

**TRIPLE TORSION OF A GIANT RIGHT OVARIAN ENDOMETRIOTIC CYST IN A
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Article Received on 19/11/2022

Article Revised on 09/11/2022

Article Accepted on 29/11/2022

ABSTRACT

Ovarian endometriotic cyst is one of the most frequent localizations of endometriosis that can affect female fertility. We describe the case of a triple torsion of a giant remodeled endometriotic cyst of the right ovary measuring 15cm /12cm diagnosed in time in the emergency room of our maternity hospital by careful clinical examination, pelvic ultrasound and abdominopelvic computed tomography. An emergency surgical detorsion of the right ovary by exploratory laparotomy was carried out in the first instance, allowing immediate restoration of its vascularisation and consequently prevention of possible ischaemia. A cystectomy was carefully carried out in the second instance after detorsion, allowing preservation of the patient's fertile future.

KEYWORDS: Endometriotic cyst; endometriosis; triple torsion of ovarian cyst; surgical detorsion; cystectomy; fertility, infertility, ovarian cancer.

INTRODUCTION

Ovarian endometrioma, also known as endometriotic cyst, is defined by the invasion of the ovary by endometriosis.

It is a lesion of endometriosis that develops in contact with the ovary and causes the ovary to wrap around the lesion. This leads to the formation of an "ovarian cyst".

It is more frequent on the left side. Endometriotic cyst of the ovary is difficult to diagnose due to the fact that the symptoms are not very specific for the diagnosis and the location, and the clinical examination is often rather poor. Diagnostic certainty is only confirmed after histological analysis of a surgical biopsy or of the surgical specimen.

Its treatment is essentially surgical, and must be careful not to damage the ovary on contact and thus not to alter fertility.

The triple torsion of a large right ovarian endometriotic cyst described in our case constitutes a diagnostic and surgical emergency requiring emergency detorsion and cystectomy.

CASE REPORT

39-year-old patient, history of abortion and stillbirth, gravida 4, para 2, first pregnancy ended with a vaginal delivery of a newborn 15 years ago, second pregnancy ended with an uncured abortion at 3 months 8 years ago, third pregnancy ended with a vaginal delivery of a newborn 7 years ago, fourth pregnancy ended at 6 months with a stillbirth one year ago.

The history of the disease goes back to the day of her admission with the sudden onset of pain in the right iliac fossa, of increasing intensity with periods of calm under analgesic treatment. As the pain persisted, the patient consulted a general practitioner in Temara, 15 km from our maternity hospital, who referred her urgently to our institution on suspicion of torsion of the ovarian cyst.

Clinical examination on admission found a normotensive patient with a heart rate of 83 beats per minute, afebrile, normocolored conjunctiva, abdominal palpation found tenderness and a mass in the right iliac fossa. There was no notion of dysmenorrhea, no menorrhagia or metrorrhagia.

The patient was admitted to hospital as an emergency, with a pelvic ultrasound showing a septated right ovarian cyst measuring 14.44cm/11.34cm. An abdominopelvic computed tomography was done showing a right ovarian cystic mass with individualization of three turns of spire in favor of a right adnexal torsion on a large ovarian cyst.

A complete biological workup was performed urgently. The plasma bHCG level is negative.

An emergency exploratory laparotomy was performed. On exploration, a triple torsion of a large right ovarian cyst measuring 15cm/12cm with citrine yellow fluid content was found. The right ovary was not necrotic and its parenchyma remained normally coloured. A detorsion of the right ovary was performed as an emergency. Exploration of the left annex was unremarkable, there was no intraperitoneal effusion, the omentum was normal in appearance as was the rest of the abdominal cavity.

A cystectomy was then performed with aspiration of the cyst contents, and a sample of this fluid was taken for cytological study. On examination, the cyst was

binocular and there were no intracystic vegetations. Careful dissection and meticulous adhesiolysis of the cyst shell was performed which is removed in its entirety. The residual cyst cavity was washed with 0.9% saline serum, and bleeding vessels in the residual cavity were stitched. Two nodules found in contact with the cyst were removed.

At the end of the operation, hemostasis was perfectly ensured, and then the right ovary with the residual cavity was reintegrated into the abdomen.

The ovarian cyst shell, the cyst fluid sample and the two nodules were sent to the anatomopathology laboratory for cytological study and histological confirmation.

Anatomopathology shows a remodeled ovarian endometriotic cyst, the cyst shell wall is smooth with no endo or exophytic vegetation, and no histologic evidence of malignancy. Microscopic examination of the two nodules showed a tubal wall with fibrous and hemorrhagic remodeling and no evidence of malignancy. Cytology of the ovarian cyst contents showed a citrine yellow and macrophagic fluid, numerous macrophagic cells and neutrophils with no suspicious cells.

The postoperative course was unremarkable.



Figure 1: Right ovarian cyst, roughly oval and well limited, with regular contours and anechoic content, enclosing a septum without vascularization on Doppler, measuring 14.44 cm/11.34 cm.



Figure 2: Left ovary of normal size and morphology measuring 2.80cm /2.07cm.



Figure 3: Uterus of normal size and morphology measuring 6.51cm/4.49cm/7.5cm.



Figure 4: fine endometrium measuring 4mm.

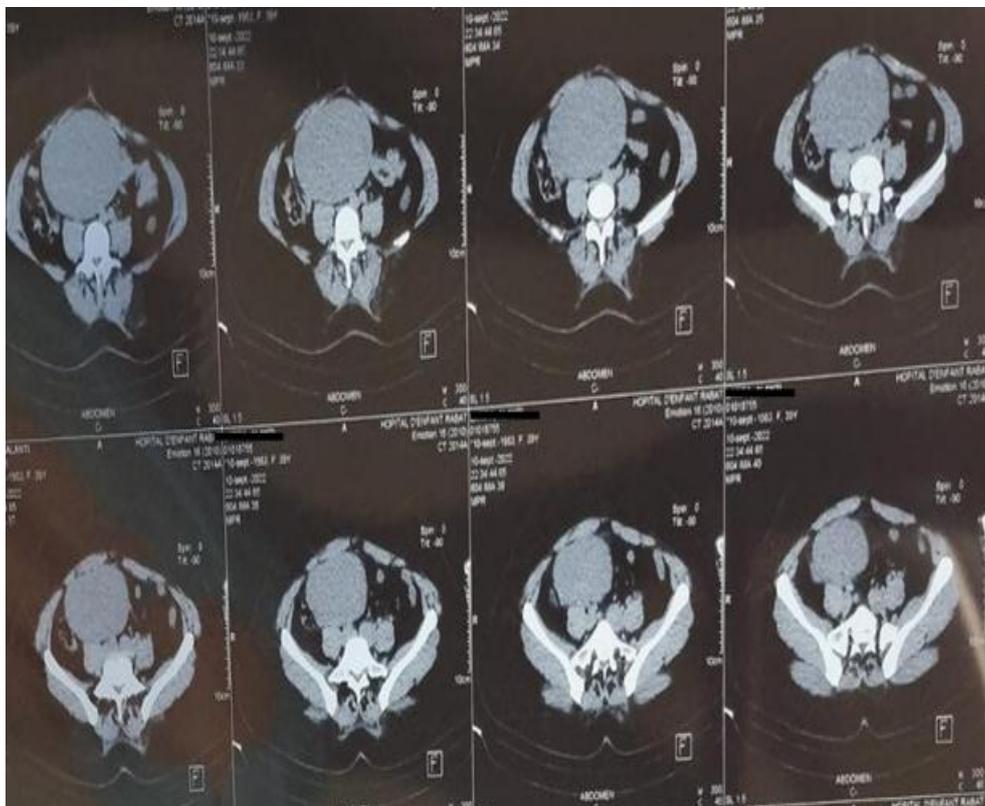


Figure 5: Abdominal and pelvic computed tomography without injection of contrast medium showing a right ovarian cystic mass with individualization of three turns of spiral in favor of a right adnexal torsion on a voluminous ovarian cyst.

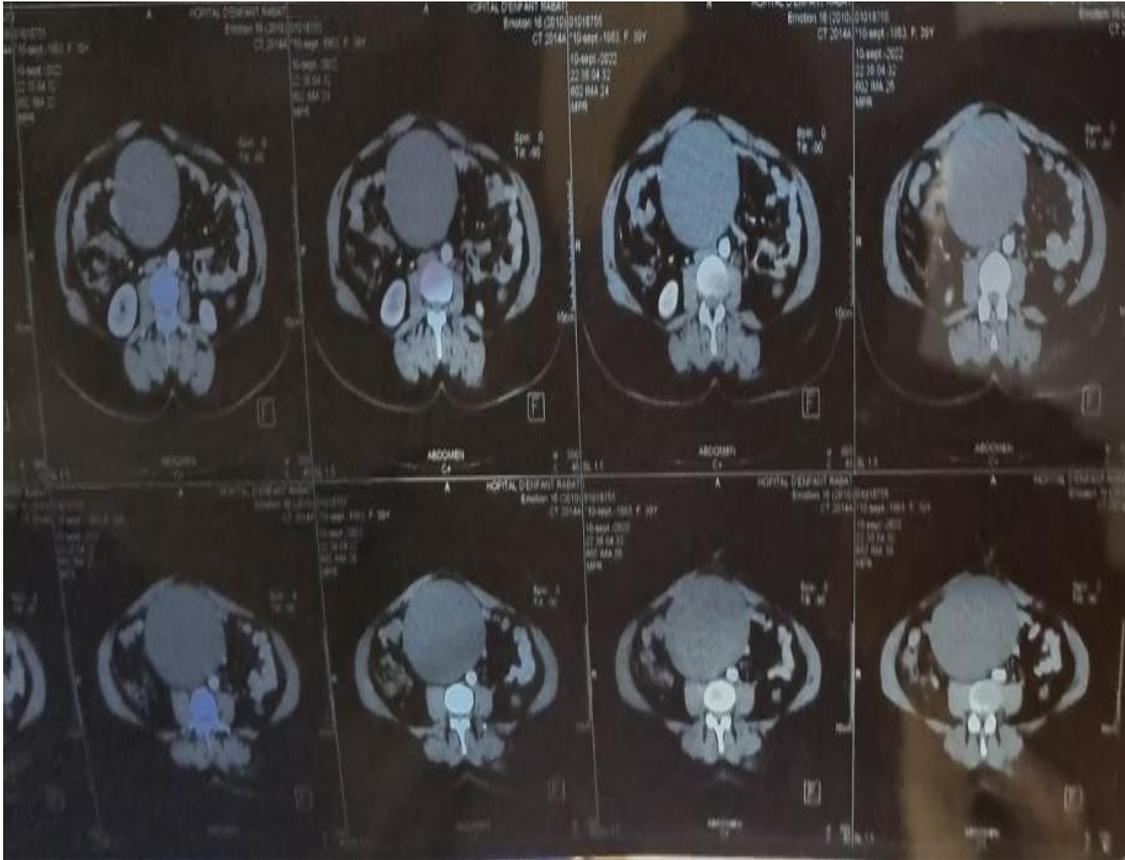


Figure 6: Abdominopelvic computed tomography with injection of contrast medium showing triple torsion of the right ovarian cyst.



Figure 7: Triple torsion of a giant right ovarian endometriotic cyst measuring 15cm /12cm after exploratory laparotomy performed in emergency.

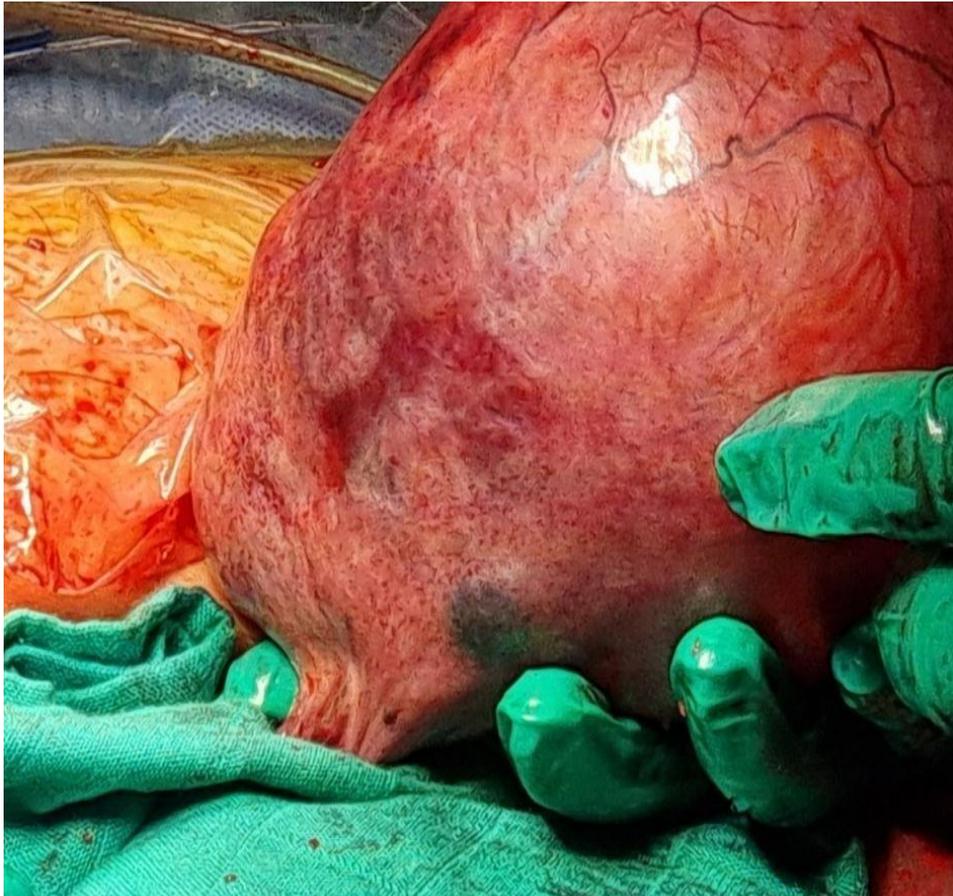


Figure 8: detorsion of the right annex.



Figure 10: Opening of the cyst wall.

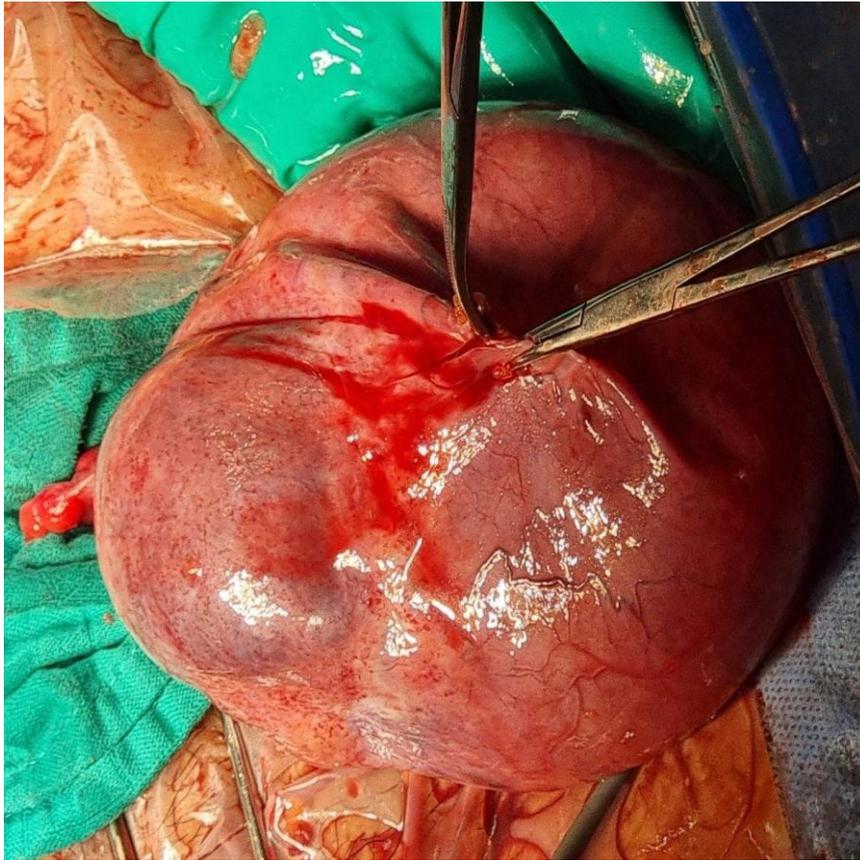


Figure 11: aspiration of the citrine yellow liquid content of the cyst, collection of the cyst liquid which is sent for cytological study. cystectomy with dissection and adhesiolysis of the cyst shell.

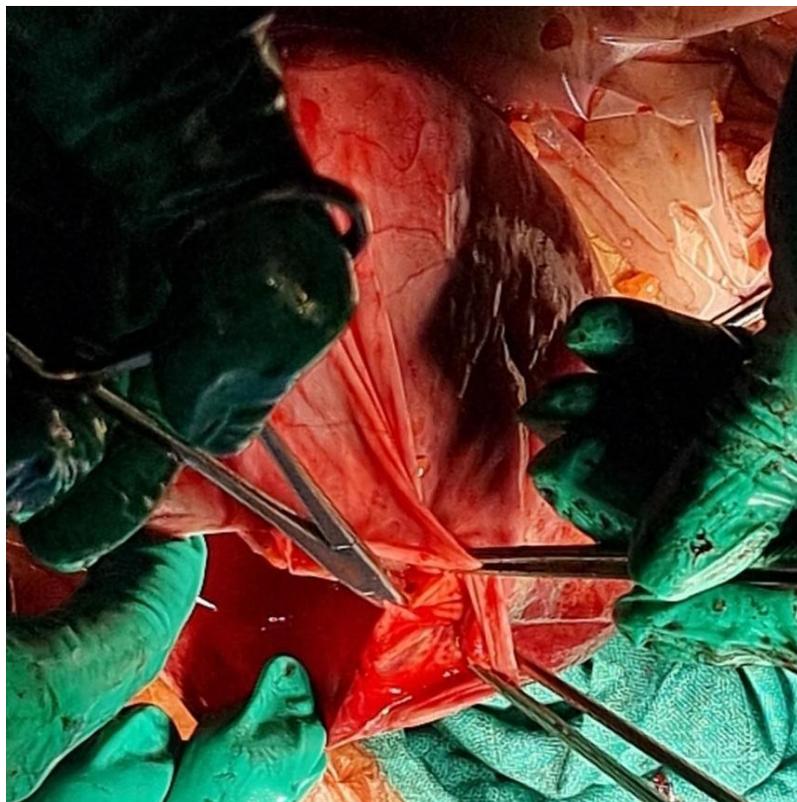


Figure 12: Binocular cyst without intracystic vegetations, complete aspiration of the cyst contents with the beginning of the detachment of the shell.

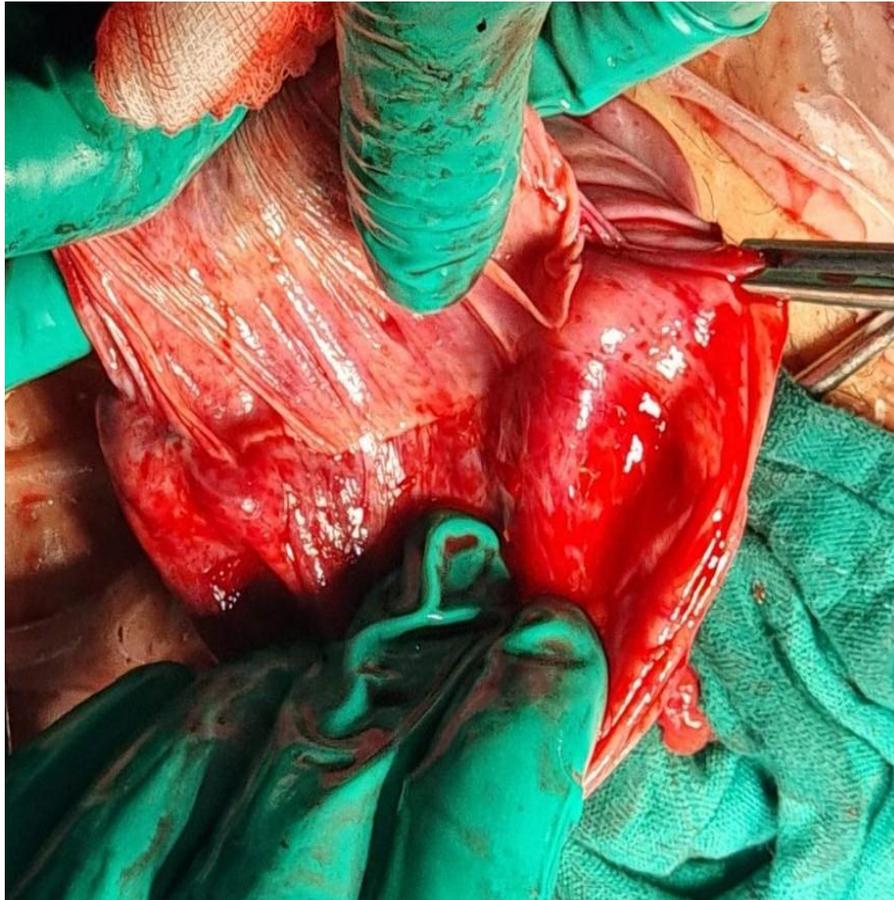


Figure 13: Adhesiolysis and detachment of the cyst shell.

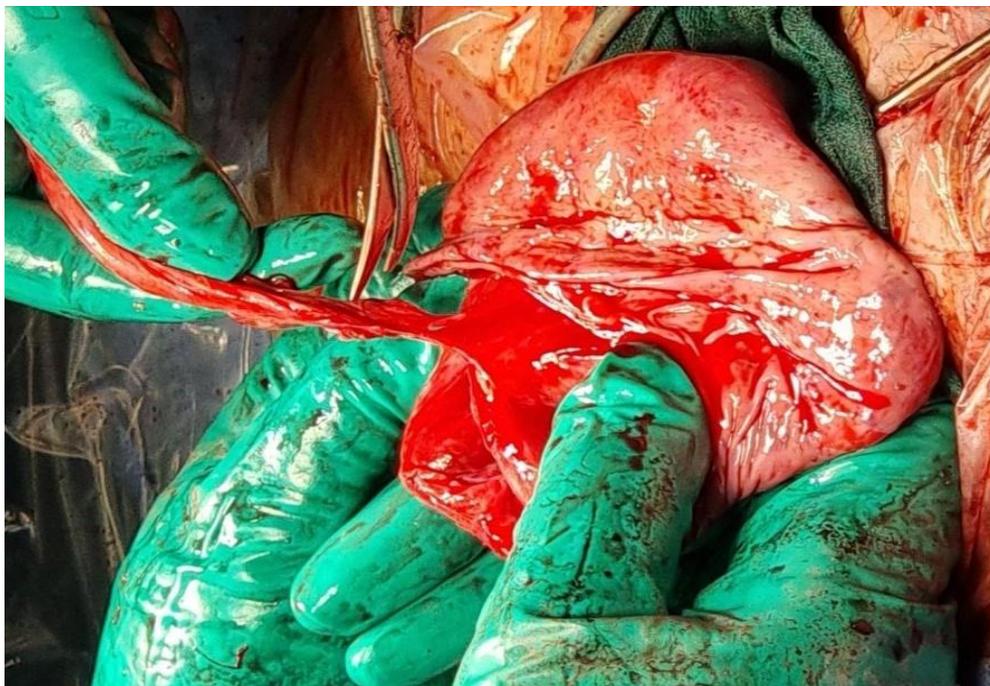


Figure 14: Complete detachment of the cyst shell and removal of two nodules incontact with the cyst.



Figure 15: Washing of the residual cyst cavity with saline serum and aspiration of the wash fluid.

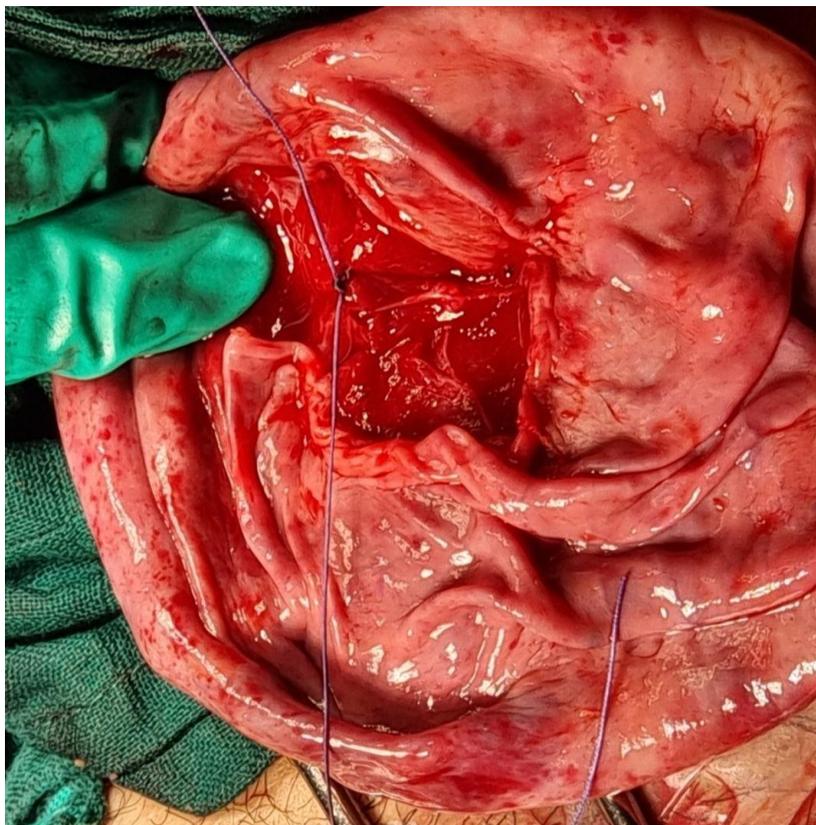


Figure 16: control of the hemostasis of the residual cyst cavity by suturing the bleeding vessels (stitches).



Figure 17: hemostasis of the residual cyst cavity is perfectly assured.

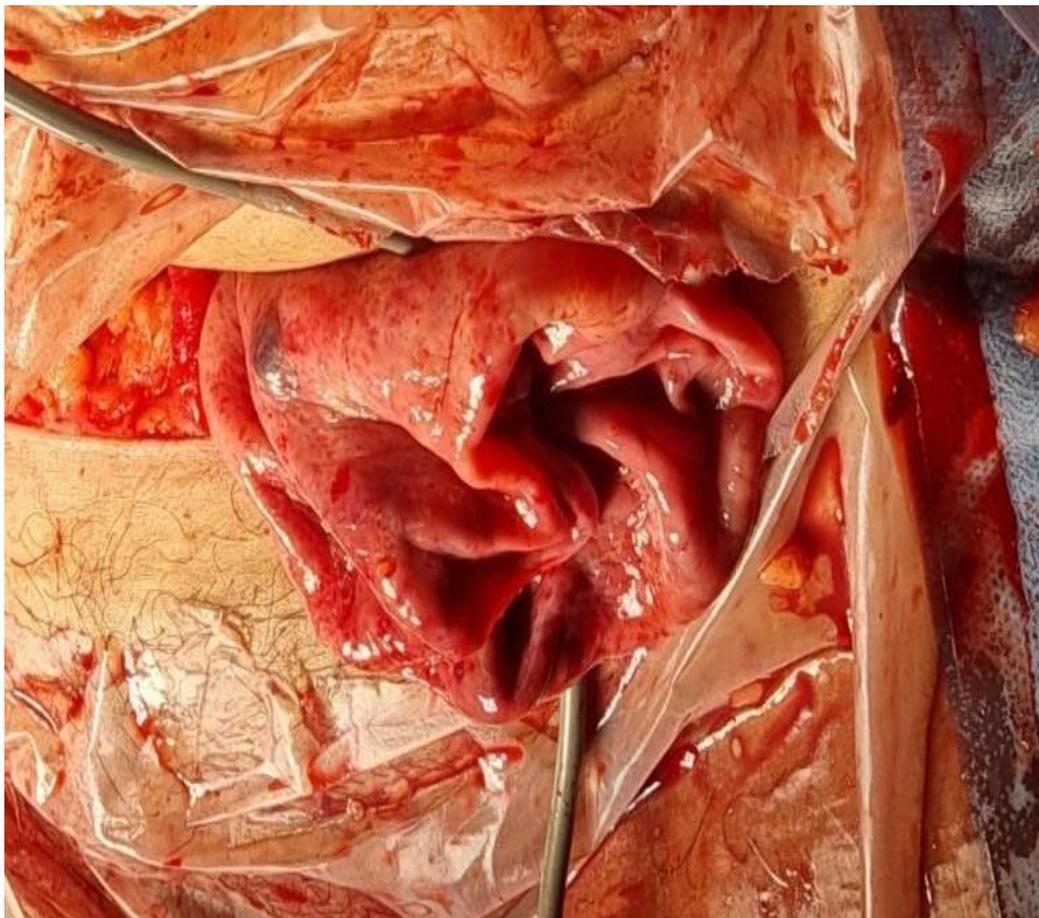


Figure 18: reintegration into the abdomen of the right ovary with the residual cyst cavity and closure of the abdominal wall.



Figure 19: The shell and the collection of the cyst content fluid, as well as the two nodules are sent to the anatomopathology laboratory for cytological and histological study.

DISCUSSION

Endometrial cyst of the ovary cause the formation of fragments of tissue resembling the uterine lining, but outside the uterus.

This tissue is as sensitive to hormones as the endometrium in the uterine cavity, so it grows like the endometrium in the first half of the cycle and then bleeds at the time of menstruation if fertilization has not occurred. As the bleeding cannot be evacuated, it stagnates and creates cysts (or other lesions caused by endometriosis), which form on the ovaries but not only.

These cysts can grow up to 15cm. They can appear alone or in groups and form on one or both ovaries.

The prevalence of endometriosis in the general population is estimated to be 10% of women in age of procreation.^[1] The proportion of ovarian involvement is 40 to 50%.^[2,3]

The pathogenesis of endometriomas remains controversial. They may arise from progressive invagination of endometriotic implants of the ovarian cortex,^[4] colonization of functional ovarian cyst by endometriotic implants,^[5,6] or metaplastic evolution of the coelomic epithelium covering the ovary.^[7,8]

The strongest physiopathological hypothesis is based on the theory of menstrual regurgitation.^[9] This theory envisages that viable endometrial fragments may be regurgitated at the time of menstruation, through the fallopian tubes, and then be discharged into the peritoneal cavity, where they will implant, grow, and sometimes invade the pelvic structures. It is possible that several phenomena intervene jointly in this

physiopathology, such as inflammation, altered immunity or else genetic factors.^[10]

The implication of the regurgitation theory in the development of endometriosis lesions is also reinforced by the fact that all factors that increase menstrual reflux, in its amplitude or frequency, are also risk factors for endometriosis, namely: early menarche, prolonged and heavy menstruation, short menstrual cycles.^[11]

Ovarian endometrioma has its own anatomical characteristics that are different from other ovarian cysts. It can be considered as a "pseudocyst" insofar as its origin seems to be extra-ovarian. In fact, ovarian endometrioma could form around a hemorrhagic peritoneal lesion to which the ovary would secondarily adhere and around which the ovarian parenchyma would duplicate.^[12] Ovulation also seems to be crucial for the development of ovarian endometriomas, in particular through hemorrhagic corpora lutea which could evolve more frequently into an ovarian endometrioma.^[13,14] These data explain why surgical removal of ovarian endometriomas may result in the removal of part of the ovarian cortex and therefore of the follicles, running the risk of altering the ovarian reserve.^[15]

Ovarian endometrioma, although it may be isolated (in 1% of cases.^[16]), is considered a marker of the severity of deep endometriosis when it is large and/or bilateral.

The endometriotic cyst affects the fertility of the patient, the ovarian cortex surrounding the endometrioma is less often of normal architecture, less well vascularized, with endometriotic implants in more than 80% of cases.^[17] Oxidative stress also seems to be more important around endometriomas, leading to an alteration of the ovarian

parenchyma.^[18] with a decrease in the number of oocytes.^[19] These data could explain the lower response, outside of surgery, to stimulation of endometriotic ovaries. The mechanisms are probably multifactorial with a direct destruction effect of the cyst on the ovary (which could be proportional to its size, the number of cysts and the presence of adhesions due to the cyst.^[20]) and an effect of the endometriosis disease.

The direct link between ovarian endometrioma and infertility is questioned.^[21,22] Spontaneous pregnancy rates of up to 43% have been reported in patients with ovarian endometrioma.^[21]

The causes of infertility associated with ovarian endometrioma are multiple. There may be an obvious role for anatomic distortions and the adhesion process associated with endometrioma lesions, particularly impacting tubal patency. However, infertility is generally considered to be the consequence of chronic inflammation.^[23] Surgical removal of endometrioma lesions may increase the chances of natural conception by decreasing pelvic inflammation.^[24]

The impact of endometrioma surgery on ovarian reserve is now well accepted and should be considered in the surgical indication.^[25,26] Endometrioma is mentioned as a risk factor, or even the precursor lesion of certain type 1 ovarian tumors: endometrioid carcinoma, clear cell carcinoma and mixed Mullerian carcinoma.^[27] This association has in fact been recognized since 1925, when Sampson described the criteria establishing the causal relationship between endometriosis and cancer.^[28,29]

This last factor is probably the most important. Indeed, if the protective effect of the estrogen-progestin pill on the risk of ovarian cancer is well demonstrated,^[30] complete removal of endometriotic lesions could reduce the risk of ovarian cancer by 70%.^[31] Enfin, association not being causation, the association between endometriosis and ovarian cancer could perfectly be explained by the fact that they share common risk factors.^[32] This is in fact the case, since endometriosis and ovarian cancer share certain risk factors (early menarche, uninterrupted ovulation and menstruation, and probably chronic or surgical stress.^[33,34]) and protective factors (bilateral tubal ligation, hysterectomy, oral contraception, physical activity).^[35]

The endometriotic cyst may be manifested by dysmenorrhea, bleeding outside the period, dyspareunia, urinary or digestive disorders. These signs may be absent, as in the case of our patient, until discovered by a complication such as torsion of the giant ovarian cyst.

Compared with peritoneal endometriosis, ovarian endometriosis is less often painful.^[36] and very rarely isolated.^[37]

Pelvic ultrasound helps in the diagnosis of endometriotic cysts. Computed tomography or magnetic resonance imaging (MRI) are sometimes used.^[38] when there is diagnostic doubt.

The CA-125 level has been described as correlated with the volume of endometriomas and the prognosis for infertile women treated surgically,^[39] however, there is no indication for this assay in the diagnosis of endometriosis. In practice, it should be carried out if there is any doubt about the nature of the cyst.

To date, there is no definitive treatment that can stop the creation of a cyst caused by endometriosis. However, hormone therapy and/or surgery (during which the cyst is punctured or extracted) can slow down the evolution of the disease for several years, depending on the woman.

Hormonal treatment prevents periods from occurring. A continuous estrogen-progestin pill, progesterone, or a hormonal intrauterine device can be prescribed to limit the pain that occurs mainly during menstruation. After 3 to 6 months, an ultrasound is suggested to see if the cysts have resolved. However, surgery should be considered if the cysts have not resolved.

The reference treatment for an endometriotic cyst of the ovary is cystectomy, which consists of adhesiolysis and careful dissection of the cyst shell.^[40] The majority of authors agree to propose a surgical procedure only for cysts larger than 3 cm.^[41,42]

Surgical treatment must be careful not to damage the ovary on contact and thus not to impair fertility.

Any surgical treatment applied to the ovary may be deleterious.^[43,44] A good knowledge of the physiopathology of endometrioma, surgical techniques and the current literature often allows a rational choice to be made.^[45,46]

Adnexal torsion requires rapid surgical management in order to allow conservative surgery. It must therefore be diagnosed as early as possible. It is easily evoked in the face of a young woman, who comes to consult because of the appearance of acute pelvic pain. This diagnosis is easily helped by pelvic ultrasound or, failing that, computed tomography or better still pelvic magnetic resonance imaging.

Our case described recalls the possible coexistence of a chronic pathology such as the giant endometriotic cyst of the ovary with an acute event of torsion of the right annex constituting a surgical emergency which consists of detorsion of the twisted annex before the ischemia and necrosis set in.

CONCLUSION

The torsion of a giant ovarian endometriotic cyst is a major gynaecological emergency which requires early

and precise diagnosis from the first clinical signs and urgent surgical detorsion to protect the ovary from ischaemia and necrosis. However, a cystectomy must be meticulously performed at the same time to better preserve the patient's fertility.

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