

Fibro-osteochondroma of the uterus – A case report with brief review of literature

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Abstract: A very rare case of fibro-osteochondroma arising in the uterine musculature is being presented here. This is the third reported case of this tumor. A 42 years old woman with a history of irregular vaginal bleeding, on ultrasonography revealed an intramural calcified mass in the uterus. On gross examination a well circumscribed firm to hard nodule measuring 1.0 cm in diameter was seen within the myometrium. Histologically the tumor showed a triphasic composition. Mature hyaline cartilage was seen peripherally in lobules surrounding bony trabeculae and a central hypocellular fibro-myxoid core with mature fibroblast-like spindle cells. Fibro-osteochondroma has to be differentiated from non-neoplastic heterotrophic cartilage and bone formation in myometrium, dystrophic calcification or ossification in a hematoma, implanted fetal tissue, mature teratoma and metaplasia within a preexisting leiomyoma or uterine sarcoma. So in a uterine lesion showing mature cartilage and bone formation, possibility of uterine fibro-osteochondroma should be considered. Diagnosis requires detailed clinical history and thorough histopathologic examination. This tumor is histologically and clinically benign and is not known to recur. Complete excision is curative. This case is being reported because of its extreme rarity.

Key Words: fibro-osteochondroma, osteochondroma, uterus

I. Introduction:

Presence of bone or cartilage within the uterine myometrium is an unusual phenomenon. This can be due to heterotrophic cartilage or bone formation as a part of a non-neoplastic process, implantation of fetal tissue or as a component of mature teratoma. Metaplastic changes in a preexisting tumor, most notably leiomyomas or uterine sarcomas have also been reported. Only two previous cases of the tumor designated as fibro-osteochondroma of the uterus have been reported.^[1,2] This benign tumor showing a triphasic histopathological appearance composed of mature chondroid, osseous and fibrous tissues have not been reported in any other site outside the uterus, although primary osteochondromatous tumors have been reported in variety of extraskelatal sites.^[1,2,3] Here we report the clinical presentation and histopathological features of fibro-osteochondroma of the uterus with a brief review of literature considering its differential diagnosis and possible histogenesis. This is the third reported case of this exceedingly rare tumor.

II. Case Report:

A 42 years old woman presented with a history of irregular, intermittent vaginal bleeding for 6 months. The patient is para 2+0, last child birth being 19 years back. On examination the patient showed pallor and the uterus was bulky. Ultrasonography revealed a small intramural fibroid with calcification measuring 1.0 cm in diameter. Haemoglobin was 8.9 gm % and peripheral smears showed a microcytic hypochromic blood picture. Other haematological investigations were within normal limits. The patient underwent hysterectomy. On gross

examination the uterus measured 8.5 cmx3.5 cmx3.0 cm and showed a well circumscribed firm to hard intramural nodule measuring 1.0 cm in diameter. Cut section of the nodule was whitish. The specimen was fixed in 10% buffered formalin, paraffin embedded and stained with haematoxylin and eosin stain. Microscopic examination of the nodule showed a tumor with a triphasic composition of mature fibrous, osseous and cartilaginous elements. Section showed peripheral area of mature hyaline cartilage in lobules surrounding bony trabeculae and a central hypocellular fibro-myxoid core. The cartilaginous tissue showed minimal pleomorphism of the chondrocyte nuclei. Central area showed mature fibroblast-like spindle cells with elongated bland nuclei. Smooth muscle, epithelial tissue, giant cells, mitosis or pleomorphism were not identified in the tumor. The patient was asymptomatic during the 3 months' follow-up period.

III. Discussion

Fibro-osteochondroma is a rare, histologically benign tumor which exhibits three types of cellular proliferation- fibroblastic, cartilaginous and osseous. Only two previous cases have been reported in the uterus by Fukuoka et al (1987) and Bates A.W et al (2012) who found similar histopathological picture of a triphasic tumor.^[1,2] A case of extraskeletal chondroma of fallopian tube was reported by Varras et al (2008) who suggested that mesenchyme of the myosalpinx of either smooth muscle or fibrous tissue lineage could represent the cell of origin^[4]. Fibro-osteochondroma of uterus is presumed to arise from similar undifferentiated precursor mesenchymal cells which undergoes metaplasia to form fibroblasts, chondrocytes and osteoblasts.^[1] The clinical features in our case and ultrasonographic findings which suggested a calcified fibroid were similar to the previously reported cases^[1,2].

Fibro-osteochondroma shows histological similarities with extraskeletal osteochondromas which usually arises near tendons and joints but have also been reported to arise from soft tissues^[3,5] However fibro-osteochondromas, unlike osteochondromas show fibro-myxoid component as seen in our case. Extraskeletal osteochondroma have no reported cases in the uterus.

Other differential diagnosis of chondroid and osteoid tissue in the uterus includes non-neoplastic heterotopic cartilage and bone formation, implanted fetal tissue, and metaplasia within a preexisting neoplasm.

Heterotrophic ossification or cartilage formation is rare in female genital tract with less than 100 reported cases^[6,7]. Possibility of dystrophic calcification or ossification in a hematoma is excluded in our case by absence of evidence of hemorrhage, hemosiderin deposition, and foreign body giant cell reaction.

In women of child-bearing age with history of instrumental abortion, possibility of "fetal homografts" have to be considered. This condition results in occurrence of mature cartilage, bone, epidermis or glia within uterine wall. Our patient has no history of recent amenorrhoea, pregnancy or abortion. However, the interval between pregnancy and diagnosis can be as long as 18 years^[8]. Both these conditions typically present with multiple deposits of a single tissue type within the myometrium^[2] rather than a solitary heterogeneous mass as was the presentation in our case.

Uterine leiomyomas may show benign or malignant osseous metaplasia. Osteosarcomatous differentiation has been reported in malignant mesenchymoma of uterus.^[9,10] Our tumor contained neither mature smooth muscle nor sarcomatoid areas; there were no features of malignancy and no epithelial component characteristic of a mature teratoma.

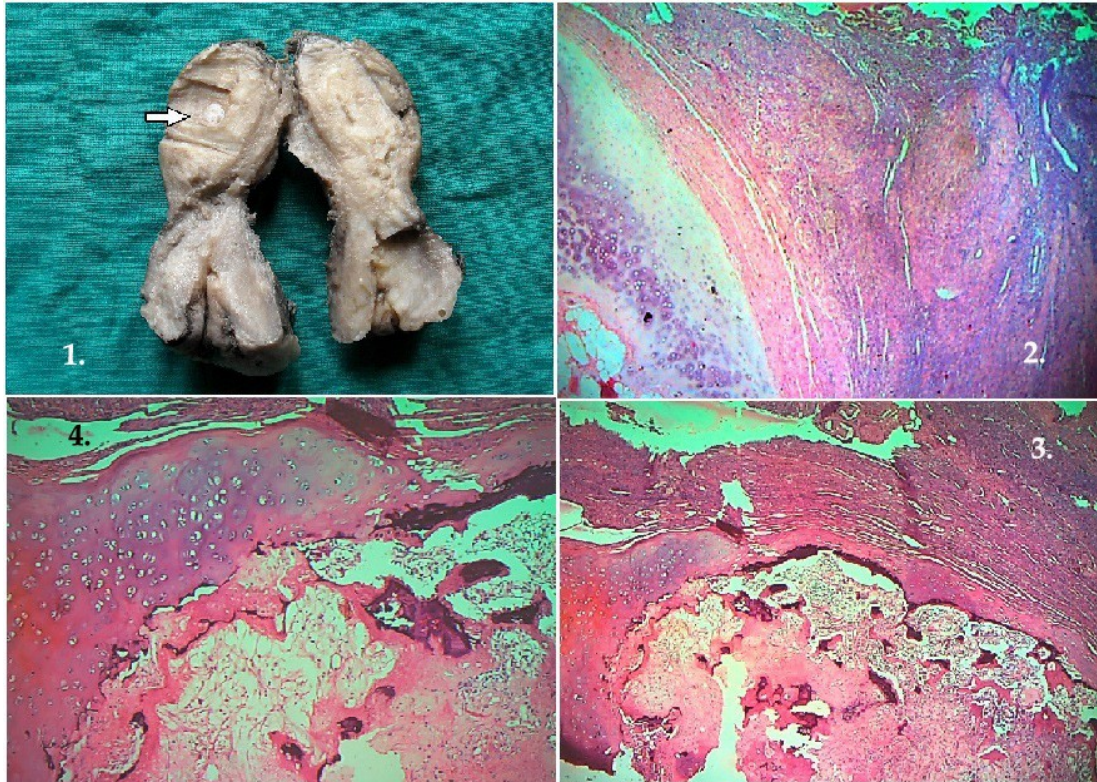
So in a uterine lesion showing mature cartilage and bone formation, possibility of uterine fibro-osteochondroma should be considered in the differential diagnosis. This tumor is histologically and clinically a benign and was not reported to recur.^[1,2] Complete excision is curative. This case is being reported because of its extreme rarity.

References:

- [1]. Fukuoka M, Fujii S, Konishi I, Mori T, Parmley TH, Woodruff JD. Fibro-osteochondroma of the uterus. *Obstet Gynecol* 1987;70:517-21.
- [2]. Bates AW, Ram M. Fibro-osteochondroma of the uterus. *Indian J Pathol Microbiol* 2012;55:555-6.
- [3]. Lim SC, Kim YS, Kim YS, Moon YR. Extraskeletal osteochondroma of the buttock. *J Korean Med Sci* 2003;18:127-30.
- [4]. Varras M, Akrivis Ch, Tsoukalos G, Plis Ch, Karadaglis S, Ladopoulos I. Tubal ectopic pregnancy associated with an extraskeletal chondroma of the fallopian tube: Case report. *Clin Exp Obstet Gynecol* 2008;35:83-5.
- [5]. Singh R, Sharma AK, Magu NK, Kaur KP, Sen R, Magu S. Extraskeletal osteochondroma in the nape of the neck: a case report. *J Orthop Surg (Hong Kong)*. 2006 Aug;14(2):192-5.

- [6]. Gerbie AB, Greene RR, Reis RA. Heteroplastic bone and cartilage in the female genital tract. *Obstet Gynecol* 1958;11:573-8.
- [7]. Ogbonmwan SE, Cher G. Endometrial ossification in postmenopausal women. *J Obstet Gynaecol* 2011;31:774-6.
- [8]. Grönroos M, Meurman L, Kahra K. Proliferating glia and other heterotopic tissues in the uterus: Fetal homografts? *Obstet Gynecol* 1983;61:261-6.
- [9]. Kefeli M, Baris S, Aydin O, Yıldız L, Yamak S, Kandemir B. An unusual case of an osteosarcoma arising in a leiomyoma of the uterus. *Ann Saudi Med* 2012;32:544-6.
- [10]. Den Bakker MA, Hegt VN, Sleddens HB, Nuijten AS, Dinjens WN. Malignant mesenchymoma of the uterus, arising in a leiomyoma. *Histopathology* 2002;40:65-70.

FIGURE:



LEGEND TO FIGURE:

1. Gross appearance of uterus with well circumscribed whitish intramural mass ; **2** Scanner view showing a well-circumscribed tumor within myometrium with overlying endometrium. **3.** Low power view- mature hyaline cartilage with benign appearing chondrocytes surrounding bone and central hypocellular fibro-myxoid tissue. **4.** High power view showing fibro-myxoid tissue with fibroblast-like spindle cells. (H & E stain)