Isolated broad ligament leiomyomatosis

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Abstract

Leiomyomas of an extraterine nature are rare and often present a more challenging diagnosis process for the clinician. Our patient presented with lower abdominal pain associated with menorrhagia over a period of 6 months. Laparoscopic surgery revealed a right-sided broad ligament myoma consisting of multiple soft and diffused fibroids, of more than 400 in total. The myomas were enucleated and completely removed by laparoscopic surgery with minimal blood loss. Histopathological examination and analysis revealed the multiple nodules to be benign leiomyomatosis. The finding of isolated broad ligament leiomyomatosis rates this case as a unique kind of leiomyoma.

Key words: broad ligament, extrauterine, laparoscopy, leiomyoma, leiomyomatosis.

Introduction

Fibroid formation can be kaleidoscopic, from minute myomatous nodules to huge myomas; be it singular, isolated or even multiple. Clinically presenting with a variety of symptoms, such as abnormal uterine bleeding, pelvic discomfort, urine frequency, dyspareunia, and constipation; its severity is dependent on the number of tumors, size and location. Myomectomy remains the ideal therapeutic option for the removal of symptomatic leiomyoma in women with a desire to preserve their fertility. The option is even more attractive since laparoscopic approach has become increasingly feasible. A broad ligament leiomyoma is categorized as an extrauterine leiomyoma. Although it is believed that uterine leiomyomas commonly affect 20–30% of women over 35 years of age, leiomyomas of an extrauterine nature are even rarer and often present a more challenging diagnosis process.1 The finding of isolated broad ligament leiomyomatosis rates this case as a unique kind of leiomyoma. We compared the various existing defined and documented uterine smooth-muscle tumors and concluded that although there are similarities to certain extents, our case is different. We suggest that this be considered as a new pathological entity, which will hopefully will entice more reports of a similar nature in the future to give us a clearer understanding of this condition.

Case Report

A 42-year-old Taiwanese woman, para 2, came to our center seeking treatment for lower abdominal pain over a period of 6 months. Her menstruation cycles had been regular; nevertheless, she stated mildly prolonged menorrhagia. A gynecological ultrasonography (USG) (Fig. 1a) revealed an extrauterine solid mass measuring 9 cm in diameter. A supporting computed tomography (CT) scan (Fig. 1b) detected a heterogeneous and hypodense mass of similar size with an indistinct margin on the right lateral wall of the uterus with no masses detected within the uterine cavity or uterine wall. There were no other abnormal findings and clinical tumor markers were all within the accepted normal range.
Laparoscopic surgery was performed. A 10-mm principal cannula was introduced midpoint between the umbilicus and the xiphoid process (Lee-Huang point). In our experience, the Lee-Huang point approach provides safe and practical placement of the primary trocar, especially in patients with large pelvic masses. The median longitudinal incision through the linea alba prevents penetrative injury to major blood vessels and provides the operator with a wider access to the abdominal cavity, increases the working distance and supports a better visual angle. Two 5-mm ancillary cannulas were then placed under laparoscopic visualization: one in the right lower quadrant lateral to the inferior epigastric arteries and one in the left lower quadrant. Another 5-mm cannula was placed at the left paramedian line at the level of the umbilicus, to complete the two-hand assisted technique. Intraoperatively we found a right-sided broad ligament myoma (Fig. 2). No other leiomyomas were present on the uterus and no other pelvic masses were detected. Taking into consideration that a broad ligament leiomyoma may commonly distort the normal anatomical route of the ureter; we carefully traced and identified the right ureter before proceeding further. Respecting the mesoureter and its blood supply, the ureter was not dissected or skeletonized, but was merely identified and lateralized away slightly. An incision was then made onto the serosa overlying the tumor using a unipolar electrode. The incision was extended until the characteristically pearly white substance of the leiomyoma was exposed. Upon further dissection, we noticed that encompassed within the broad ligament were multiple soft and diffused minute fibroids (Fig. 3). Using endoscopic forceps, blunt dissection was performed separating the mini cleavage planes; gently pulling and removing the minute leiomyomas, depositing all of them into a specimen retrieval system (Endobag 10 mm; United States Surgical/Tyco Healthcare Group, Norwalk, CT, United States).
USA). We successfully enucleated and removed all the leiomyomas with minimal blood loss. These multiple myomatous nodules were each smaller than 1 cm (ranging from 0.1–1.0 cm) and numbered over 400 pieces in total (Fig. 4). Electrocoagulation of vessels and bleeding points were performed using bipolar mode Kleppinger forceps. The remaining uterine surgical defect was then sutured using a 1/0 monofilament poliglecaprone 25 (Monocryl; Ethicon, Somerville, NJ, USA).

A continuous running non-lock suture and intracorporeal knot-tying method was used. By slightly enlarging the right lower quadrant skin incision, the Endobag could be removed easily from the abdominal cavity. The operative procedure consumed approximately 90 min with neither intra- nor postoperative complications and the patient was discharged in a good condition on the second postoperative day (as per hospital protocol). The histopathological examination and analysis revealed the multiple nodules to be benign leiomyomatosis.

Discussion

Gynecologists have long established that benign smooth-muscle tumors (leiomyomas) are the most common tumors found in the female genital tract. With a possibility of leiomyomas arising from anywhere in the genitourinary tract (vulva, ovaries, urethra, urinary bladder), these histologically benign tumors can still arise in nearly any anatomic site. Although the diagnosis is usually clear-cut, infrequently we encounter quizzical presentations requiring clarification. As a diagnostic armamentarium, pathologists have reviewed and structured a division of three major groups: (i) smooth-muscle tumors with clearly recognized smooth-muscle differentiation – these include benign, malignant and of uncertain malignant potential; (ii) smooth-muscle tumors with no clearly recognized smooth-muscle differentiation, such as myxoid, epithelioid, clear cell and granular cell leiomyomas, as well as leiomyomas with sex cord-like pattern; and (iii) smooth-muscle tumors with unusual growth patterns, such as diffuse uterine leiomyomatosis, intravenous leiomyomatosis, benign metastasizing leiomyoma, disseminated peritoneal leiomyomatosis, perinodular hydropic leiomyoma, multinodular hydropic leiomyoma, infiltrative uterine leiomyoma and cotyledonoid dissecting leiomyoma.4

We believe that our case predominantly falls into the third grouping. In a recent supporting review, intravenous leiomyomatosis (IVL), benign metastasizing leiomyoma (BML) and leiomyomatosis peritonealis disseminate (LPD) have been identified to represent the three major primary neoplasm of uterine smooth-muscle tumors with unusual growth patterns;5 four other less common conditions; diffuse uterine leiomyomatosis, parasitic leiomyoma, cotyledonoid leiomyoma and retroperitoneal growth, have also been reported.15–9 Our patient’s clinical presentation was very interesting because it did not fit into any of the
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aforementioned conditions, despite closely mimicking diffuse uterine leiomyomatosis (DUL). DUL is a rare condition in which the entire myometrium is embedded with innumerable, ill-defined, small and confluent myomas, specifically noted to cause symmetrical enlargement of the uterus. Typically, the diffuse nature denotes difficulty in performing a complete myomectomy, hence hysterectomy was always recommended. This is a stark difference when compared to our case as the featured leiomyomatosis was isolated, located within the right side of the broad ligament with neither uterine enlargement nor involvement of the entire corpus uterus (further supported and confirmed by computed tomography scans). IVL primarily is defined as the presence of smooth-muscle cells growing in venous and lymphatic vessels without invading them. The etiology has been thought to arise from the uterine leiomyoma entering into the venous system, while another theory suggests a direct extension from the walls of the uterine veins. Hence, we cannot categorize this presentation as IVL as our patient did not have a primary myoma in the uterus.

It is also unlikely for the condition to be categorized as BML as it is usually characterized as the presence of multiple smooth-muscle nodules frequently located in the lung, lymph nodes or abdomen; furthermore, BML is commonly diagnosed 3 months to 20 years after a hysterectomy due to fibroids. LPD is possible; however, in that scenario, the lesion is usually characterized by the presence of multiple hard rubbery small nodules on the abdominal and pelvic peritoneal surfaces, including the bowel serosa, which is not so in our case as all 400-odd myomatous nodules were well-circumscribed and encapsulated within the broad ligament. Consideration was given to cotyledonoid leiomyoma as a probable neoplasm given its multinodular, pedunculated appearance, which often extends into the broad ligament or peritoneal cavity. However, it was ruled out as this ‘grapelike’ tumor is usually large, reddish, exophytic, fungating and histologically has marked hydropic change and rich vascularity. Through laparoscopic surgery, we were able to resect all nodules and confirm visually that there were no other leiomyomas in the uterus; neither was there any invasion of myomatous tissue into the surrounding blood vessels. The etiology of such a fibroid formation is still unknown, although some reports indicate its association to genetic abnormalities. Clonality analysis suggests the independent origin of neoplastic clones for each single myomatous nodule and rejects the possibility of a single clonal origin of all tumor cells. Reported cases of leiomyomatosis have not demonstrated a fast regrowth of the corpus, neither rapid nor new proliferation of other nodules. This suggests that the potential for relapse is low. Nevertheless, in the follow-up period of more than 4 years, our patient has not had a relapse. Future studies directed towards the understanding of its pathophysiology, pathogenesis and etiology will be attractive and will be very much appreciated by the academic cohort.

References

