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ORIGINAL ARTICLE

Leiomyomatosis peritonealis disseminata as a possible result of laparoscopic myomectomy—report of four cases

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Abstract Between 2008 and 2010, four patients were treated for leiomyomatosis peritonealis disseminata, three were symptomatic and in one pelvic tumor was found during a routine pelvic examination. All four patients were in reproductive age and had never used any hormonal medication. Possible etiological factors were analyzed. The only common feature we found in these patients was laparoscopic myomectomy (in one additional laparoscopic supracervical hysterectomy 1 year after the first procedure) performed 3–11 years previously. Laparoscopy confirmed multiple myoma-like tumors and was converted to laparotomy in two patients; in the two patients, the tumors were removed laparoscopically. Laparoscopic myomectomy with morcellation could be an etiological factor for leiomyomatosis peritonealis disseminata. In selected cases and in experienced hands, the disease can be treated laparoscopically.

Keywords Leiomyomatosis peritonealis disseminata · Unclear etiology · Potential risk factor · Laparoscopic myomectomy

Introduction

Leiomyomatosis peritonealis disseminata (LPD) is a rare, usually benign tumor, and varying in size and number of smooth muscle cells that grow along the subperitoneal surface and can mimic a disseminated malignancy. The condition was first described in 1952 by Wilson and Peale [1]. LPD is usually discovered incidentally during a cesarean section or laparotomy for unrelated reasons or when searching for the cause of nonspecific pain or menorrhagia related to uterine myomas. Its etiology is poorly understood. This condition has mostly been described in case reports; therefore, different possible etiological factors have been proposed for its occurrence. They can be divided into hormonal, subperitoneal mesenchymal stem cells, metaplasia, genetic, or iatrogenic [2]. Increasing number of reports on LPD as a complication of laparoscopic myomectomy and morcellation support the iatrogenic theory [2–4], according to which small pieces of a myoma remaining after morcellation and their subsequent growth are supposed to be etiological factors.

Moreover, possible underlying factors have also been investigated. Several treatment modalities have been proposed such as discontinuation of hormonal medication, medical treatment, and surgery.

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Patients and methods

Laparoscopic myomectomy was introduced at our department in 1997 and laparoscopic supracervical hysterectomy (LASH) in 2000. Four patients with LPD were treated at the department between 2008 and 2010, the longest time interval since the initial procedure was 7 years. Three patients were symptomatic; one abdominal tumor was detected by transabdominal ultrasound examination done for unrelated reasons.

Informed consent was obtained from all patients for being included in the study.

Case 1

A 37-year-old woman, para 1, presented at the department 3 years after laparoscopic removal of a 10-cm myoma. She complained of abdominal pain lasting 2 months. Abdominal ultrasound scan and MRI revealed multiple tumors measuring from 1 to 4 cm in the abdominal cavity. Diagnostic laparoscopy revealed a uterine myoma and several myoma-like tumors in the rectosigmoid colon, appendix, and omentum and was

converted to laparotomy. Total hysterectomy with bilateral adnexectomy, appendectomy, and omentum resection were performed; on the request of a general surgeon, bowel resection was performed as well. Due to enlarged lymph nodes along the iliac vessels, lymphadenectomy was also performed, but frozen sections of the tumors and lymph nodes did not confirm malignancy.

Case 2

A 36-year-old woman, para 1, presented to the gynecologist who performed laparoscopic removal of a 10-cm myoma 6 years earlier for recurrent menorrhagia. Bimanual examination revealed an enlarged uterus of irregular shape, whereas abdominal and transvaginal ultrasound revealed tumors with sonographic characteristics of myomas. The patient was admitted to the hospital for hysterectomy. Laparoscopy revealed a 6-cm myoma on the anterior abdominal wall which was removed. On further exploration of the small pelvis and abdominal cavity, four myoma-like tumors 1 to 3 cm in size were found in the rectosigmoid colon and two small subserous myomas on the uterus. Laparoscopy was converted to laparotomy which further revealed three 2 mm myoma-like tumors and a myoma also in the abdominal fascia at the suprapubic trocar site. Biopsy and frozen sections excluded malignancy. The tumors were removed; the uterus and the adnexa were not.

Case 3

A 50-year-old patient, para 1, referred to the gynecologist who performed laparoscopic myomectomy 7 years earlier and laparoscopic supracervical hysterectomy without adnexectomy for recurrent myomas 6 years ago. She was asymptomatic but the transabdominal ultrasound examination performed for an unrelated reason detected a 7×6 cm large tumor in the small pelvis. A transvaginal ultrasound confirmed the tumor with ultrasonographic characteristics of a myoma. The preoperative diagnosis was a recurrent myoma on the cervical stump, but to evaluate the true nature of the tumor, laparoscopy was performed. The cervical stump was found normal, but behind it were two myomas measuring 5 and 4 cm large, three small myomas measuring 2 cm arising from the rectosigmoid colon were detected, and another one, 4 cm large arising was located retroperitoneally above the right tube. Biopsy and frozen sections excluded malignancy. Myomas were removed laparoscopically. Because of the patient's age, bilateral oophorectomy was performed as well despite apparently functional ovaries.

Case 4

A 50-year-old woman, para 2, was referred to the department for a 5-cm tumor in the pelvis behind the cervix. The transperitoneal involvement of the colon was excluded by colonoscopy as requested by the general practitioner. The patient underwent laparoscopic myomectomy and tubal sterilization 11 years earlier. She never missed her annual gynecological examination, but the tumor was first detected at the last visit only. She reported discomfort in the small pelvis lasting about 3 months. Preoperative diagnosis, based on bimanual examination and transvaginal ultrasound scan, was a cervical myoma. Laparoscopy revealed a 5-cm myoma-like tumor in the subperitoneal space of the rectum and 5 and 4 cm nodules on the sigmoid colon. All tumors were removed laparoscopically. The final pathologic diagnosis—benign myoma agreed with frozen section biopsy.

None of the four patients was ever taking any hormone therapy.

The tumors removed at primary surgery were diagnosed by the pathologist as benign myomas and described as densely cellular spindle-shaped smooth muscle tumors in all four patients. Histological description of LPD nodules was completely identical.

There were no intra- or postoperative complications. Currently, all patients are symptom free and no recurrence has been registered (follow-up period from 2 to 5 years).

Tumors were removed laparoscopically using electric morcellator; the defects on the site of enucleation were peritonized, after which the abdominal cavity was extensively washed performed to remove any remaining small particles.

Discussion

LPD is most prevalent in women of reproductive age, particularly in those subjected to altered hormonal milieu, such as prolonged use of oral contraceptives [5, 6], hormonal replacement therapy [7], pregnancy [8], estrogen secreting tumors [9], and ovarian stimulation [10]. Al-Tahib and Tulandi encountered 132 of LPD in the English literature, 113 were described in premenopausal women, 7 in postmenopausal women, 6 in males, 1 in a horse, 1 in a fetus, and 4 were not classified [2].

None of our four patients was exposed to increased exogenous or endogenous estrogen concentrations. The only common factor was previous laparoscopic myomectomy done with the aid of electric morcellator. Laparoscopic myomectomy or laparoscopic supracervical hysterectomy with morcellation has been reported as a possible cause of LPD in recently published papers [2, 11–15]. Therefore, multiple tumors in the abdominal cavity should raise suspicion of LDP in patients with a previous history of laparoscopic myomectomy or LASH. Tumors are usually detected in symptomatic patients or at regular checkups with imaging techniques such as ultrasound, CT, or MRI [16–18]. The reports on the use of these techniques in LPD are rare because many reports are old and because many LPD cases were usually found incidentally at laparotomy for unrelated reasons. Regardless of the

sensitivity and specificity of imaging techniques, the differential diagnosis between malignant and benign nature of the disease can be made only by biopsy and histological evaluation, particularly if we take into account that LPD can mimic peritoneal carcinomatosis [19, 20] or can be complicated, although extremely rarely, by sarcomatous transformation [21, 22], and that the treatment of malignancy is significantly different from that of a benign disease.

To evaluate the nature of preoperatively diagnosed tumors and the extent of the disease, we performed diagnostic laparoscopy and intraoperative frozen sections of myoma-like tumors to exclude malignancy in all four patients before taking the decision on the type of treatment. Spontaneous regression of LPD has been reported after discontinuation of hormone therapy, such as oral contraceptives or after pregnancy [23, 24]. Reports on medical treatment are rare, a successful treatment of LPD with the aromatase inhibitor anastrazole and with gonadotropin-releasing hormone agonists has been reported [25, 26]. Surgical treatment of LPD is usually the method of choice, particularly in symptomatic patients with no history of exogenous hormonal therapy, but there is no consensus on the type of the surgical procedure, although hysterectomy with oophorectomy is suggested in women with completed childbearing. Hysterectomy with oophorectomy was performed in one of our four patients and oophorectomy in one, whereas ovaries were left in place in two patients, although it is still debatable whether ovaries should be removed or not. The purpose of oophorectomy is to deprive patients from estrogen stimulus which is supposed to play the most important role in tumor growth. Unfortunately, this theory cannot explain the occurrence of LPD in postmenopausal women [27–29], in women after abdominal hysterectomy [22, 30], and in men [31]. The same is true of the procedures of the bowel. Bowel resection is rarely indicated because tumors usually involve the subserosa only and rarely cause bowel obstruction, a rare indication is obstruction of the small bowel [32].

Looking back, we may conclude that the extremely radical procedure applied in our first case was unnecessary, but it was the general surgeon's decision due to lack of knowledge and experience with the disease of both the general surgeon and the gynecologist, as this was the first case they met within their carrier. Due to frequent involvement of the bowel, the general surgeon and oncologist are usually the first to meet with the disease and they may not be always familiar with its nature, although a minimally invasive approach in the treatment of LPD was reported and proposed already in 1994 by Krueczynsky [33]. Among all the cases treated surgically, laparoscopic approach was used in two only [12, 33]; in all others, laparotomy was performed. In cases 3 and 4, we removed tumors laparoscopically. Surgery was performed by gynecologists experienced in advanced laparoscopic surgery such as pelvic and paraaortic lymphadenectomy and bowel and urinary tract endometriosis. In our experience, the laparoscopic approach is safe and feasible, although the procedure can be time-consuming in case of multiple tumors, but not extremely demanding because the encapsulated nodules grow subperitoneally and do not invade the lumen of the adjacent organs.

An increasing number of reports on laparoscopic myomectomy with morcellation suggest morcellation as a possible cause of the development of LPD. Small particles of the minced tissue remaining in the abdominal cavity grow to become LPD under exposure to steroid hormones and growth factors. The only common factor that could be pointed out in the four cases here presented was laparoscopic myomectomy and no other previously mentioned potential risk factor in their previous history. Regarding thousands of laparoscopic myomectomies and LASHs performed all over the world, over 2,500 at our department only, the occurrence of LPD as a complication of myomectomy is rare. In addition to the particles that remained in the abdominal cavity and the iatrogenic theory, there must be some underlying etiological factors in these women. Some authors have suggested the genetic theory and abnormalities of chromosomes X, 8, 12, and 17 [13]; the hormonal theory explains the occurrence of LPD with proliferation of subcelomic submesenchymal cells and their differentiation into myoblasts, myofibroblasts, and fibroblasts under estrogen stimulation [34]. The diagnosis is usually based only on histological evaluation confirming benign myoma, whereas immunohistochemical evaluation has been rarely reported. Danikas and co-workers explained the occurrence of LPD in postmenopausal women with the presence of LH receptors [35], Butnor and co-workers found progesterone receptor activity in pre- and postmenopausal women as well [24], Takeda and co-workers found progesterone receptors positive in leiomyomas that were histologically almost identical to the myoma at primary surgery [12], and Ruscalleda and co-workers found negative estrogen and positive progesterone receptors [17].

Conclusion

Despite the above mentioned theories, the etiology of the occurrence of LPD remains unclear. The iatrogenic theory is supported by increasing number of reports on LPD as a complication of laparoscopic surgery with morcellation. Because small particles of minced tissue remaining in the peritoneal cavity after morcellation are supposed to be much more frequent as is the occurrence of LPD, these procedures are probably only a co-factor for LPD development and further research is required to find possible underlying factors. LPD occurs only rarely, but due to increasing number of laparoscopic myomectomies and LASHs, the number of cases is expected to increase; therefore, physicians faced with LPD must be familiar with the disease. If surgery is indicated and malignancy excluded, minimally invasive surgical approach is, according to our experience, feasible and safe, but should be

performed by surgeons skilled in advanced laparoscopic procedures.

Conclusion for practice

LPD is a rare condition, the etiology of which is unknown; laparoscopic myomectomy and morcellation can be a possible risk factor for LPD. Extensive washing of the abdominal cavity at the end of the procedure is proposed to reduce the risk of the occurrence of LPD.

Conflict of interest The authors report no conflicts of interest. The authors alone are responsible for the contents and writing of the paper.

References

1. Wilson JR, Peale AR (1952) Multiple peritoneal leiomyoma associated with a granulose-cell tumor of the ovary. Am J Obstet Gynecol 64:204–208
2. Al-Talib A, Tulandi T (2010) Pathophysiology and possible iatrogenic cause of leiomyomatosis peritonealis disseminata. Gynecol Obstet Invest 69:239–244
3. Hilgar WS, Magrina JE (2008) Removal of pelvic leiomyomata and endometriosis 5 years after laparoscopic supracervical hysterectomy. Obstet Gynecol 108:772–774
4. Kumar S, Sharma JB, Verma D et al (2008) Disseminated peritoneal leiomyomatosis: an unusual complication of laparoscopic myomectomy. Arch Gynecol Obstet 278:93–95
5. Dallenbach C, Miklaw H, Dallenbach FD et al (1989) Disseminated peritoneal leiomyomatosis: a rare disease picture. Geburtshilfe Frauenheilkd 49:201–204
6. Wise LA, Palmer JR, Harlow BL et al (2004) Reproductive factors, hormonal contraception, and risk of uterine leiomyomata in African-American women: a prospective study. Am J Epidemiol 159:113–123
7. Heinig J, Neff A, Cirkel U, Klockenbusch W (2003) Recurrent leiomyomatosis peritonealis disseminata after hysterectomy and bilateral oophorectomy during combined hormonal replacement therapy. Eur J Obstet Gynecol Reprod Biol 111:216–218
8. Izadi-Mood N, Azordegan N (2003) Disseminated peritoneal leiomyomatosis: a case report. Arch Iran Med 6:216–218
9. Drake A, Dhundee J, Buckley CH et al (2001) Disseminated leiomyomatosis peritonealis in association with estrogen secreting ovarian fibrothecoma. Br J Obstet Gynaecol 108:661–664
10. Deering S, Miller B, Kopelman J et al (2000) Recurrent leiomyomatosis peritonealis disseminata exacerbated by in vitro fertilization. Am J Obstet Gynecol 182:725–726
11. Epstein JH, Nejat EJ, Tsai T (2009) Parasitic myomas after laparoscopic myomectomy: case report. Fertil Steril 91(3):13–14
12. Takeda A, Mori M, Sakai K et al (2007) Parasitic peritoneal leiomyomatosis diagnosed 6 years after laparoscopic myomectomy with electric morcellation: report of a case and review of the literature. J Minim Invasive Gynecol 14:770–775
13. Miyake T, Enomoto T, Ueda Y et al (2009) A case of disseminated peritoneal leiomyomatosis developing after laparoscope-assisted myomectomy. Gynecol Obstet Invest 67:96–102
14. Osterzensky A (1997) Uterine leiomyoma particle growing in an abdominal wall incision after laparoscopic retrieval. Obstet Gynecol 89:853–854
15. Paul PG, Koshy AK (2006) Multiple peritoneal parasitic myomas after laparoscopic myomectomy and morcellation. Fertil Steril 85:492–493
16. Guarigilia L, Conte M (1996) Sonographic findings in a case report of benign leiomyomatosis peritonealis disseminata. Acta Obstet Gynecol Scand 75:804–805
17. Ruscalleda N, Eixarch E, Pages M et al (2006) Leiomyomatosis peritonealis disseminata. Eur Radiol 16:2879–2882
18. Demir MK (2007) Disseminated peritoneal leiomyomatosis: magnetic resonance imaging and differential diagnosis. Australasian Radiol 51:B268–B271
19. Altinok G, Usubütün A, Küçükali T et al (2000) Disseminated peritoneal leiomyomatosis. A benign entity mimicking carcinomatosis. Arch Gynecol Obstet 264:54–55
20. Ramesh L, Edozien LC (2005) Not all disseminated intra-abdominal lesions are malignant: a case of leiomyomatosis peritonealis disseminata. J Obstet Gynecol 25:409
21. Fulcher AS, Szucs RA (1998) Leiomyomatosis peritonealis disseminata complicated by sarcomatous transformation and ovarian torsion: presentation of two cases and review of the literature. Abdom Imaging 23:640–644
22. Sharma P, Chaturvedi KU, Gupta R et al (2004) Leiomyomatosis peritonealis disseminata with malignant change in a postmenopausal woman. Gynecol Oncol 95:742–745
23. Düe W, Pickartz H (1989) Immunohistological detection of estrogen and progesterone receptors in disseminated peritoneal leiomyomatosis. Int J Gynecol Pathol 46–53
24. Butnor K, Burchette JL, Robboy SJ (1999) Progesterone receptor activity in leiomyomatosis peritonealis disseminata. Int J Gynecol Pathol 18:259–264
25. Takeda T, Masuhara K, Kamiura S (2008) Successful management of a leiomyomatosis peritonealis disseminata with an aromatase inhibitor. Obstet Gynecol 112(2 Pt 2):491–493
26. Hales HA, Petrerson CM, Jones PK, Quinn JD (1992) Leiomyomatosis disseminate treated with a gonadotropin-releasing hormone agonist. Am J Obstet Gynecol 167:515–516
27. Komatsu M, Tsuchya S, Kuroda T (1996) Leiomyomatosis peritonealis disseminata occurring in a postmenopausal woman: report of a case. Jpn J Surg 26:700–703
28. Strinić T, Prusac-Kuzmić I, Eterović D et al (2000) Leiomyomatosis peritonealis disseminata in a postmenopausal woman. Arch Gynecol Obstet 264:97–98
29. Nigojević S, Kapural L, Scukanec-Spoljar S et al (1997) Leiomyomatosis peritonealis disseminata in a postmenopausal woman. Arch Gynecol Obstet Scand 26:893–894
30. Rajab KE, Aradi AN, Datta BN (2000) Postmenopausal leiomyomatosis peritonealis disseminata. Int J Gynecol Obstet 68:271–272
31. Yamaguchi T, Imamura Y, Yamamoto F et al (2003) Leiomyomatosis peritonealis disseminata with malignant change in a man. Radiographics 53:179–185
32. Ghosh K, Dorigo O, Bristow R, Berek J (2000) A radical debulking of leiomyomatosis peritonealis disseminata from colonic obstruction: a case report and review of the literature. J Am Coll Surg 191:212–215
33. Krueczynsky D, Merz E, Knapstein PG (1994) Minimal invasive therapie bei peritoneal leiommyomatose. Geburtshilfe Frauenheilkd 167:181–183
34. Tavassoli FA, Norris HJ (1982) Peritoneal leiomyomatosis (leiomyomatosis peritonealis disseminata): a clinicopathologic study of 20 cases with ultrastructural observations. Int J Gynecol Pathol 1:59–74
35. Danikas D, Goudas VT, Rao CV et al (2000) Luteinizing hormone receptor expression in leiomyomatosis peritonealis disseminata. Obstet Gynecol 95:1000–1011