Ovarian Fibroma with Metaplastic Bone Tissue: Case Report

Kemik Metastazı İçeren Over Fibromu

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ABSTRACT Fibromas are benign tumors of the ovary and arising from the stromal component of the ovary. Nearly half of the cases (40%) have tumor bigger than 10 cm in diameter. Only one third of the cases have smaller than 3 cm and they are rarely bilateral (5% of cases) In microscopic examination fibromas consist of cellular bundles and intersecting strips of hyaline-appearing collagen and fibrous tissue. Mitoses are rare, but occasionally fibromas may be hypercellular and/or show substantial mitotic activity. Dystrophic calcifications, focal necrosis and hemorrhage are common. In this report we described a case of unilateral fibroma of ovary that was found incidentally by laparoscopic surgery for chronic pelvic pain with pathologically consisting metaplastic bone tissue.

Key Words: Fibroma; bone and bones; laparoscopy

ÖZET Fibromlar overin stromal komponentinden kaynaklanan bening tümörlerdir. Olguların yaklaşık yarısı (%40) 10 cm'den daha büyüktür. Yanlızca üçte biri 3 cm'den daha küçüktür ve nadiren (olguların %5'i) bilateraldir. Fibromların mikroskobik bulguları hücre kümeleri ve hiyalen görünümlü kollajen ve fibröz doku şeritlerinden oluşur. Mitoz nadirdir, ancak bazen fibromlar hipersellüler olabilir ve/veya önemli mitotik aktivite gösterebilir. Distrofik kalsifikasyonlar, fokal nekroz, hemoraji ise yaygındır. Sunulan olguda, kronik pelvik ağrı nedeni ile laparoskopik cerrahi sırasında rastlantısal olarak saptanan ve patolojik değerlendirmesinde metastatik kemik dokusu içeren tek taraflı ovarian fibrom tanımlanmıştır.

Anahtar Kelimeler: Fibrom; kemik ve kemikler; laparoskopi

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Fibromas are benign tumors of the ovary and arising from the stromal component of the ovary. They represent a subgroup of the granulosa-theca cell tumors and belong to the thecoma-fibroma group according to the World Health Organization (WHO) classification of ovarian neoplasms.[1,2] Fibromas are the most commonly encountered subtype of the sex cord-stromal tumors fibromas, account for almost two-thirds of neoplasms in this group. The mean age at diagnosis is 48 years and 90% of patients are at least 30 years old when they are diagnosed with fibroma.[1,3] Although other sex cord-stromal tumors secrete hormonal products, fibromas are rarely associated with estrogen production. In macroscopic determination nearly half of the cases (40%) have tumor size bigger than 10 cm in diameter.[3] Only one third of the cases have smaller than 3 cm.[1-3] They are

rarely bilateral (5% of cases). Large tumors have a smooth or slightly irregular serosal surface and are generally solid. Small lesions may be seen as polypoid nodules over the ovary or as non-capsulated nodules in the ovary. The cut surface may sometimes demonstrate areas of cystic degeneration. Dystrophic calcifications may also occur, especially if the fibromas are seen in association with basal cell nevus syndrome[4] (features of this syndrome include basal cell carcinomas appearing early in life, and bilateral multinodular calcified ovarian fibromas also known as Gorlin syndrome). Ovarian fibromas have also been described in association with Maffucci and Sotos syndromes.[4-6]

In microscopic examination fibromas consist of cellular bundles and intersecting strips of hyaline-appearing collagen and fibrous tissue. Tumor cells have spindle shaped nuclei with no signs of atypia and produce collogen.[1-3] The cellularity varies inversely with the amounts of collagen production and stromal edema, both of which may be quite intense. Mitoses are rare, but occasionally fibromas may be hypercellular and/or show substantial mitotic activity. Cellular fibromas contain one to three mitoses per 10 high-power fields. Dystrophic calcifications, focal necrosis and hemorrhage are common.

Clinically they rarely cause symptoms.[3] These tumors are often an accidentally finding on radiological examination for various reasons. However, symptoms related to ascites and pleural effusion may develop, especially in those cases with large tumors. Ascites or classic Meig's syndrome with additional hydrothorax should be seen in 10% of all cases.

CASE REPORT

A 40 years old woman, gravida 2, parity 1, abortion 1 admitted our clinic with pelvic pain 7 month duration. Her menstural history was irregular and medical history was unremarkable except previous carpal tunnel operation. Gynecologic examination and transvaginal ultrasonography were also unremarkable. In preoperative period tumor markers and other biochemical parameters were normal.

Patient was operated by laparoscopically for chronic pelvic pain. In pelvic visualization uterus and right adnexa were normal. In left adnexa left ovary was strictly adherent to the left ovarian fossa. In the pouch of douglas some peritoneal adhesions were seen but not occluded (Figure 1 a,b,c). There was also left paratubal cyst in 1.5 cm diameter. After pelvic visualization an attemp to mobilize the left ovary and pelvic adhesiolysis was done. During the mobilization of the left ovary a solid mass which was 2.5-3 cm diameter, firm, irregular and adherent to the fossa was seen (Figure 2). This tumor was dissected from fossa and left ovary by preserving left ovarian function (Figure 3 a,b). Left salpingectomy and right tubal ligation was done

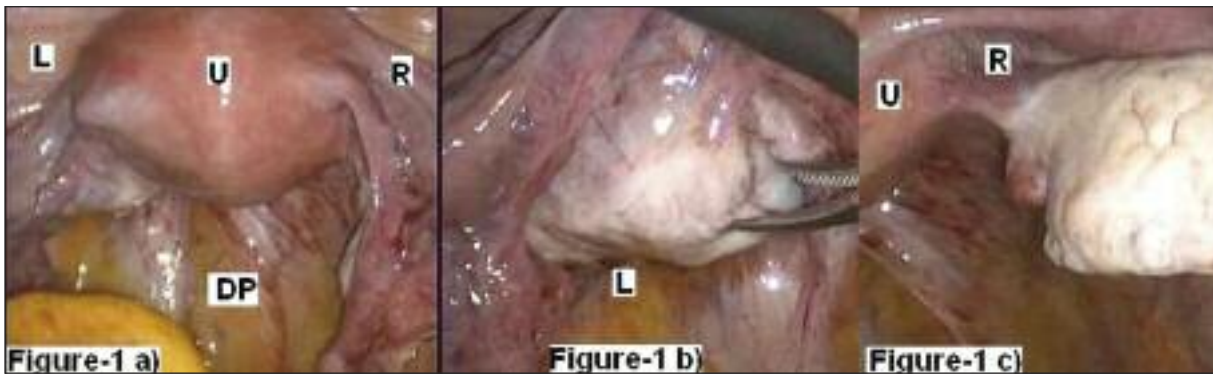


FIGURE 1: In left adnexa left ovary was strictly adherent to the left ovarian fossa. In the pouch of douglas some peritoneal adhesions were seen but not occluded. Pelvic vizualisatior.

a) Uterus and right adnexa were normal. In left adnexa left ovary was strictly adherent to the left ovarian fossa. In the pouch of douglas some peritoneal adhesions were seen but not occluded, **b)** In left adnexa left ovary was strictly adherent to the left ovarian fossa, **c)** Right adnexa were normal.

R: Right hand side, L: Left hand side, U: Uterus, PD: Pouch of douglas.

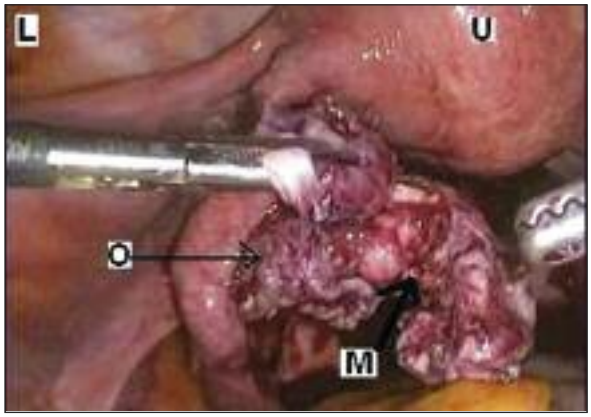
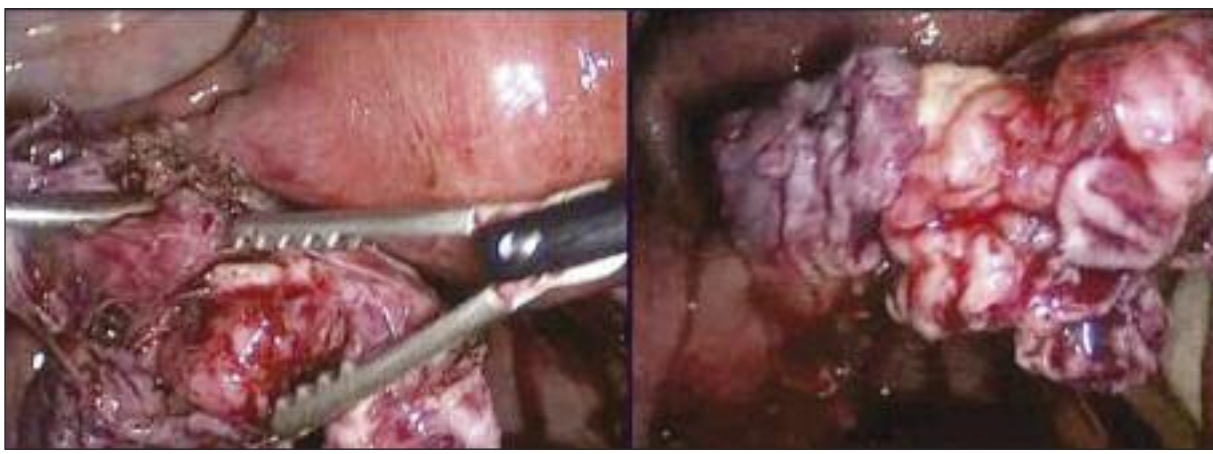


FIGURE 2: During the mobilization of the left ovary a solid mass which was 2.5-3 cm diameter, firm, irregular and adherent to the fossa was seen.
L: Left hand side, U: Uterus, O Over, M: Mass.

by the patients sterilization offer. Tumor was analyzed by frozen section but because of calcified nature of tumor, pathology revealed nothing intraoperatively. After peritoneal and omental biopsies, operation was terminated. The histopathologic examination revealed the presence of metaplastic bone and bone marrow over the fibroma tissue (Figure 4). Pelvic peritoneal and omental tissue were normal.

DISCUSSION

Fibromas are the most common benign solid neoplasms of the ovary. They account for approximately 5% of benign ovarian neoplasms and approximately 20% of all solid tumors of the ovary. Fibromas are tumors that contain spindle cells and grow slowly, with an average diameter of 6 cm. Some fibromas have been as large as 30 cm in diameter. Fewer than 10% of fibromas calcify. In our case fibroma was unilateral, solid, 3 cm in diameter and calcified.



a **b**

FIGURE 3: This tumor was dissected from fossa and left ovary by preserving left ovarian function.
a) The tumor was dissected from fossa and left ovary by preserving left ovarian function, **b)** A solid mass.
L: Left hand side, U: Uterus, M: Mass.

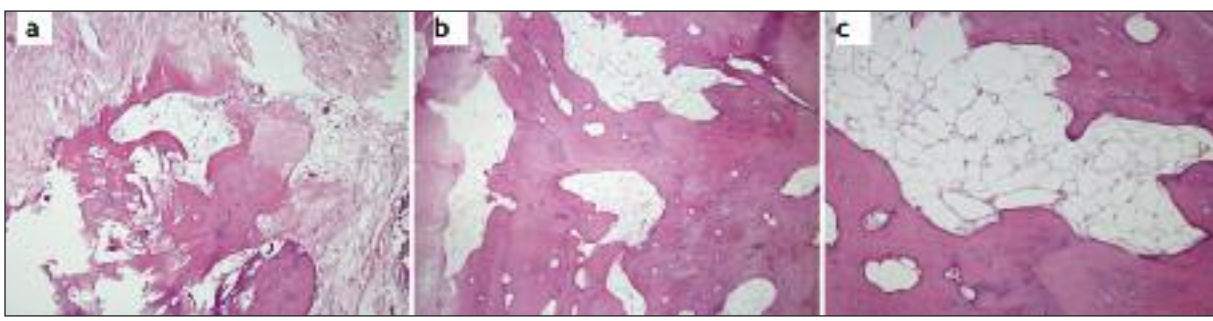


FIGURE 4: The histopathologic examination revealed the presence of metaplastic bone and bone marrow over the fibroma tissue
a) HE, x100, **b)** HE, x200, **c)** HE, x400.

Meigs' syndrome represents a benign condition which is described by the presence of a benign ovarian mass, associated with ascites and pleural effusion that resolve after the resection of the adnexal mass.[7] Despite earlier similar reports, Meigs' properly described the triad of the syndrome, initially in his book "Tumours of the female pelvic organs". Subsequently he published along with Cass a series of 7 cases with fibromas of the ovaries and the associated symtomps in 1937.[8] The ascite related to fibroma is thought to be caused by excessive transudate from the tumours surface in an amount that the peritoneum cannot absorb.[9] There are various theories about the mechanism of pleural effusion of which one supports the quick transfer of the ascitic fluid via transdiaphragmatic lymphatic channels or stomas.[10] The incidence of associated ascites is directly proportional to the tumor size. So in our case related to the small size of tumor there was no ascite or pleural fluid.

Level of CA 125 in fibromas are thougt to be related amount of ascites, not tumor itself directly. Irritation of the peritoneal surface may also explain the increased CA 125 levels associated with fibroma/fibrothecoma. Indeed, on immunoperoxydase staining, CA 125 is found to be localized within the peritoneum and not in the tumor.[11,12] In our case we did not see neither ascite nor rising levels of CA 125.

Nevoid basal cell carcinoma (NBCC) syndrome, also known as basal cell nevus syndrome and Gorlin syndrome, is a rare autosomal dominant condition with incidence of between one in 57.000-164.000 individuals.[13,14] This syndrome consists of developmental anomalies and susceptibility to cancer, especially basal cell carcinoma. NBCC syndrome is caused by mutations in the *PTCH* gene located on chromosome 9.[15] There many kinds of anomalies with this syndrome and one of them is ovarian fibromas. There are three large series that are published in 1990s about the prevalence of fibromas in Gorlin syndrome. According to these studies prevalence of fibromas in this syndrome is 14-24%. It was also reported that ovarian fibromas are more likely to be bilateral and calcified in patients with NBCC syndrome.[16] In our case patient and her relatives do not have history of any malignancy, her fibroma was unilaterally located and apart from this syndrome she do not have any malformation.

In pathologic examination fibromas consist of cellular bundles and intersecting strips of hyaline-appearing collagen and fibrous tissue. Tumor cells have spindle shaped nuclei with no signs of atypia and produce collogen.[1-3] The cellularity varies inversely with the amounts of collagen production and stromal edema, both of which may be quite intense. Mitoses are rare, but occasionally fibromas may be hypercellular and/or show substantial mitotic activity. Cellular fibromas contain one to three mitoses per 10 high-power fields. Dystrophic calcifications, focal necrosis and hemorrhage are common. Our case's pathologic examination revealed that no signs of malignancy in peritoneal and omental biopsies but metaplastic bone tissues were seen in fibromas tissue. As we described over calcifications may be seen but this case is first case in literature that bone tissue was seen in fibroma. Because of any sign of malignancy, surgery was limited to fibroma extirpation, peritonal and omental biopsies.

In conclusion we described a case of unilateral fibroma of ovary that was found incidentally by laparoscopic surgery for chronic pelvic pain with pathologically consisting metaplastic bone tissue.

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