Leiomyomatosis Peritonealis Disseminata Associated with Pregnancy: Case Report

GEBELİKLE BİRLİKTE GÖRÜLEN LEIOMYOMATOSIS PERITONEALIS DISSEMINATA: VAKA SUNUMU

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Abstract

Leiomyomatosis peritonealis disseminata (LPD) is characterized by multiple subperitoneal spread of nodules. A few cases of LPD associated with pregnancy have been reported in the literature. The histopathology of LPD is that of a benign leiomyoma, probably originating from the multipotent subcoelemic mesenchymal cells, but cellular origin of LPD is still not known exactly. LPD is usually a benign condition for which conservative management is indicated. A case report associated with pregnancy is presented below.

Key Words: Leiomyomatosis peritonealis disseminata, pregnancy

Özet

Leiomyomatosis peritonealis disseminata (LPD) çoklu subperitoneal nodüllerle karakterizedir. Literatürdeki az sayıdaki LPD vakası gebelikle birliktedir. LPD'nin histopatolojik orijini muhtemelen multipotent subçölomik mezenşimal hücrelerden oluşan benign leiomyomdur ancak LPD'nin selüler orijini hala tam olarak bilinmemektedir. LPD konservatif tedavi gerektiren benign bir durumdur. Aşağıda gebelikle ilişkili bir LPD olgusu sunulmuştur.

Anahtar Kelimeler: Leiomyomatosis peritonealis disseminata, gebelik

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Leiomyomatosis peritonealis disseminata (LPD) is a rarely found abdomino-pelvic and peritoneal pathology and it can stimulate disseminated abdominal carcinoma.[1] LPD is most common in reproductive ages and also was reported during or following pregnancy.[2] LPD is characterized with subperitoneal nodules seemingly composed of smooth muscle "disseminated" throughout the abdominal cavity. It is a benign condition for which conservative management should be recommended if fertility is desired.[3]

Case Report

HY, a 27 year old primigravid woman was admitted to the hospital at 41 weeks gestation with 2 centimeters cervical dilatation. On admission, transabdominal ultrasonographic examination revealed that a hypo and hyperechoic large mass measuring approximately 15-17 cm. in diameter like leiomyoma on antero-superior wall of the uterus. Laboratory measurements were in normal limits. After successful oxytocin challenge test, contractions were induced using intravenous oxytocin by thirteen hour. She underwent caesarean section because of cephalopelvic disproportion that was diagnosed after six centimeters cervical dilatation. Caesarean section was immediately performed. A low transverse uterine incision was made to deliver a living 4670 gr. female infant. During surgery, extending from uterine fundus to umbilicus with a thick stalk, a nodular, round polypoid degenerating tumour, around 16 cm. in diameter, resembling "atomic bomb" was found and multiple small hard nodules were noted scattered through the peritoneal surfaces. The nodules were 2-5 mm in diameter. Neither ascites nor

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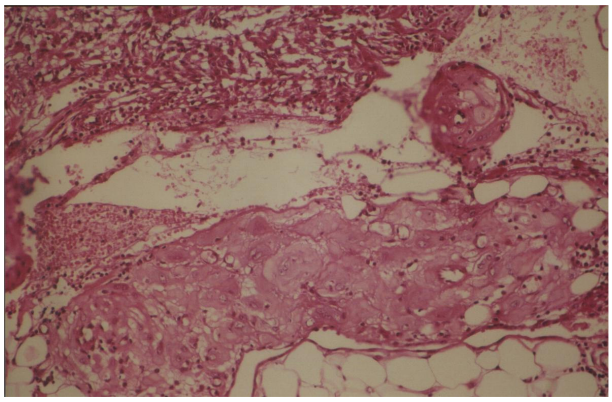

Figure 1. Peritoneal biopsy (H&E X 100).

hepatic involvement were observed. After dissecting the adhesions from transverse colon, omentum and umbilicus, large tumor with many nodules as possible of the small nodules removed and partial omentectomy were performed. During laparotomy, it could not be distinguished macroscopically from carcinomatous dissemination. The postoperative course was stable and she was discharged on fifth postoperative day. Pathologic examination has showed many gry-white nodules measuring 3-4 mm. in diameter in the omentum, peritoneum and the intestinal surfaces, and whorled bundles of spindle cells with no significant nuclear pleomorphism and mitotic activity (Figure 1). There was a dilated capillary vasculature, particularly in the periphery of the nodule, and an associated inflammatory reaction. Vascular invasion has not been found. Electron microscopy has confirmed the presence of the spindle cells, which are admixed with myofibroblasts, fibroblasts and decidual cells.

Discussion

LPD remains a condition primarily of the reproductive years. The clinical behaviour of LPD is generally agreed to be benign. LPD can be an incidental finding during caesarean section. The most common preoperative diagnosis is uterine leiomyomata.

Association with pregnancy, endometriosis and oral contraceptives suggest some influence of sex hormones on the pathogenesis of LPD.[4] The pathogenesis and cellular origin of LPD is still controversial. Leiomyomas have been induced experimentally in animals. LPD is probably derived from the stromal elements lying beneath the mesothelium lining the peritoneal cavity.[5] The mesothelium and the adjacent stroma have maintained multipotentiality even after embryological state. Noagles and Coll thought that the cells represented a transitional stage between mesenchymal and smooth muscle cells.[4] These authors suggested that the multicentric, metaplastic development into LPD is determined by an abnormal response to elevated hormone levels. Parmley and associates proposed an alternative pathogenesis for LPD. They suggested the nodules are not smooth muscle but fibrous tissue and speculated that an exaggerated decidual reaction may occur throughout the visceral and parietal peritoneal surfaces. Despite a benign histopathological view disseminated nature of LPD can give rise to diagnostic problems that some cases of LPD can be misdiagnosed classified as leiomyosarcoma.[6]

Total abdominal hysterectomy, bilateral salpingo-oopherectomy, omentectomy, myomectomy and debulking of the nodules can be performed at the time of diagnosis. However, treatment of LPD should be conservative since the disease can regress spontaneously after termination of pregnancy.[7] Spontaneous regression has often been described in LPD by reducing the estrogen exposure (surgical, postpartum, stopping oral contraceptives).[2] A conservative approach is recommended when LPD is diagnosed in association with pregnancy.[2] Follow up should include a pelvic examination, ultrasound and maybe laparoscopy.[8]

LPD of malignant change, adipocytic differentiation or sex-cord like pattern have been rarely reported and does not occur in our case.[9]

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