Laparoscopic management of a true broad ligament leiomyoma in a patient with advanced endometriosis and a solitary kidney – A case report and literature review

Vishal Bahall *, Lance De Barry

Department of Obstetrics and Gynaecology, San Fernando, General Hospital, South-West Regional Health Authority, Trinidad and Tobago

ARTICLE INFO

Keywords: Broad ligament Leiomyoma Fibroid Minimally invasive surgery Case report

ABSTRACT

Leiomyomas are the most common benign tumours of the female genital tract, and almost always arise from the uterine myometrium. Although extraterine leiomyomas are rare, they usually develop in sites such as the ovary, broad ligament, round ligament, cervix or abdominal wall. The broad ligament is the most common site of extraterine leiomyoma involvement, and this unique clinical entity may prove to be a diagnostic or therapeutic challenge, particularly in patients with advanced endometriosis and distorted pelvic anatomy. Herein, we report the case of a large true broad ligament leiomyoma that was discovered during a total laparoscopic hysterectomy and bilateral salpingo-oophorectomy in a 47-year-old patient with stage IV endometriosis and a congenital left kidney and left ureter. This case highlights a rare occurrence of a true broad ligament leiomyoma, the challenges associated with preoperative diagnosis, and the laparoscopic approach to its management.

1. Introduction

Leiomyomas are the most common benign tumours of the female genital tract and are present in up to 80% of women of reproductive age [1]. Leiomyomas typically originate from the clonal proliferation of smooth muscle cells of the uterine myometrium [1]. However, they can also develop in several extraterine sites such as the ovary, round ligament, broad ligament, cervix or abdominal wall [2]. While extraterine leiomyomas are extremely rare, they most commonly occur in the broad ligament, with an overall incidence of less than 1% [2].

Clinically, broad ligament leiomyomas may be asymptomatic or may present as an extraterine pelvic mass that compresses the bladder, ureter, or bowel, leading to varying degrees of chronic pelvic pain, urinary or bowel dysfunction [3]. Broad ligament leiomyomas may be detected on routine pelvic ultrasonography; however, abdominopelvic computed tomography (CT) or magnetic resonance imaging (MRI) can more accurately delineate the size, location, and number of leiomyomas and assess their spatial relationship with nearby pelvic structures [4].

Due to their rarity, broad ligament leiomyomas pose a diagnostic challenge as they are easily mistaken for benign or malignant ovarian masses, tubo-ovarian abscess, broad ligament cysts or lymphadenopathy [4]. Herein we report a rare case of a large true broad ligament leiomyoma in a 47-year-old patient with a history of advanced endometriosis and distorted pelvic anatomy. In this case, the left broad ligament myoma presented a significant diagnostic and therapeutic challenge due to its resemblance to a suspicious adnexal mass on preoperative assessment and its proximity to the ureter in a patient with a congenital solitary left kidney and left ureter.

2. Case Presentation

A 47-year-old biparous woman presented to the gynaecology clinic with chronic pelvic pain, abnormal uterine bleeding and urinary incontinence for 2 years. She described the pelvic pain as intermittent and severe, associated with the onset of menstruation and deep dyspareunia. The patient denied experiencing a vaginal discharge, symptomatic anaemia and gastrointestinal symptoms. She was diagnosed with stage III endometriosis after undergoing a diagnostic laparoscopy with left ovarian cystectomy secondary to an ovarian endometrioma 4 years earlier. She had a history of type II diabetes mellitus, hypertension, and a congenital left solitary kidney and left ureter with preserved renal function. Her past gynaecological and obstetric history was otherwise unremarkable, although she was non-compliant with medical treatment for endometriosis. The patient had no personal or familial history of

Abbreviations: CT, Computed tomography; MRI, Magnetic resonance imaging.

* Corresponding author.

E-mail address: vbahall@gmail.com (V. Bahall).

https://doi.org/10.1016/j.crwhe.2022.e00436

Received 13 July 2022; Received in revised form 1 August 2022; Accepted 3 August 2022

Available online 5 August 2022

2214-9112 © 2022 The Authors. Published by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).
malignancy.

On clinical examination, a 10-week-sized non-tender, retroverted uterus was appreciated with a healthy-appearing cervix. Laboratory investigations, including a complete blood count, and renal and liver function tests, were within normal parameters; however, a CA-125 level was mildly elevated at 53 U/ml. Pelvic ultrasonography demonstrated a retroverted uterus measuring 10.1 cm × 8.5 cm and a 3.4 cm × 3.9 cm suspicious left adnexal mass with heterogeneous solid components. There were no signs of abdominopelvic ascites, left hydroureter or hydronephrosis.

Computed tomography (CT) of the abdomen and pelvis was requested to further characterize the adnexal mass and plan treatment. Abdominopelvic CT demonstrated a heterogeneous left pelvic nodule measuring 2.6 cm suggestive of an endometriotic deposit (Fig. 1). The uterus appeared unremarkable and there was no pelvic lymphadenopathy. The right kidney and right ureter were absent while compensatory hypertrophy of the left kidney and left ureter was observed. These findings suggested a likely left endometriotic deposit. Treatment options were discussed with the patient, and she consented to a total laparoscopic hysterectomy and bilateral salpingo-oophorectomy. Pre-operative left ureteric stenting was discussed with patient. A decision was made to proceed without a pre-operative ureteric stent with the option to stent if necessary intraoperatively by the on-site urologist.

Intraoperatively, a 5 mm optical access trocar was placed at the umbilicus and two 5 mm accessory trocars were placed in the left lower quadrant. A panoramic inspection of the pelvic cavity highlighted dense adhesion bands that involved the uterine serosa, bladder peritoneum, Fallopian tubes, and anterior abdominal wall with widespread evidence of endometriotic deposits. The pelvic mass identified on preoperative imaging was in fact a 2.6 cm discrete mass suggestive of an endometriotic deposit (Fig. 1). The pelvic mass was incised using the LigaSure™ (Medtronic, USA), and the anterior leaf was dissected down to the level of the cervix (Fig. 2B). The peritoneum was stripped of the anterior aspect of the leiomyoma to its base and the myoma was carefully dissected off the left ureter. After excision, the specimen was stowed in the pouch of Douglas and the standard procedure for a total hysterectomy and bilateral salpingo-oophorectomy was completed. The specimens were removed transvaginally and the vaginal vault was closed using 2/0 delayed absorbable barbed suture. The procedure was completed without injury to the ureter, bowel or bladder in 110 min, with an estimated blood loss of 100 ml.

Histopathology later confirmed a true broad ligament leiomyoma comprising thin spindle cells in a whorled arrangement. Additionally, there was histologic confirmation of endometriosis involving the uterine serosa, cervix, Fallopian tubes and ovaries. The patient’s postoperative course was unremarkable, and she was discharged in satisfactory condition one day later.

3. Discussion

Uterine leiomyomas are the most common benign tumours of the female genital tract [1]. However, leiomyomas can also arise in smooth muscles at extraterine sites such as the ovary, round ligament, broad ligament, ovarian ligament, cervix or abdominal wall [2]. According to several epidemiological studies, the broad ligament is considered the most common site of involvement for extraterine leiomyoma, although its estimated incidence is less than 1% [2]. Broad ligament leiomyomas may be characterized as true or false based on anatomical delineation. True broad ligament leiomyomas arise from smooth muscle cells of the mesometrium and are independent of the uterus [5]. In contrast, false broad ligament leiomyomas are more common, originate from the lateral walls of the uterus or cervix, and involve the broad ligament [5].

The aetiology of true broad ligament leiomyomas remains unelucidated. In general, leiomyomas develop from the clonal proliferation of a single smooth muscle cell, most often in the estrogen-sensitive uterine myometrium [1]. Several theories have been proposed for the development of extraterine leiomyomas. These include mesenchymal stem cell metaplasia secondary to hormonal or genetic influences, parasitic implantation and growth of leiomyomas because of iatrogenic morcellation or endomyometriosis leading to smooth muscle metaplasia and hyperplasia [6]. First described by Cozzutto et al in 1981, endomyometriosis is a rare condition characterized by the presence of endometrial tissue outside the endometrial cavity that contains smooth muscle cells [7]. Like endometriosis, this endomyometriotic tissue may implant onto pelvic structures and the smooth muscle cell component undergoes subsequent hyperplasia to form a fibroid mass. [7,8] Considering our patient’s long-standing history of advanced endometriosis, endomyometriosis may be a plausible explanation for the development of the true broad ligament leiomyoma in her case.

Broad ligament leiomyomas have significant clinical and diagnostic implications. In most cases, broad ligament leiomyomas are asymptomatic and are discovered only on pelvic imaging or incidentally during surgery [3]. However, their unique location and ability to attain a large size may cause local mass effects on the ureter, bladder, or bowel to produce varying degrees of chronic pelvic pain, ureteric obstruction, urinary incontinence, constipation or bowel obstruction [3].
Furthermore, broad ligament leiomyomas are difficult to diagnose on preoperative imaging [4]. Due to its apparent adnexal location, broad ligament myomas are often mistaken for benign or malignant ovarian masses, tubo-ovarian abscesses, broad ligament cysts, lymphadenopathy, or pedunculated uterine leiomyoma on ultrasonography [4]. Pelvic MRI is associated with superior sensitivity in assessing the precise location, number, and size of leiomyomas, their spatial relationship with other pelvic structures and the uterine topography [4,9]. Although pelvic MRI may improve the positive predictive value for assessing broad ligament leiomyomias, its implementation in routine clinical practice is limited by its cost and availability [9]. Broad ligament leiomyomas may also produce an elevation of the tumour marker CA-125 [3]. However, CA-125 is a non-specific marker that is often elevated in endometriosis or serous ovarian tumours and thus may contribute to the diagnostic uncertainty [10].

The definitive management of true broad ligament leiomyomas involves surgical excision [5]. Modern advancements in minimally invasive surgical techniques and expertise have facilitated the safe and effective removal of increasingly large leiomyomas with several advantages over conventional laparotomy [11,12]. According to Hassan et al, laparoscopy is the best method for removal of broad ligament leiomyomas as patients experience a shorter duration of hospitalization, faster recovery times, better cosmetic outcomes and lower procedural costs [12,13]. The standard surgical approach involves the division of the broad ligament into its anterior and posterior leaves with careful dissection of the leiomyoma off its peritoneal attachments after proper identification and exposure of the ureter [5]. The specimen may be removed by contained morcellation within an endoscopic bag or removed transvaginally if a hysterectomy is also planned [5]. The perioperative utilization of gonadotropin-releasing hormone (GnRH) analogues such as goserelin, leuprolide or nafarelin may reduce the size and vascularity of broad ligament leiomyomas [14]. However, these agents may produce an ill-defined fibroid pseudocapsule that may obscure cleavage planes and is also limited by its cost and adverse effects [14].

Leiomyoma enucleation may be challenging due to the rich vascularity of the broad ligament and the proximity of the ureter and uterine vessels [15]. Ureteric and uterine vessel injuries and concealed haematoma are the most common complications associated with surgical management [15]. Moreover, the likelihood of iatrogenic injury is further compounded in patients with distorted pelvic anatomy from prior surgical procedures, advanced endometriosis, or radiotherapy.

According to Sizzi et al, broad ligament leiomyomas are associated with an 18.8% increased risk of complications during surgical removal and an odds ratio of 2.43 for developing procedural complications [16]. In this regard, such an operation should only be attempted by experienced gynaecological surgeons. As noted in our case, the patient's history of advanced endometriosis and a congenital solitary left kidney and ureter demanded meticulous attention to dissection to avoid debilitating injury.

In conclusion, a true broad ligament leiomyoma is a rare clinical entity that is often not diagnosed or mistaken for an alternative aetiology of a pelvic mass. Broad ligament leiomyomias are difficult to diagnose by clinical assessment and imaging alone and in most cases the diagnosis is confirmed intraoperatively. Clinicians should consider broad ligament leiomyoma as a differential diagnosis for an adnexal mass or if a uterine leiomyoma is reported as lateral. In this regard, collaboration with the radiologist should be considered when planning treatment. Broad ligament leiomyomas can be safely and effectively managed by laparoscopic surgery, particularly when performed by an experienced surgeon to confer the best outcomes.

Contributors

Vishal Bahall conceived, supervised and drafted the manuscript and performed the total laparoscopic hysterectomy and bilateral salpingo-oophorectomy with broad ligament myomectomy.

Lance De Barry drafted, edited, revised the manuscript, and performed the literature review.

Both authors revised the draft and approved the final manuscript.

Funding

No funding from an external source supported the publication of this case report.

Patient consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

Provenance and peer review

This article was not commissioned and was peer reviewed.
Acknowledgements

The authors would like to thank all anonymous reviewers and editors for their helpful suggestions for the improvement of this paper.

Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

References

[1] A. Zimmermann, D. Bernuit, C. Gerlinger, M. Schaefers, K. Geppert, Prevalence, symptoms and management of uterine fibroids: an international internet-based survey of 21,746 women, BMC Womens Health 12 (2012), 6.

[2] A.S. El-Agwany, Huge broad ligament fibroid with paracervical extension: a safe approach by same setting myomectomy before hysterectomy, J. Med. Ultrasound. 26 (1) (2018) 45–47.

[3] P. Bansal, D. Garg, A case of massive broad ligament leiomyoma imitating an ovarian tumour, J. Clin. Diagnost. Res. JCDR 8 (3) (2014) 136–137.

[4] D.K. Rajanna, V. Pandey, S. Janardhan, S.N. Datti, Broad ligament fibroid mimicking an ovarian tumor on ultrasonography and computed tomography scan, J. Clin. Imag. Sci. 3 (2013) 8.

[5] P. Pandit, S. Chandak, Laparoscopic management of broad ligament fibroids, J. Gynecol. Endosc. Surg. 2 (1) (2011) 64–66.

[6] H. Chin, X.H. Ong, P.K.L. Yam, B.S.M. Chern, Extraterine fibroids: a diagnostic challenge and a long-term battle, BMJ Case Reports. 2014 (2014), 2014: bcr2014204928.

[7] C. Cozzutto, Uterus-like mass replacing ovary: report of a new entity. (0003–9985 (Print)), Arch Pathol Lab Med, 1981.

[8] G. La Greca, C. Colarossi, P. Di Martia, C. Gorzo, M. De Zuanzi, E. Piombino, et al., Endometriometrosis of the rectum with disseminated peritoneal Leiomyometrosis 8 years after laparoscopic myomectomy: a case report, Front. Surg. 8 (2021) 666147.

[9] J.D. Oliveira, T.M. Cunha, A. Teresó, Tumors of the broad ligament: what and when to suspect such rare location, Radiol. Bras. 53 (5) (2020) 349–355.

[10] V. Bahall, L. De Barry, S.S. Harry, M. Bobb, Gross Ascites Secondary to Endometriosis: A Rare Presentation in Pre-Menopausal Women. (2168–8184 (Print)), Cureus, 2021.

[11] I.M. Kindinger, T.E. Sitchell, T.S. Misik, Broad ligament fibroids—a radiological and surgical challenge, Gynecol. Surg. 11 (1) (2014) 19–22.

[12] T.D. Theodoridis, L. Zeppiridis, G. Griimbizis, J. Bonits, Laparoscopic management of broad ligament leiomyoma, J. Minim. Invasive Gynecol. 12 (6) (2005) 469.

[13] K.M. Al-Hussaini, Al-Isa, Is it an ovarian tumor or broad ligament leiomyoma? W J. Gynecol. Women’s Health 4 (5) (2021). WJGWHM.ID.000596.: W J Gynecol Women’s Health; 2021.

[14] I. Chen, T. Motan, D. Fau-Kiddoo, D. Kiddoo, Gonadotropin-releasing hormone agonist in laparoscopic myomectomy: systematic review and meta-analysis of randomized controlled trials. (1553–4669 (Electronic)), J Minim Invasive Gynecol (2011).

[15] M. Sakanaka, K. Kohno, Y. Arai, M. Nishida, Management of Seven cases of broad ligament fibroids via laparoscopic surgery, Japanese J. Gynecol. Obstet. Endosc. 29 (1) (2013) 79–83.

[16] O. Sizzi, A. Rossetti, M. Fau-Malzoni, M. Malzoni, L. Fau-Minelli, L. Minelli, F. Fau-La Grotta, F. La Grotta, L. Fau-Soranna, L. Soranna, S. Fau-Pampanzi, et al., Italian multicenter study on complications of laparoscopic myomectomy. (1553–4650 (Print)), J Minim Invasive Gynecol (2007).