

Endometriosis infiltrating the pelvic floor muscles with histopathological correlation—A case report

Claudio P. Crispi Jr¹, Claudio P. Crispi¹, Fernanda de Paula Crispi¹, Leon Cardeman², Alice C. C. Brandão Salomao² and Marlon de Freitas Fonseca³

¹Crispi Institute of Minimally Invasive Surgery, ²Felippe Mattoso - Grupo Fleury and ³Department of Women's Health - Fernandes Figueira National Institute for Women, Children and Youth Health - Oswaldo Cruz Foundation, Rio de Janeiro, Brazil

Abstract

We report the case of a 29-year-old woman with deep infiltrating endometriosis who underwent robotic nerve-sparing surgery for resection of all visible lesions infiltrating pelvic and extrapelvic sites. Painful symptoms included severe dysmenorrhea, menstrual dyschezia and stranguria, with no improvement in response to hormonal treatment. The location on physical examination of a painful retrocervical nodule was identified by magnetic resonance imaging to be infiltrating the right parametrium/paracervix. During surgery, this nodule was recognized as an important retrocervical/rectovaginal lesion infiltrating the pelvic floor (i.e. levator ani and coccygeus), and was histopathologically confirmed as endometriosis infiltrating the skeletal pelvic floor muscles. A Pubmed search of the MEDLINE database in March (2019) found no publication reporting histopathologic confirmation of endometriosis infiltrating the pelvic floor muscles.

Key words: histopathology, laparoscopy, levator ani, pelvic pain, robotic surgery.

Introduction

Endometriosis is a prevalent condition that affects women's health-related quality of life worldwide. Dysmenorrhea, deep dyspareunia, dyschezia and dysuria are the most frequently reported symptoms, and they may occur independently or in combinations.¹ Besides pain,² infertility,³ sexual disorders⁴ and lower urinary tract dysfunction,⁵ endometriosis is also associated with mental health problems, some of which correlate with the severity and chronicity of the pelvic pain.⁶ Endometriosis is one of the principal causes of chronic pelvic pain in young women. Different medical and surgical treatments have been adopted, and in some patient both drugs and surgery are necessary.⁷ For women with symptomatic deep infiltrating endometriosis (DIE), for example, surgery is often the therapy of choice when the symptoms attributed to deep lesions do not improve with a medical management.⁸ When multiple deep lesions are identified during

diagnostic investigations, complex surgeries designed to excise multifocal lesions may be necessary and an experienced multidisciplinary team should perform the surgery because the risk of complications.

Here we report the case of a symptomatic 29-year-old woman who underwent robotic nerve-sparing surgery in a private practice setting for resection of multiple endometriotic lesions scattered in several gynecological and nongynecological sites. Histopathologic assessment concluded that deep infiltrating endometriosis has penetrated the pelvic floor skeletal muscles.

Case Report

This case report was approved by the institutional review board, the Research Ethics Committee (CAAE 88572718.5.0000.5269 IFF-FIOCRUZ). Written patient consent for the publication of this case was obtained

Received: January 29 2019.

Accepted: June 22 2019.

Correspondence: Dr Claudio P. Crispi Jr, Crispi Institute of Minimally Invasive Surgery, Avenida das Américas, 3434 - bloco 4 - Barra da Tijuca, Rio de Janeiro - RJ, 22640-102, Brazil. Email: claudin.jr@gmail.com

and is on file at our institution. To improve transparency and reporting quality, the SCARE Guideline was used as checklist.⁹

A 29-year-old nulliparous Caucasian nonsmoking lawyer was referred to our institution reporting pelvic pain due endometriosis. The patient's body mass index was 21.4 kg/m² (weight 67 kg; height 177 cm). Percentage body fat estimated by bioelectrical impedance analysis was 30.2%.

Her dysmenorrhea had begun in adolescence; from the age of 15 her symptoms were controlled with oral contraceptive pills, which she used for 12 years. When she stopped the oral contraceptive on her own for about 10 months, the menstrual cycles remained normal, but the intense dysmenorrhea returned. The intensity of specific painful pelvic symptoms was assessed using a 0–10 verbal analogue scale, the reported scores were dysmenorrhea 8, deep dyspareunia 2, acyclic pelvic pain 2, menstrual strangury 7, menstrual dyschezia 7. No obstructive bowel complaints were reported. The patient complained of moderate catamenial right lower back pain radiating to the external aspect of the right lower limb and abdominal distension. She acknowledged current lower urinary tract dysfunctions: straining to void and the sensation of incomplete emptying.

On physical examination, the uterus demonstrated minimal mobility. A very tender large retrocervical nodule that extended to the uterosacral ligaments and the right parametrium was palpable. Serum CA-125, measured after initiating dienogest 2 mg, taken once daily preoperatively for 12 weeks, was 16.7 units/mL.

Magnetic resonance imaging, which was performed in a specialized center by an experienced radiologist (A.C.C.B.S.), revealed multicompartamental endometriotic involvement. Lesions compatible with endometriosis were identified on the right ovary, in the bilateral ovarian fossa, on the bladder, round ligaments, uterosacral ligaments, vagina, retrocervical region, rectum (exerting mild stenosis), perirectal fat and right parametrium with extension to the lumbosacral trunk and piriformis muscle. Details of the pelvic region are shown in Figure 1.

During the routine evaluation for intestinal endometriosis by rigid rectosigmoidoscopy, stenosis by angulation and external traction was observed 13 cm from the anus.

Due to the important involvement of the anterior compartment, the preoperative evaluation also included a urodynamic study and cystoscopy. The main urodynamic finding was major infravesical

obstruction with a high opening pressure (62 cmH₂O). Cystoscopy identified a 3-cm lesion infiltrating the bladder mucosa in vesicouterine fold topography, which exerted retraction on the two round ligaments.

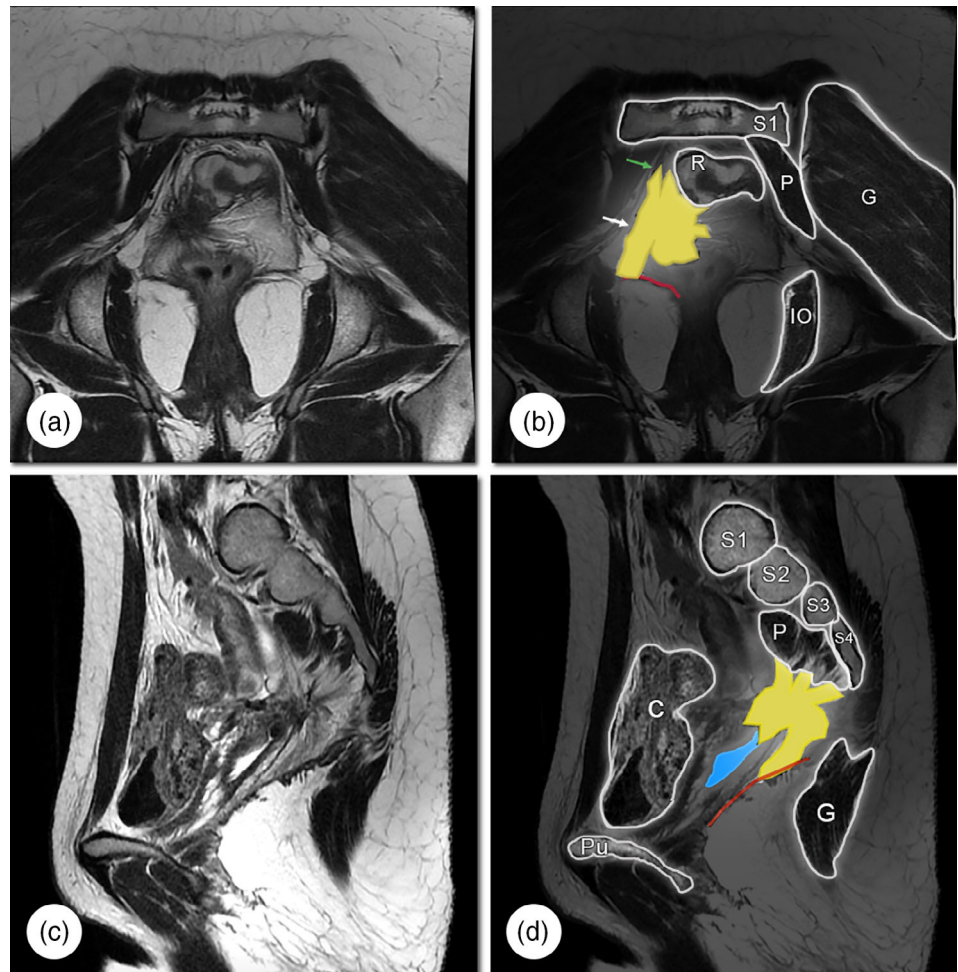
Spinal anesthesia was combined with general anesthesia to achieve transoperative hemodynamic stability and optimal postoperative analgesia. After lumbar (L3-L4) spinal anesthesia with isobaric bupivacaine and morphine, the general anesthesia was induced with propofol, alfentanil and rocuronium. Airway manipulation and placement of an orogastric tube under general anesthesia were smooth and atraumatic. The general anesthesia was then maintained with inhalatory sevoflurane associated with a continuous infusion of rocuronium and alfentanil.

The harmonic scalpel-assisted robotic cytoreductive surgery using the da Vinci Si Surgical System was performed by an experienced multidisciplinary team led by a gynecologist (C.P. C.). The peritoneal cavity was entered through direct trocar insertion under direct camera visualization, with the camera inside the trocar at insertion. In addition to the optic puncture performed on the umbilical scar, three accessory punctures were used (two robotic and one laparoscopic). The surgery was performed using pressure pneumoperitoneum of 12 mmHg. During the exploration of the abdominal cavity, all lesions previously identified by physical examination and MRI were visually confirmed. Additional typical endometriotic lesions were also identified in the right diaphragmatic dome.

The procedures performed followed a cytoreductive surgical strategy, which included fulguration of the diaphragmatic lesions; segmental rectosigmoidectomy (primary anastomosis); bilateral resection of uterosacral and round ligaments; excision of the retrocervical lesion with superior colpectomy; partial cystectomy; resection of the peritoneum of the ovarian fossa; right oophoroplasty (stripping technique); and release of the lumbosacral trunk.

For the parametrial lesions, we sought to preserve the pelvic anatomy by taking care to isolate and protect the hypogastric nerves and ureters through meticulous bilateral neurolysis and ureterolysis. The right parametrial lesion was found to be very extensive, penetrating the ipsilateral paracervix and paracolpium until reaching and infiltrating the levator ani muscle where it constitutes part of the pelvic floor (Fig. 2). Right parametrectomy with a 'shaving' resection of the affected portion of the pelvic floor musculature was performed.

Figure 1 Coronal T2-weighted image (a) and (b) AND Sagittal T2-weighted image (c) and (d). Regarding the black–white contrast in the endometriotic nodule area (highlighted in yellow in the photo on the right), the lighter the image, the more glandular the tissue and the darker the image, the more fibrous the tissue. S1, S2, S3 and S4: sacral vertebrae. P: piriformis. C: cecum. G: gluteus. Pu: pubis. S: sacrum. R: rectum. P: piriformis. IO: internal obturator. Blue: vagina. Red line: levator ani. White arrow: right inferior hypogastric plexus. Green arrow: right hypogastric nerve.



Total surgical time for this complex multicompartiment surgery – really an assortment of procedures – was 380 min (docking time of 15 min not included). The surgery was uneventful; blood loss (40 mL) was negligible. The patient remained hospitalized for 3 days; there were no postoperative complications.

Since a partial cystectomy was performed, an indwelling Foley catheter was kept in the bladder for 16 days. The postvoid residual urine volume was minimal after the Foley catheter was removed (30 mL).

The surgical specimens, including a retrocervical/rectovaginal endometriotic nodule affecting the right parametrium, paracervix, paracolpium and uterosacral ligament adhered to a small portion of the pelvic floor musculature, were evaluated by an experienced pathologist (L.C.). The presence of ectopic endometrial glandular epithelium was confirmed in all specimens. However, when the resected retrocervical/rectovaginal

endometriotic nodule (including the parametrial region) was assessed, not only were endometrial cells found in the nodule, but the presence of endometrial glands infiltrating skeletal muscle fibers of the pelvic floor was also histopathologically confirmed (Fig. 3).

Discussion

A Pubmed search of the MEDLINE database was conducted on March 1, 2019 by two of the authors (C.P. C.Jr. and M.F.F.) using the following combination of keywords: endometriosis AND ('pelvic floor' OR 'levator ani' OR ischiococcygeus OR coccygeus OR pubococcygeus). The search yielded 90 publications, which were then reviewed and read if potentially relevant. None of these publications mentioned

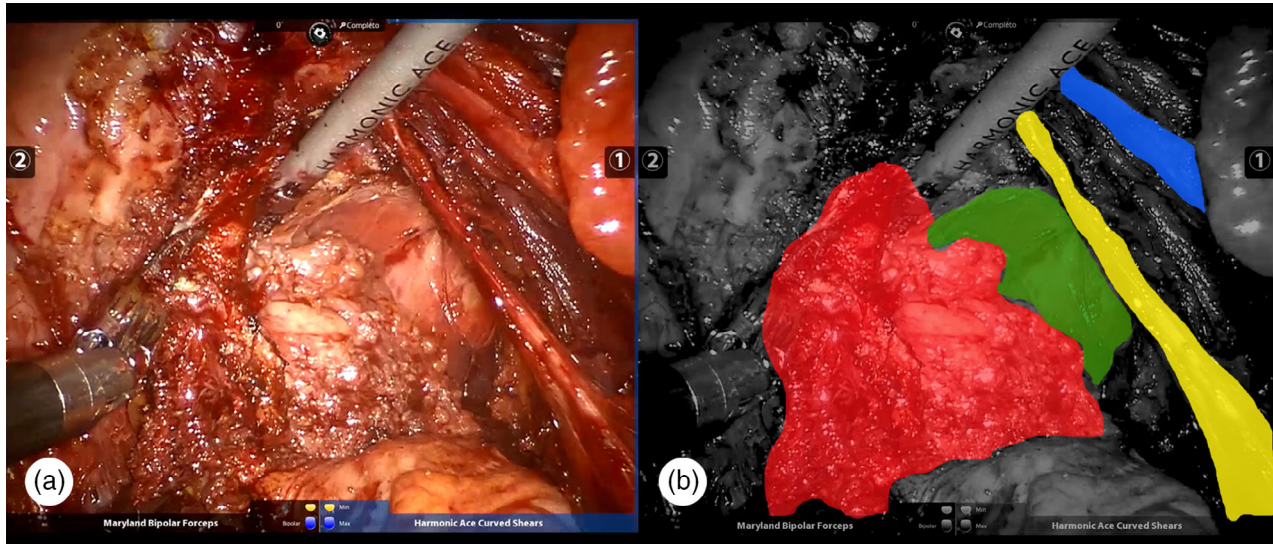


Figure 2 Robotic view of the posterior and right lateral pelvic compartment during nerve-sparing resection of a retrocervical/rectovaginal endometriotic nodule affecting the right parametrium, paracervix, paracolpium and uterosacral ligament. Anatomical reference points highlighted: right ureter (blue), right hypogastric nerve pathway (yellow), pelvic floor musculature after shaving (green), resected nodule (red).

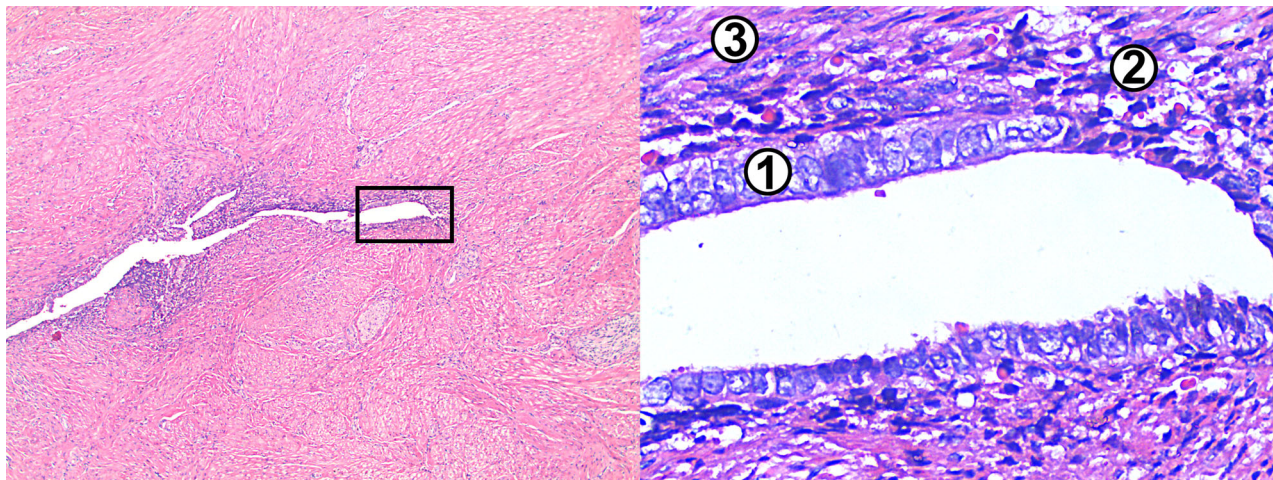


Figure 3 Microscopic view of a 2 μ section of an endometriotic nodule. The rectangle in the left (original magnification $\times 40$) was augmented in the right ($\times 100$) in order to evidence deeply located endometrial glands (i) and cytotrogen stroma (ii) surrounded by pelvic floor skeletal muscles (iii). Hematoxylin has a deep blue–purple color and stains nucleic acids whereas eosin is pink and stains proteins nonspecifically.

histopathologic confirmation of endometriosis infiltrating one or more pelvic floor muscles.

When adenomyosis (the presence of endometrial glands and stroma within the myometrium) is focal and located outside the uterus, it has been called

extrauterine adenomyoma.¹⁰ The present report, however, presents a case in which endometrial cells were found infiltrating skeletal muscles. Therefore, our hypothesis has been that an endometriosis nodule present, for example, in the retrocervical region

or in the uterosacral ligament can evolve and, by contiguity, reach deeper lateral retroperitoneal sites such as paracervix, paracolpium and pelvic floor (e.g. levator ani and/or coccygeus muscles). We think this condition, albeit uncommon, may occur in individuals with a greater predisposition to severe forms of DIE.

Surgery for deep infiltrating endometriosis is one of the most complex in gynecology. Especially when the DIE lesions involve the distal gastrointestinal and lower urinary tracts, minimally invasive multicompartiment cytoreductive procedures should be carried out by experienced multidisciplinary teams.

Minimally invasive techniques – including robotic assisted laparoscopy – has largely supplanted laparotomic surgery for cases such as these due to shorter hospital stays, better peri-operative outcomes, and reduced postoperative pain and bleeding. Moreover, laparotomic surgery also has the disadvantage of the difficulty of intraoperative identification of all foci of endometriosis, when compared to what can now be visualized with cameras during minimally invasive techniques. For all these reasons, we consider that minimally invasive techniques should generally be the surgical treatment of choice for deep infiltrating endometriosis.

Robotic-assisted laparoscopy is the most advanced technology in minimally invasive surgery and recovers the three-dimensional vision lost in laparoscopy (even knowing that there are 3D laparoscopic monitors). It offers the surgeon better ergonomics; indeed, endowrists now make it easier to work in regions of the pelvic anatomy that were extremely difficult to reach.

However, regarding key surgical outcomes in gynecology, robotic surgery has not yet been shown to be superior to conventional laparoscopy. At the present time, surgical outcomes with laparoscopy or robotics are comparable.^{11,12}

Acknowledgments

The authors are grateful to Dr Leigh J. Passman for reviewing the manuscript and Carlos Eduardo Burgos for image editing.

Disclosure

The authors declare that there is no conflict of interest regarding the publication of this article.

References

1. De Freitas Fonseca M, Sessa FV, de Carvalho Aragão L, de Resende Júnior JAD, Crispi CP. The association between dyspareunia and dysmenorrhea in women with deep endometriosis: A pre-planned observational study. *Ann Public Health Res* 2015; **2**: 1018.
2. Fonseca MF, Aragao LC, Sessa FV, Resende JAD Jr, Crispi CP. Interrelationships among endometriosis-related pain symptoms and their effects on health-related quality of life: A sectional observational study. *Obstet Gynecol Sci* 2018; **61**: 605–614.
3. Macer ML, Taylor HS. Endometriosis and infertility: A review of the pathogenesis and treatment of endometriosis-associated infertility. *Obstet Gynecol Clin North Am* 2012; **39**: 535–549.
4. Barbara G, Facchin F, Buggio L *et al.* What is known and unknown about the association between endometriosis and sexual functioning: A systematic review of the literature. *Reprod Sci* 2017; **24**: 1566–1576.
5. de Resende Júnior JAD, Crispi CP, Cardeman L, Buere RT, Fonseca MF. Urodynamic observations and lower urinary tract symptoms associated with endometriosis: A prospective cross-sectional observational study assessing women with deep infiltrating disease. *Int Urogynecol J* 2018; **29**: 1349–1358.
6. Vannuccini S, Lazzeri L, Orlandini C *et al.* Mental health, pain symptoms and systemic comorbidities in women with endometriosis: A cross-sectional study. *J Psychosom Obstet Gynaecol* 2017; **13**: 1–6.
7. Collinet P, Fritel X, Revel-Delhom C *et al.* Management of endometriosis CNGOF/HAS clinical practice guidelines short version. *J Gynecol Obstet Hum Reprod* 2018; **47**: 265–274.
8. Abrão MS, Petraglia F, Falcone T, Keckstein J, Osuga Y, Chapron C. Deep endometriosis infiltrating the recto-sigmoid: Critical factors to consider before management. *Hum Reprod Update* 2015; **21**: 329–339.
9. Agha RA, Fowler AJ, Saeta A *et al.* The SCARE Statement: Consensus-based surgical case report guidelines. *Int J Surg* 2016; **34**: 180–186 Erratum in: *Int J Surg.* 2016;36(Pt A):396. *Int J Surg.* 2017; **47**:151.
10. Paul PG, Gulati G, Shintre H, Mannur S, Paul G, Mehta S. Extrauterine adenomyoma: A review of the literature. *Eur J Obstet Gynecol Reprod Biol* 2018; **228**: 130–136.
11. Berlanda N, Frattaruolo MP, Aimi G *et al.* 'Money for nothing'. The role of robotic assisted laparoscopy for the treatment of endometriosis. *Reprod Biomed Online* 2017; **35**: 435–444.
12. Chen SH, Li ZA, Du XP. Robot-assisted versus conventional laparoscopic surgery in the treatment of advanced stage endometriosis: A meta-analysis. *Clin Exp Obstet Gynecol* 2016; **43**: 422–426.