



RESEARCH ARTICLE

GIANT ENDOCERVICAL LIPOLEIOMYOMA OF CERVIX IN A YOUNG FEMALE, PRESENTING WITH PROLAPSE – AN UNUSUAL PRESENTATION

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ABSTRACT

Lipoleiomyoma as we know it, accounts for merely 0.03- 0.2 % of uterine leiomyomas. Predominantly described in the uterine corpus, there have been reports describing their occurrence at uncommon sites like ovary and broad ligament. Cervical leiomyomas however account for 1-2% of all the fibroids encountered clinically and lipoleiomyomas arising from the cervix are even rarer. Our case is unique since it is a giant lipoleiomyoma of cervix in a young patient who presented with prolapse. A 37 year old, female presented with continuous vaginal bleeding from past 3 months, abdominal pain since 1 month and a mass protruding out of vagina since last 1 week. On further investigations while ultrasound was suggestive of an adnexal mass, MRI suggested the presence of cervical leiomyoma. Total abdominal hysterectomy with unilateral salpingo-oophorectomy was performed on histopathology; a final diagnosis of cervical lipoleiomyoma was given. We report this case due to its rarity in terms of uncommon presentation, deceptive characteristics on radiology and its unusually large size and we also emphasize the role of imaging and histopathology with immunohistochemistry in diagnosis of this rare variant of leiomyoma of cervix .

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INTRODUCTION

Leiomyomas of the uterus are the most frequently encountered benign smooth muscle neoplasms of female genital tract. However lipomatous differentiation in these tumors is quite uncommon. Lipoleiomyoma as we know it, accounts for merely .03- 0.2 % of uterine leiomyomas. (Sonam Sharma et al., 2015) Although most commonly described in the uterine corpus, they have occasionally been reported in various other locations including cervix, ovary, broad ligament and retroperitoneum as well. (Sonam Sharma et al., 2015; Manjunatha et al., 2010) When in the cervix, these tumors are usually small, ranging in size from 0.5 to 1 cm. Moreover the lipomatous differentiation is seen to occur most commonly in post menopausal obese women. Our case is unique since it is a giant lipoleiomyoma of cervix in a young patient who presented with prolapse. Due to its rarity in terms of uncommon presentation, deceptive characteristics on radiology and its unusually large size, we report this case. We also emphasize the role of imaging and histopathology with immunohistochemistry in diagnosis of this rare variant of leiomyoma of cervix.

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Case report

A 37 year old, Gravida 2, Para 2, Live 1 and Abortion 1 (G2P2L2A1 ), premenopausal female presented to the Gynecology Out Patient Department with chief complaints of continuous vaginal bleeding from past 3 months, abdominal pain since 1 month and a mass protruding out of vagina since last 1 week. She is a known case of Type II diabetes mellitus. Her general and systemic examinations along with the hematological parameters were within normal limits. Per vaginal examination revealed a large mass of about 8x9cm occupying the entire vagina. On Per speculum examination there was a large mass protruding through external os and no cervical rim was identified around this mass. Ultrasonography (USG) was suggestive of a heterogeneous hyperechoic ill defined area of 9x8.8 cm, showing a mass affect in the left adnexal region. However exact nature and origin of the lesion could not be established due to poor acoustic window. An impression of ill defined heterogeneous left pelvic Space occupying lesion (SOL) was rendered. (Figure 3) MRI showed a large well defined polypoidal heterogeneously enhancing mass arising from the posterior wall of cervix, mildly invading into the posterior fornix and lower part of uterine myometrium. Bilateral fallopian tubes and ovaries were normal. Fat planes with rest of anterior and posterior vaginal

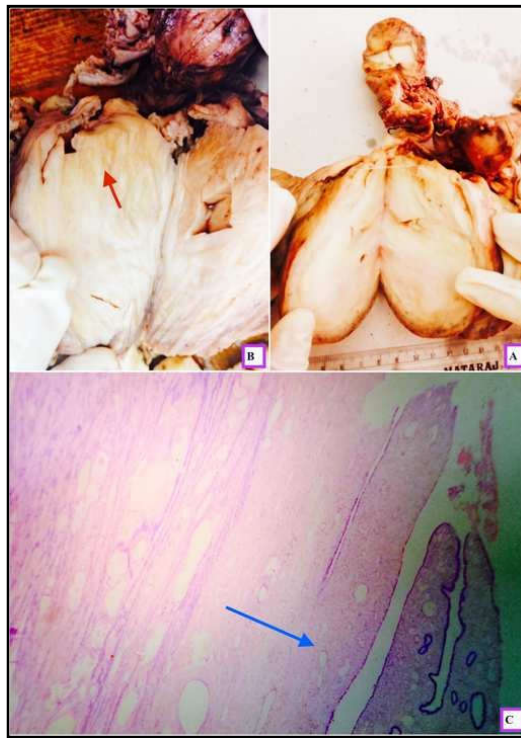


Figure 1. {A} Gross image of hysterectomy specimen showing cervical lipoleiomyoma originating from cervix. {B} Cut section of leiomyoma shows yellowish areas of lipomatous differentiation (red arrow). {C}. Low power microscopy image showing the leiomyoma originating from the endocervical region. (Blue Arrow) (H and E; 40X)

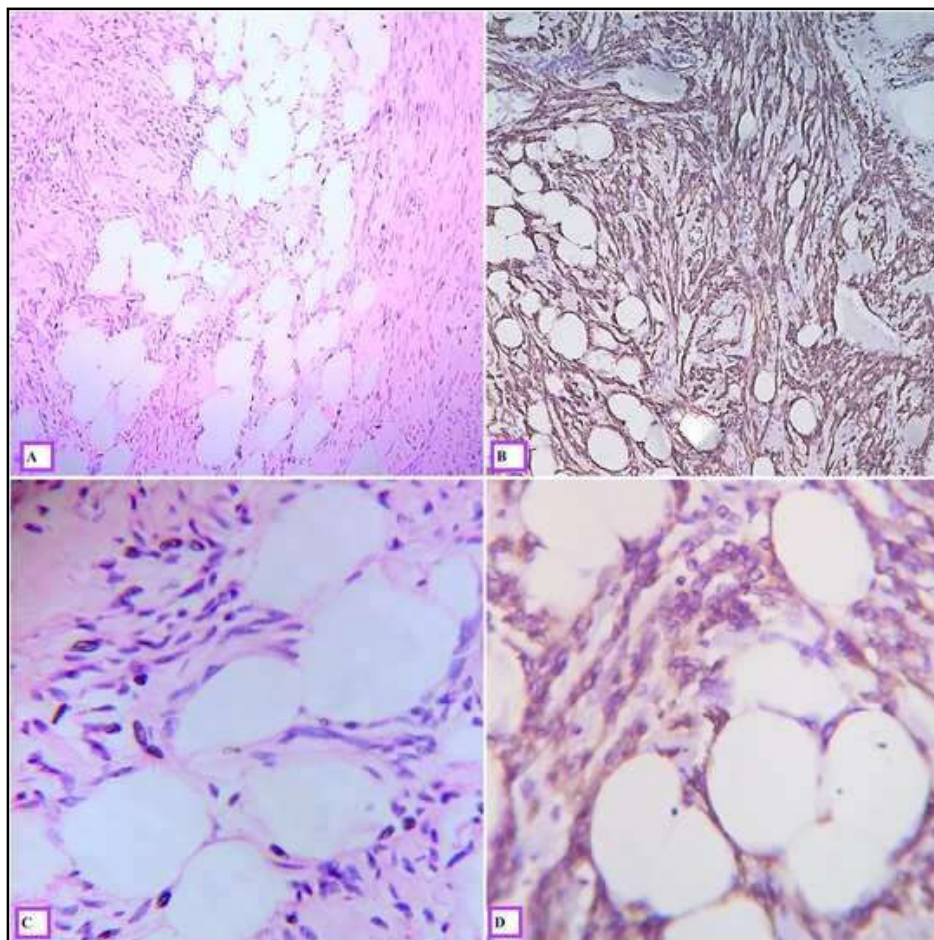


Figure 2: {A} Low power microscopy image of leiomyoma with areas of lipomatous differentiation.(H and E; 200x) {B} Smooth Muscle Actin cytoplasmic positivity seen in the smooth muscle cells on immunohistochemistry. (IHC; 200X). {C} High power image showing adipocytes scattered amongst the smooth muscle bundles. (H and E; 400X). {D} Smooth Muscle Actin Cytoplasmic positivity of smooth muscle cells as seen on high power (IHC;400X).



**Figure 3. Ultrasonography image showing a heterogeneous, hyperechoic ill- defined area of 9x8.8cm in the left adnexal region (red arrow in upper left pic)**

walls, rectum and urinary bladder were well maintained. Based on the above features the patient underwent total abdominal hysterectomy with unilateral salpingo-oophorectomy and the specimen was sent for histopathology. On gross examination, the specimen of uterus with cervix and unilateral adnexa measured 15x13x7 cm. Cervix showed the presence of polypoidal, well-circumscribed grey brown mass measuring 13x8x5 cm arising from the posterior lip of the cervix. On Cut surface the tumor was predominantly grey-white and homogeneous with an ill-defined whirling pattern. However some pale yellow areas were also noted at some places on cut section. No areas of hemorrhage and necrosis were seen. The attached ovary and fallopian tube were unremarkable grossly.

(Figure 1) On Microscopy the sections from the cervical growth showed a well circumscribed tumor tissue comprising predominantly of spindle shaped cells in interlacing fascicles and bundles along with interspersed areas of mature adipocytes in groups and singly scattered throughout the tumor tissue. (Figure 2A) The spindle shaped cells had moderate amount of cytoplasm, elongated cigar shaped blunt ended nuclei, fine chromatin, and inconspicuous nucleoli. There was no nuclear atypia, mitosis or necrosis. On Immunohistochemistry the spindle shaped cells were positive for Smooth Muscle Actin (SMA) (Figure 2B). Based on the above findings, a diagnosis of lipoleiomyoma was given. Sections from ovary and tubes were unremarkable

and endometrium was in proliferative phase. She had an uneventful postoperative course, recovered satisfactorily and is currently doing well under follow up for the last 3 months.

## DISCUSSION

Lipoleiomyoma is a rarely encountered histological subtype of leiomyoma, a benign smooth muscle tumor. They merely account for 0.03-0.2% of all the leiomyomas occurring in the uterus. Predominantly described in the uterine corpus, there have been reports describing their occurrence at uncommon sites like cervix, ovary and broad ligament. (Warty *et al.*, 1982; Pounder, 1982; Bajaj *et al.*, 2000) Cervical leiomyomas account for 1-2% of all the fibroids encountered clinically. Depending on their location in the cervix they can either be interstitial, supravaginal or polypoidal. Most of these patients are asymptomatic however when symptoms occur they correlate with the direction in which the tumor is enlarging and causing pressure symptoms like dysuria, ureteral obstruction or obstruction of the cervix. There are Only few case reports like ours where the patient presented with prolapse due to the large size of this tumor. The average size of these tumors is 5-10 cm, but cases with masses as large as 30 cm in diameter have also been reported in the past. (Kitajima *et al.*, 2007) According to the literature, uterine lipoleiomyomas occur at an average age of 50-70 years in post-menopausal women unlike our patient who was relatively young being 37 years of age. (Dharkar *et al.*, 1981) The exact incidence of cervical lipoleiomyoma however is still unknown due to the rare occurrence of this disease. However as per the series of Aung *et al.* (Aung *et al.*, 2004), Wang *et al.* (Wang *et al.*, 2006) and Akbulut *et al.* (Akbulut *et al.*, 2014) the mean ages reported in their study were 59.9, 53.9 and 55.49 years respectively.

The pathogenesis of Lipoleiomyoma of cervix is still controversial, as various theories have been proposed by different researches for the origin of lipomatous component of these tumors. In 1916, Lopstein highlighted the detailed histopathology of lipoleiomyoma. But, the term 'lipoleiomyoma' was brought into notice decades later, by Willen *et al.* and Pounder, when they classified the uterine fatty tumors into 'lipoma' and 'mixed lipoma/leiomyoma' i.e. lipoleiomyoma. (Pounder, 1982; Willen *et al.*, 1978) According to the later studies however it was proposed that the lipomatous lesion of the uterus arose from metaplasia of smooth muscle or connective tissue cells into fat cells. The other theories that were also proposed but are less accepted now included misplaced embryonic fat cells, lipocytic differentiation of a specific primitive connective tissue cell, fatty infiltration or degeneration of connective tissue itself or very rarely perivascular fat cells accompanying the blood vessels into the uterine wall during surgery (Sonam Sharma *et al.*, 2015; Bolat *et al.*, 2007). These tumors are quite often detected incidentally postoperatively as a chance pathological findings in most of the cases Although histopathology is the gold standard for diagnosis, today's modern day diagnostic modalities like CT and MRI play a significant role in the diagnosis of cervical lipoleiomyomas. (Wang *et al.*, 2006; Walid and Heaten, 2010) Unfortunately, USG, which is more affordable and accessible for poor patients in a developing country like ours, are not of much help in their diagnosis.

This is because on USG, the lesion is echogenic and is usually partially encased by a hypoechoic rim. The hypoechoic rim is thought to represent a layer of myometrium surrounding the fatty component but these findings are not considered to be specific for such tumors. The findings of CT and MRI on the other hand are more specific for detecting the fatty component of lipoleiomyomas. In our case also while the USG findings of this patient were inconclusive, MRI gave us a clearer view of the preoperative status of the tumor. Microscopically, lipoleiomyomas is composed of a variable admixture of muscular and adipose tissue. It is further classified into grades I-III depending on the amount of adipocytic abundance. (Akbulut *et al.*, 2014) Our case was of grade II lipoleiomyoma. The Differential diagnosis of adipocytic tumor in female pelvis includes spindle cell lipoma, angiolipoma, angiomyolipoma and well-differentiated liposarcoma. (Bolat *et al.*, 2007) However, in our case the absence of lipoblasts, atypia, mitotic activity or necrosis; and paucity of angiomatous elements helped us in making a final diagnosis of lipoleiomyoma.

As per the literature, there are only a limited number of case reports that demonstrated a histologic transition of a benign leiomyoma into a leiomyosarcoma. The age at presentation is an important factor however a study done by Gard *et al.* in 1999 concluded that the incidence of leiomyosarcoma was approximately one in one million with mean age of 55 yrs. (Gard *et al.*, 1999) Cytogenetic studies have exhibited that leiomyosarcoma occur de novo and might be unrelated to benign myomas. (Walker and Stewart, 2005) Similar to Leiomyoma the treatment of lipoleiomyoma is usually by hysterectomy as was done in our case. However other treatment modalities like Uterine artery embolization and myomectomy could also be performed depending on patient's symptoms, desire of fertility, the site of the mass, and other associated uterine pathologies (Agrawal *et al.*, 2014). These tumors share a good prognosis and usually do not recur after removal.

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