenesis. The origin of the lipomatous lesions of the uterus have various proposed theories that include misplaced embryonic fat cells, metaplasia of muscle or connective tissue cells into fat cells, lipocytic differentiation of a specific primitive connective tissue cell, perivascular fat cells accompanying the blood vessels into the uterine wall during surgery, or fatty infiltration or degeneration of connective tissue [1]. But, because of the relative paucity of smooth muscle fibers in the cervical stroma, the majority of myomas that appear to be cervical actually arise from the isthmus of the uterus [3]. Our immunohistochemical study has revealed reactivity of adipocytes for vimentin confirming the hypothesis of their direct transformation from smooth muscle cells

into adipose cells. A number of various lipid metabolic disorders or other associated conditions with estrogen deficiency which occur in peri or post menopausal period, possibly promote abnormal intracellular storage of lipids which may play a role in development of lipoleiomyomas [7]. The current case is of a premenopausal female who had mild elevated serum triglycerides and cholesterol levels which might have led to its occurrence.

They are usually detected incidentally postoperatively as a chance pathological findings in most of the cases but for its diagnosis imaging can play an important role in determining its location and fatty nature .On USG, the lesion is echogenic and is usually partially encased by a hypoechoic rim. The hypoechic rim is thought to represent a layer of myometrium surrounding the fatty component. However, USG findings are not specific to the diagnosis [4,5]. CT and magnetic resonance imaging (MRI) findings are more specific and can assist in the preoperative diagnosis of these lesions as it helped in our case too. Histopathology is still the gold standard for its diagnosis and IHC plays an dynamic role as has been suggested by many authors [1,5,8]. In our case, the smooth muscle fibers intervening the adipose tissue were positive for desmin and smooth muscle actin. Some researchers have also interpretated that the adipose tissue element of lipoleiomyomas show cell proliferative activity and that it is positive for estrogen receptor and progesterone receptor , suggesting that the adipose element was not degenerative tissue but actively proliferative tissue. The presence of these receptors in the adipose element suggests that the adipose element was specific fat tissue related to female genital organs [9]. Though, in our case the ER and PR were negative and did not support there finding.

The differential diagnosis of fat containing mass in female pelvis is quite limited. The vast majority of these lesions are benign cystic ovarian teratoma [10]. Others include uterine fatty tumors, ovarian lipomas or possibly ovarian lipoleiomyoma. Other histological differentials of uterine tumors with similar morphology include; spindle cell lipoma, angiolipoma, angiomyolipoma, myelolipomas, atypical lipoma, and well-differentiated liposarcomas [1].

Treatment of lipoleiomyoma is similar to typical leiomyoma. Usually asymptomatic lipoleiomyoma requires no treatment. Management of these tumors is usually by hysterectomy as they pose surgical difficulty being close to urinary bladder and uterus. Uterine artery embolization and myomectomy can be performed depending on patient's symptoms, fertility desire, the site of the mass, and associated uterine fibroids [3,6]. These donot recur or cause any major fatality.

Conclusion

Cervical lipoleiomyomas are extraordinary rarer than uterine lipoleiomyomas with clinical symptoms similar to typical leiomyomas. They frequently occur in perimenopausal and postmenopausal women but can occur in premenopausal women. The final diagnosis is established on the basis of a histopathological examination with radiology playing important role in preoperative diagnosis. The IHC findings suggest a complex histogenesis for these tumors, in that they might arise from mesenchymal immature cells or from direct transformation of smooth muscle cells into adipocytes by means of progressive intracellular storage of lipids.

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