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CASE REPORT

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Villars nodule: An incidental finding with uterine fibroids and infertility—A case report

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Key Clinical Message

Villar's nodule is a rare presentation of endometriosis. Only a few cases report coexistence with uterine fibroids and infertility. We report an incidental discovery of a bleeding umbilical lesion confirmed as Villar's nodule in a 37-year-old woman with primary infertility who presented for myomectomy on account of leiomyomas.

KEYWORDS

case report, fibroids, infertility, umbilical endometriosis

1 | INTRODUCTION

Villars nodule or umbilical endometriosis (UE) is defined by the presence of endometrial glands and stroma in the umbilicus.[1,2] It is estimated to be prevalent in less than 1% of extra-pelvic endometriosis.[3,4] Endometrial ectopia, which characterizes endometriosis, is prevalent in 5%–20% of women in their reproductive and perimenopausal ages.[5] Some cases have also been described in postmenopausal women.[5] Aside the umbilicus, other extra-pelvic sites include the abdominal wall, appendix, intestines, lungs, and brain. Pelvic endometriosis, which is found in 15% of UE cases,[4] encompasses endometriosis in the perimetrium, fallopian tubes, ovaries, round ligament, and pouch of Douglas. The pathogenesis of endometriosis is unclear, but postulates including; direct spread, hematogenous spread, lymphatic spread, primary metaplasia, and other theories have been adduced in attempts to understand the etiogenesis.[4–8]

In the literature, only a few cases have cited the coexistence of uterine fibroids and UE.[2,9–11] Ours is one of such a few cases, and additionally, it presented together with infertility. Also, uniquely, this case was incidentally discovered and was without any catamenial symptoms typical in UE. In this case report, a Villars nodule in a 37-year-old patient with huge uterine fibroids and primary infertility is described with a review of the literature.

2 | CASE REPORT

A 37-year-old G0P0, presented 4 months ago to our facility with an 8-year history of an abdominal mass, heavy menstrual bleeding, and dysmenorrhea. She was asymptomatic of anemia and did not complain of umbilical pain or bleeding; she complained of heavy menstrual bleeding and dysmenorrhea. She had been married for 8 years and had no significant medical or surgical history aside from primary infertility. Examination revealed uterine fibroids equivalent to a 26-week gestation which was confirmed by ultrasound. She had a hemoglobin of 6.9 g/dL. She was managed for iron deficiency anemia secondary to heavy bleeding on Tot'hema (hematinic containing 50 mg of elemental iron manufactured by Innotera Chouzy) twice

daily and counseled for a myomectomy at a later date. She reported for a review 4 months later with no new complaints, and her hemoglobin level had improved to 11.9 g/dL. Her menses had begun 3 days prior to the presentation.

Abdominal examination findings were the same except for a dark umbilical nodule shown in Figure 1 and blood in the umbilicus, which she did not notice.

The umbilicus was tender. She had a repeat abdominal ultrasound that confirmed uterine fibroids but also reported a superficial hyperechoic mass measuring 1 × 1 cm and suggested a diagnosis of umbilical endometriosis.

She underwent a laparotomy and myomectomy via a midline incision. Aside from uterine fibroids and a kinked left fallopian tube, no other pelvic pathologies were seen, and abdominal inspection did not reveal any endometriotic lesions. After a standard myomectomy was carried out, the umbilicus was excised as shown in Figure 2.

The umbilical defect was closed in with subcutaneous vicryl-0 and the skin with nylon-0. The excised umbilicus was sent for histopathology and was reported as composed of islands of endometrial glands, which confirmed a diagnosis of primary UE. The post-operation period was uneventful, and she was discharged 5 days after surgery.

3 | DISCUSSION

Key to the diagnosis of endometriosis is evidence of endometrial ectopia.[1] The condition is present in one out of every five women in the reproductive age group.[4] Classifications vary, but the commonly used one pertains to the location of the lesions in relation to the pelvis.[3] Pelvic endometriosis is defined by all those lesions that affect the uterus, fallopian tubes, ovaries, pouch of Douglas, and bladder. Extra-pelvic endometriosis is found in the abdominal wall, intestines, appendix, liver, and lungs and has been reported in the brain.[12,13] UE is found in 21% of all cases of abdominal wall endometriosis, and 35% of cases of UE have pelvic endometriosis.[4] UE is considered secondary when there is previous abdominal surgery and primary or spontaneous when there is no previous abdominal surgery, irrespective of the presence of pelvic or extra-pelvic endometriosis. The presence of primary UE in the absence of abdominal endometriosis, like in the index case has been difficult to explain. Current theories to understand the development of UE include hematogenous spread, lymphatic spread, retrograde menstruation and transportation of endometriotic cells, primary metaplasia, and the congenital presence of endometrial tissue in the umbilicus. Secondary UE is well understood as spreading from the mechanical transfer of endometriotic cells during abdominal surgery.

There is contradiction with regards to the mean age of onset. Some authors report a mean age of 28 years.[3,5,6] Our patient presented at 37 years old, 10 years older than the mean age and in consonance with the reported age in other case series.[7,14] It must be noted that infertility in the index case started 8 years earlier and it may well be that UE may have been present from the onset but not diagnosed then. This may have been so despite the evidence that there may be no relationship between primary UE, uterine myomata, and infertility.[10,11] Additionally, in the present case, uterine myomata may partially account for her infertility.

The umbilical lesion or nodule usually demonstrates cyclical pain[1–7,15,16] despite the case that in this case report, there was no cyclical umbilical pain. She may have misunderstood any umbilical pain as pain arising from dysmenorrhea or degenerating uterine fibroids, which may be cyclical even though these pains will not localize to the umbilicus. It should be emphasized, however, that in 1

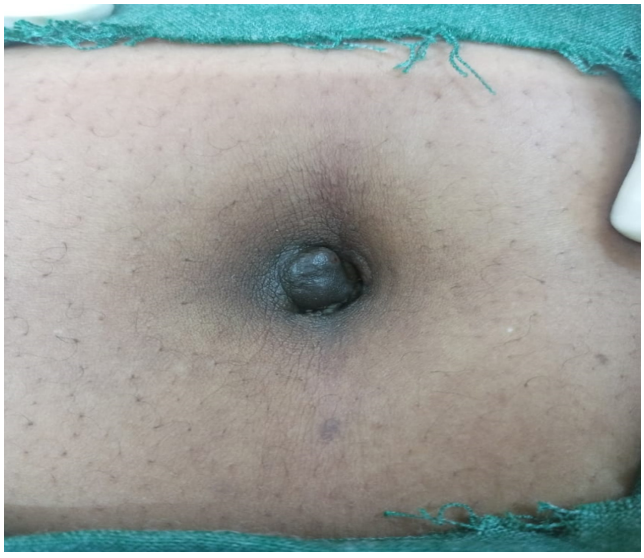

FIGURE 1 Umbilicus showing a dark nodule lesion before excision.

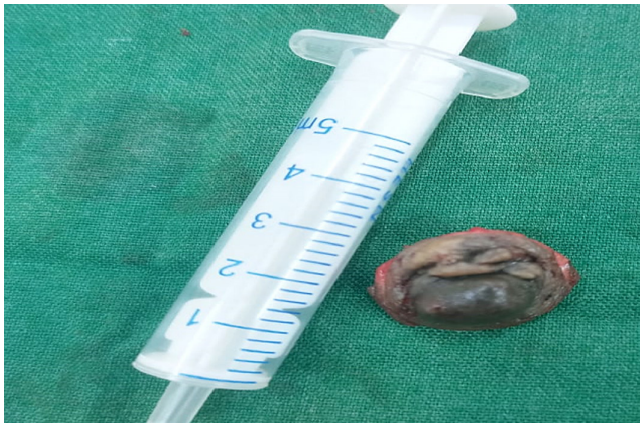

FIGURE 2 Excised umbilicus with umbilical lesion.

out of every 4 cases of UE, no pain is present[1,11,14,17] consistent with the finding in this case report. The so-called catamenial symptoms, which refer to cyclical umbilical pain and umbilical bleeding, are usually characteristic of UE. Our patient had umbilical bleeding, and that drew our attention and heightened the suspicion of UE, but this is not a universal finding. Therefore, clinicians should demonstrate a sense of critical observation during abdominal examinations so as not to miss a diagnosis. Other umbilical color changes have been described as particularly prominent during the menses.[11,18] The lesions have been variously described as dark or brown,[2,4,10,17] similar to what we encountered in this case report. They may also be bluish-purple,[1,2,4,5,11] or reddened[4] masses especially pronounced during the menses by other reports.

There is often a hard nodular swelling between 1 and 3 cm.[3,7] The index nodule measured about 1 × 1 cm, falls well within this range. These lesions may also appear as multilobulated or multinodular.[19,20] Examination findings may reveal umbilical bleeding[6,15] coinciding with the menses, similar to the observation in this report. In this case report, umbilical bleeding prompted a further detailed abdominal examination and a repeat abdominal ultrasound. Aside from the uterine myomata, the second ultrasonogram found a hyperechoic lesion and increased the suspicion of primary umbilical endometriosis. The hyperechoic description of the lesion was similarly reported by Ifeanyi et al.,[10] although isoechoic and even hypoechoic descriptions were explained to arise as a result by fibrotic changes and from the chronicity of the lesion.[16,17] Crucially, the second ultrasonogram allowed us to rule out periumbilical lymphadenopathy and any pelvic and abdominal endometriotic lesions. Additional imaging modalities such as computed tomography, magnetic resonance imaging, and PET scans could provide additional detail[3,5,8,9] but are of questionable diagnostic benefit,[1] expensive, and the patient could not afford these. Aside from imaging, fine needle biopsy, or Fine Needle Aspiration and Cytology (FNAC), and immunocytochemistry have been additional investigations used in some case reports.[1,5,18] Laparoscopy[2] and laparotomy have also been employed to exclude the presence of pelvic endometriosis as a sole investigation in some reports.

Diagnosis requires a high index of suspicion because a myriad of other lesions mimic primary UE. These include granuloma, Sister Mary Joseph nodule, umbilical hernia, umbilical polyp, melanocytic naevus, keloid, and lipoma.[3,5,8,17] The diagnosis is usually made clinically and confirmed by histopathology. The overwhelming practice regarding management entailed umbilical excision and repair.[1,3,7,8,10,15] In a few cases, nodulectomy has been the choice for management.[1] The patient had spinal anesthesia, and we elected an umbilical excision with a margin of 0.5 cm in consonance with the current practice.[3,10,15] This we considered to reduce the risks of recurrence and malignant transformation, which are small but possible.[18]

Additionally, the laparotomy, aside from allowing us to perform a myomectomy, the procedure allowed a thorough abdominal inspection that did not reveal pelvic or extra-pelvic endometriosis. Aside from Mba et al.,[8] who managed coexisting uterine fibroid with myomectomy and managed UE with umbilical excision like in this report, other cases of endometriosis and uterine fibroids did not undergo any uterine surgery.[10,11] Significantly, some other cases reported the presence of an endometrioma (chocolate cyst) in addition to uterine fibroids and UE, which was managed by only an umbilical excision without a myomectomy.[2,10] We recommend abdominal surgical exploration and umbilical excision when UE exists with uterine fibroids and infertility because of the opportunity of a myomectomy and the excision of any chocolate cysts that may be present. Laparotomy may also offer the opportunity to explore and possibly manage any pelvic and extra-pelvic endometriotic lesions.

Histopathology, which is mandatory and is the gold standard for diagnosis, in our instance, reported islands of endometrial glands and confirmed a diagnosis of Villars nodule.

Medical management used in other instances[3,5,15] was not contemplated in our case due to costs and their limited value in this very rare disorder.

Bleeding umbilical nodules in women in their reproductive ages, especially those occurring during menses, should be evaluated for UE with a histopathological confirmation. We urge clinicians to consider UE even when patients do not fully present with catamenial symptoms or present with the first episode of a bleeding umbilical nodule. A high index of suspicion is required to reduce the delay in diagnosis or misdiagnosis associated with UE.

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There was no funding for the study.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interests.

DATA AVAILABILITY STATEMENT

Data available on request from the authors

CONSENT

The authors obtained a signed informed consent for the publication of this case report.

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