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Oncology

Post-hysterectomy extrauterine cotyledonoid leiomyoma in a 42-year-old female

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Introduction

Cotyledonoid dissecting leiomyoma was originally described in 1975 in a case report by David et al. as a "grape-like leiomyoma of the uterus".[1] In 1996, Roth et al. coined the name "cotyledonoid dissecting leiomyoma", based on the tumor's resemblance to cotyledons of the placenta and the accompanying dissecting myometrial component.[2] To the best of our knowledge, this is the first case presented of cotyledonoid dissecting leiomyoma (CDL) occurring after a hysterectomy and originating from an extrauterine site. CDL consists of an exophytic component of smooth muscle which is rich in blood supply and has a multinodular gross appearance, suggestive of placental tissue. It typically protrudes from the lateral surface of the uterine cornu into the broad ligament and pelvic cavity. This exophytic component is in continuity with intramural dissecting components, which consist of sinuous processes of neoplastic smooth muscle extending between normal myometrial smooth muscle.[2] CDL is a benign process, as histologically there is no nuclear atypia, mitotic activity, or coagulative tumor necrosis, which are suggestive of leiomyosarcoma.[4]

Case presentation

A 42-year-old obese Caucasian female with history of thyroidectomy for papillary thyroid carcinoma underwent PET/CT screening in August 2014 (Fig. 1), and was found to have an incidental 8 × 6 × 5 cm pelvic mass. She had an extensive gynecologic history including recurrent uterine leiomyomas despite two polypectomies with D & C, and laparoscopic ablation of endometriosis and ultimately underwent a robotic total hysterectomy with bilateral salpingo-oophorectomy in 2012. Her clinical symptoms at the time of presentation were significant for mild urge incontinence and pelvic pain. She was taking transdermal estrogen as well as levothyroxine and liothyronine supplementation. Other past medical history included lupus, migraine headaches, fibromyalgia, and benign colon polyps. Family history was negative for malignancy except for breast cancer in her sister. Physical exam was unremarkable except for morbid obesity (BMI 44).

Further imaging by pelvic ultrasound and MRI revealed an irregular, lobulated mass extending from the vaginal cuff and bordering the bladder anteriorly and rectum posteriorly with significant mass effect. Gynecologic oncology performed a robotic assisted laparoscopy and cystoscopy at this time that was non-diagnostic. She underwent a repeat CT six months later which revealed a large pelvic mass. She was referred to urology for evaluation and treatment due to her persistent urinary symptoms and proximity of the mass to her bladder and ureter.

Our workup continued with a colonoscopy to rule out colonic origin of the mass as well as a CT-guided biopsy. Colonoscopy revealed a tubulovillous adenoma and a bluish discoloration of the anterior rectum with narrowing of the lumen secondary to mass effect. CT-guided biopsy showed benign fibroadipose tissue with no evidence of metastasis. The patient was advised to undergo surgical resection of the mass.

The patient first underwent cystoscopy, which revealed normal-appearing bladder. Ureters were stented bilaterally to facilitate intraoperative identification. The DaVinci Xi system was used. The left ureter was encased in a bluish-hued cystic mass (Fig. 2) that appeared to originate on top of the rectum and extend into the true pelvis. The ureter was carefully peeled away from the mass. Numerous arterial branches from the iliacs to the mass were taken. The mass was then peeled away from the bladder and rectum and the anterior vaginal wall was resected to free the mass. The mass was removed though the vaginal cuff. Rigid sigmoidoscopy showed no evidence of rectal injury. The patient was discharged on post-op day 1, and had an unremarkable postoperative recovery with only mild vaginal bleeding, abdominal pain, and constipation. The diagnosis of cotyledonoid dissecting leiomyoma was made based on the gross appearance of a placenta-like tumor along with the benign smooth muscle histology (see Fig. 3).

Discussion

Other cases of CDL/CL usually involve the surface of the uterus, broad ligament, adnexa, and rarely the bladder, rectum, and retroperitoneum.[3] In our case the tumor had grown extensively around the posterior bladder, anterior rectum, and left ureter, but without invasion of these structures. This is likely secondary to the unique location of the origin at the vaginal cuff and its proximity to the involved structures. Also, unlike any cases in the literature, this tumor had recruited blood supply from branches of the internal iliac artery, likely because it

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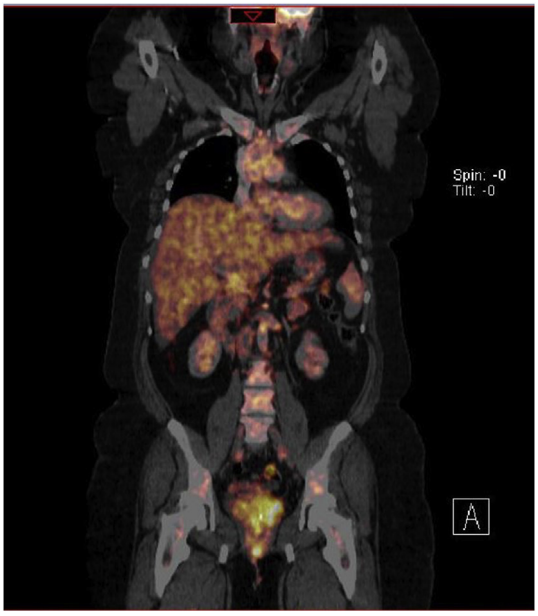

Fig. 1. Pet CT with enhancing pelvic mass.

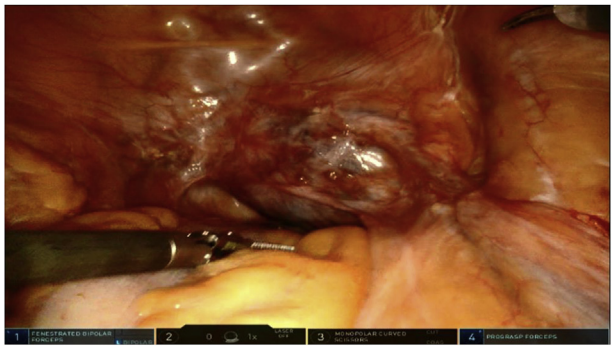

Fig. 2. Da Vinci image.

lacked the richly vascular myometrium from which CDL typically develops.[2]

Given our patient's history of multiple uterine leiomyomas including a small leiomyoma at the time of hysterectomy, and her risk factors for hyperestrogenism, we hypothesize that her vaginal cuff was seeded with leiomyoma cells. The seeding occurred either at the time of dilation and curettage or hysterectomy. Afterward, her hyperestrogenic state fostered the growth and differentiation of the tumor.

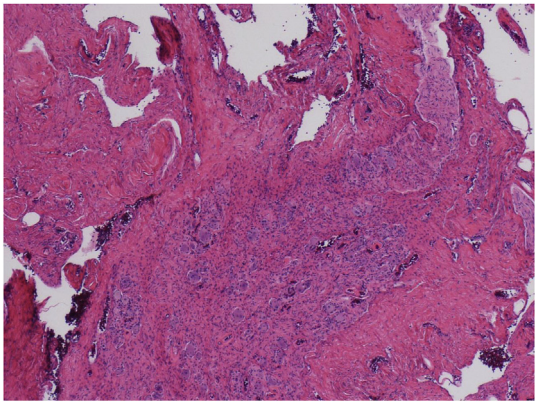

Fig. 3. Histology – H&E stain.

Conclusion

This case represents a unique occurrence of cotyledonoid leiomyoma originating from the vaginal cuff three years after hysterectomy. CDL/CL is a rare variant of leiomyoma with concerning radiologic and gross features, and is usually diagnosed based on placenta-like gross appearance in combination with benign smooth muscle histology. These are benign tumors, but can recur if not adequately excised. While these tumors are typically removed via hysterectomy by gynecologists or gynecologic oncologists, our case warranted urologic evaluation and treatment due to extensive tumor growth around the bladder and ureter. This case illustrates that even in women with prior hysterectomy, leiomyoma or its variants should be considered.

Appendix A. Supplementary data

Supplementary data related to this article can be found at http://dx.doi.org/10.1016/j.eucr.2018.03.018.

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