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Case report

Mullerian duct failure: An unusual case with adenomyosis managed with total hysterectomy

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ABSTRACT

A 45-year-old woman admitted to our hospital complaining of perimenopausal uterine bleeding not responding to medical treatment. Ultrasound evaluation revealed unicornuate uterus with adenomyosis and it was so difficult to see the distant small left rudimentary horn on ultrasound. The patient underwent laparotomy with total hysterectomy for both horns and was sent to histopathology that indicated adenomyosis and non-communicating non-cavitated left rudimentary horn.

Defeito de ducto de Muller: caso incomum com adenomiose tratada com histerectomia total

Palavras-chave:
Deficiência de duto de Muller
Histerectomia
Adenomiose

RESUMO

Mulher, 45 anos, internada m nosso hospital com queixa de sangramento uterino perimenopáusico que não respondia ao tratamento clínico. A avaliação ultrassonográfica revelou útero unicorno com adenomiose, foi difícil visualizar na ultrassonografia o pequeno e rudimentar corno esquerdo distante. A paciente foi submetida a laparotomia com histerectomia total para ambos os cornos; em seguida, o espécime foi enviado ao patologista, que indicou adenomiose e corno esquerdo rudimentar não cavitado não comunicante.

Introduction

Unicornuate uterus is a rare congenital anomaly (2%–10% of all types of uterovaginal anomalies which occurs in 2% of population)[1] in which the partial development of one Mullerian duct results in various degrees of rudimentary horns connected or not to the other horn. According to Buttram Jr. and Gibbons classification,[2] the presence of a rudimentary cavitary horn, Non-communicating with the other one, represents the A1b variant by class II of the unicornuate uterus.

We present a case of unicornuate uterus with rudimentary horn that was successfully treated by hysterectomy for adenomyosis with persistent vaginal bleeding.

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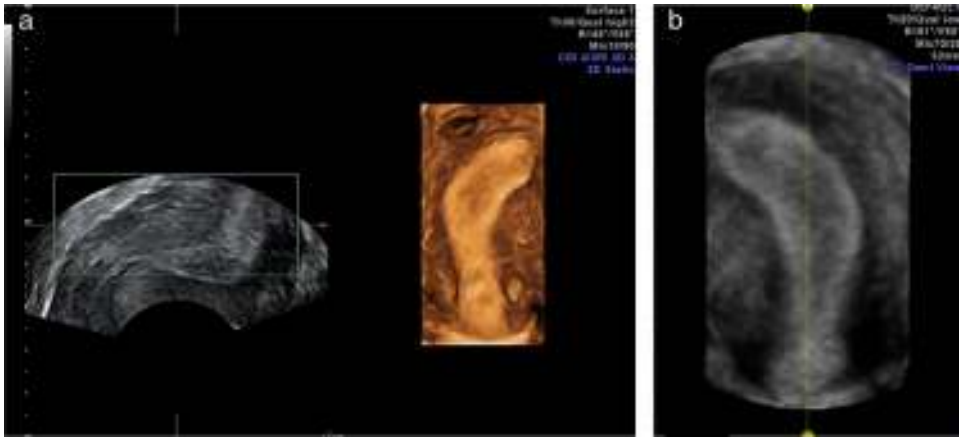

Fig. 1 – 2D and 3D ultrasound view showing unicornuate uterus.

Case presentation

A 45-year-old female referred to our department with persistent perimenopausal uterine bleeding not responding to medical treatment inform of progesterone and haemostatics since months. She had three children with history of vaginal delivery and no previous laparotomies. Ultrasound (2D and 3D views) showed a unicornuate uterus with adenomyotic features with failure to visualize the other horn with both adnexa seen, and both kidneys appeared normal (Figs. 1 and 2). The patient was not informed about that before weather on delivery or other ultrasound examinations. Under general anaesthesia, the patient underwent laparotomy for hysterectomy that revealed unicornuate uterus, but it was very unusual to see a completely separated and distant left horn, with non-communication and non-cavitated, with both adnexa attached to the both horns (Fig. 3). A rectovesical band was seen between both horns that might be the aetiology for the non-union of the 2 Mullerian ducts. Excision of this band was performed by scissors before starting the hysterectomy (Fig. 4). Hysterectomy was performed in the usual technique. Total hysterectomy with bilateral salpingo-oophorectomy was done and the specimen was sent for histopathology that confirmed adenomyosis in the right horn and non-communicating non-cavitated left rudimentary horn.

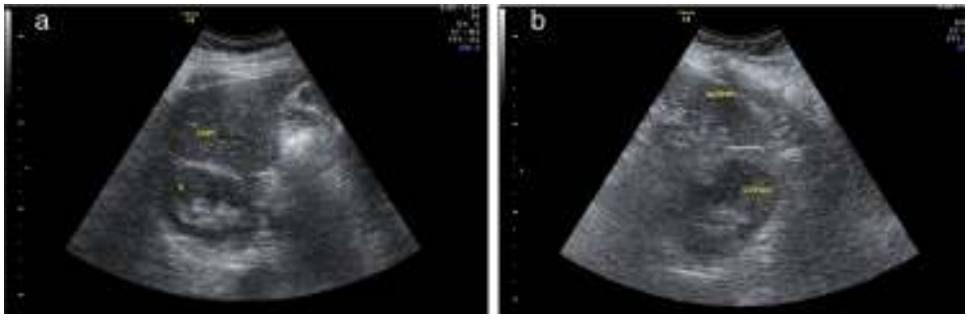

Fig. 2 – 2D ultrasound showing normal both kidneys.

Fig. 3 – Operative view showing unicornuate uterus with rudimentary left horn with adnexa attached to both horns.

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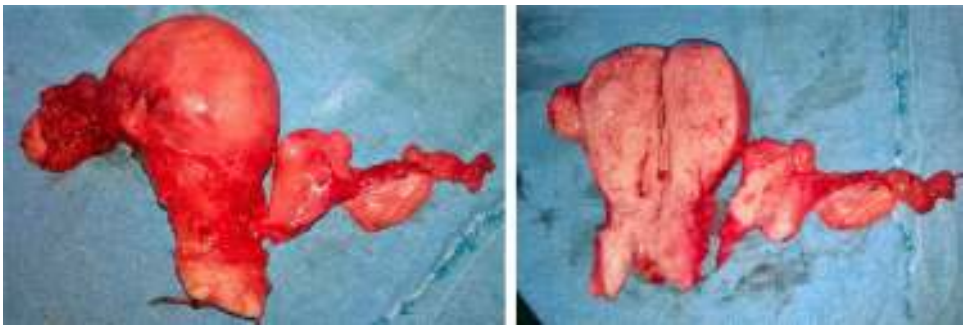

Fig. 4 – Hysterectomy specimen showing total hysterectomy with bilateral salpingo-oophorectomy with unicornuate uterus with rudimentary non communicating non cavitated left horn.

Discussion

The frequency of A1b variant by class II of the unicornuate uterus classified by Buttram Jr. and Gibbons is 7.7%–42.9% of all unicornuate uterus with a rudimentary horn.[3,4] The association with pregnancy complications is not mandatory, as shown by the obstetric history of our patient, other complications are infertility, abortion and preterm labour. Gynaecological conditions associated are dysmenorrhea (estimated in 70% of cases), hematometra (in 50%), and endometriosis (in 20%–40% of cases).[5] The diagnosis of unicornuate uterus is not so easy sometimes, and it can even be missed by inexperienced surgeons and sonographers. Proper identification and diagnosis of mullerian anomaly, however, is critical to assess the right surgical approach, because the procedure is greatly influenced by the specific subtype and by anatomical characteristics of the uterus, such as the extent of the connection between the rudimentary horn and the unicornuate uterus.[6–9]

As previously described by Fedele et al.,[6] the unicornuate uterus with rudimentary horn brings itself some intrinsic variations of the classic pelvic anatomy: frequently, the ureter lateral to the rudimentary horn has a higher course, as it lies adjacent to the vascular pedicle of the horn. For this reason, it should be mandatory to identify the ureter course firstly when the round ligament is transected and the broad ligament and retroperitoneal space are entered. Moreover, it has been suggested[3] to perform omolateral salpingectomy as the last surgical step, rather than excising it at the same time of the uterine horn.

Conclusion

In managing challenging cases of anomaly, such as the one affecting our patient, surgeon skill and decision at that point of time are very important. Operative laparoscopy, with all its advantages, and ultrasound even 3D and MRI are valid alternatives to diagnoses but the surgical procedure can be complicated by important anatomical variations.

Conflicts of interest

The author declares no conflicts of interest.

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