Journal of african clinical cases and reviews / Journal africain des cas cliniques et revues

www.jaccrafrica.com ISSN 1859-5138 Open access

Clinical case

Spontaneous bilateral ectopic pregnancy. A case report from Regional Hospital of Niamey

Grossesse extra-utérine bilatérale spontanée. A propos d'un cas au Centre Hospitalier Régional de Niamey

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Résumé

La Grossesse extra-utérine bilatérale spontanée est très rare. On estime qu'elle représente entre 1/750 à 1/580 grossesses extra-utérines et, 1/200 000 de grossesses. Cependant cette incidence est en augmentation du fait des techniques d'Assistance Médicale à la Procréation. Nous rapportons le cas d'une patiente de 28 ans à 8 semaines d'aménorrhée chez qui, le diagnostic de grossesse extra utérine rompue avec inondation péritonéale a été posé. La Laparotomie faite en urgence a permis de découvrir une GEU ampullaire droite rompue associée à une GEU gauche non rompue. Une salpingectomie bilatérale a réalisée. La patiente a reçu une poche de 450 cc de sang total en post-opératoire. Les suites ont été simples. Elle est sortie de l'hôpital à J3 post-opératoire. Nous recommandons une exploration systématique la trompe controlatérale afin d'éliminer risque d'une grossesse extra utérine bilatérale dont la méconnaissance peut être fatale.

Mots-clés : grossesse extra utérine bilatérale spontanée, salpingectomie, Centre Hospitalier Régional, Niger.

Abstract

Spontaneous bilateral ectopic pregnancy is an extremely rare event, occurring in 1/750 to 1/580 of all ectopic pregnancies, and in 1 case in 200,000 pregnancies. Nonetheless, this occurrence appears to be on the rise owing to Assisted Reproductive Technology. In this report, we present a singular case of spontaneous bilateral ectopic pregnancy in a 28-year-old patient under our care. The diagnosis was established during exploratory laparotomy, revealing an unruptured ectopic pregnancy on the left and a ruptured ampullary ectopic pregnancy on the right tub, resulting in hemoperitoneum. Bilateral salpingectomy was performed. The patient received one pack of red blood cells transfusion in postoperative period. The recovery period was simple. She left the hospital on

post-operative day 3. We recommend a systematic exploration of the contralateral fallopian tube in order to eliminate the risk of a bilateral ectopic pregnancy, which could result in a fatal outcome.

Keywords: bilateral ectopic pregnancy, salpingectomy, Regional Hospital Center, Niger.

Introduction

Ectopic pregnancy (EP) is the implantation and development of the blastocyst anywhere outside the endometrial cavity. Its incidence in developed countries has increased over the past 30 years to approximately 2% of all pregnancies in 1992 [1]. This has been attributed to the increase in sexually transmitted infections (STIs), intrauterine device (IUD) use, the rise of tubal surgery and the widespread use of reproductive technology (ART). Despite the immense improvement in detection of EP with the introduction of transvaginal ultrasound and the beta-chain human chorionic gonadotropin (beta-hCG) assay and the almost 90% decrease in associated mortality rates, EP is still responsible for approximately 9% of deaths following all teenage pregnancies in the United States [1]. While unilateral tubal pregnancy constitutes 90% of all EPs, bilateral spontaneous ectopic pregnancy is considered the rarest form [2]. Its exact frequency is difficult to estimate, approximately 1 in 200,000 pregnancies [3]. For some authors, it represents 1/750 to 1500 of all EPs [4]. Generally, there are notions of recourse to ART techniques preceding the development of bilateral secondary EP, whereas its spontaneous appearance, without induction of ovulation, is extremely rare. Hereby, we describe the management of a case of bilateral spontaneous ectopic pregnancy in order to sharpen the constant reflex of systematic exploration of the entire abdominal cavity during ultrasound examination or surgery of the EPs so as not to miss a bilateral EP, the ignorance of which can have a serious vital prognosis.

Clinical case

This was a 28-year-old patient, gravida 4, para 2, and an abortion in her third pregnancy 6 months ago. She is mother of two living children. She has no previous surgery, but has been followed by a gynecologist for pelvic inflammatory disease for 2 years She was admitted to the gynecological emergency of the Regional Hospital Center (RHC) of Niamey for pelvic pain with metrorrhagia in a context of amenorrhea of 8 weeks.

The clinical examination upon admission found a conscious but distressed patient with an unstable hemodynamic state, a BP of 103/41 mm Hg, moderately colored cutaneous, sensitivity with sharp pain on pelvic palpation. The Vaginal Touch was painful with filling of the Douglas.

Pelvic ultrasound performed urgently revealed the presence of a left ectopic pregnancy with an embryo and visible cardiac activity corresponding to the term of 7 weeks. The uterus was empty and there was fluid effusion in the peritoneal cavity.

The biological assessment revealed moderate anemia with a hemoglobin level of 9.4g/dl. The platelet count was normal (225,000 elements /mm3), PT at 83% and an A blood group positive Rhesus. The others biologicals findings were normal.

Clinical and ultrasound findings were in favor of ruptured ectopic pregnancy with maternal hemodynamic instability. So, we performed an emergency exploratory laparotomy which allowed to discover an unruptured left ampullary EP (Fig.1) as visualized on ultrasound, after aspiration of 400 ml of hemoperitoneum mixed with clots. Because of discordant pelvic effusion to unruptured EP, exploration of the contralateral tube revealed a ruptured right ampullary EP (Fig.2) with multiple pelvic adhesions and a large hematoma in the Douglas. The uterus and ovaries were normal. After careful adhesiolysis, bilateral salpingectomy was performed. After checking the hemostasis, we carried out a toilet of the abdominal cavity with isotonic saline and then we proceeded to close the abdominal wall

plane by plane. The postoperative course was simple. The patient underwent a blood transfusion with a packed red blood cell unit in postoperative. She had an uneventful early post-operative period and was discharged on day three. Pathological examination of surgical specimens confirmed the diagnosis of bilateral ectopic pregnancy (Fig. 3).

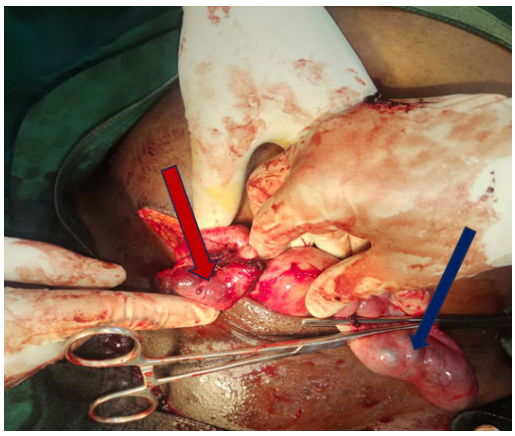

Fig 1: Intraoperative image of bilateral ectopic pregnancy. Unruptured EP located in the ampullary portion of the left uterine tube (blue arrow) and a ruptured EP located in the ampullary portion of the right uterine tube (red arrow).

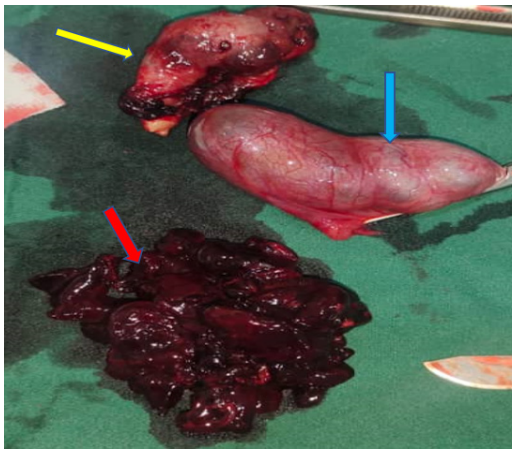

Fig 2: Surgical specimens from bilateral ectopic pregnancy: ruptured right EP (yellow arrow), unruptured left EP (blue arrow), haematoma (red arrow).

Discussion

Bilateral ectopic pregnancy is primary when it occurs spontaneously as in our case or secondary when it occurs following an IVF process. EP has well-defined risk factors that are known to cause damage to the integrity of the fallopian tubes. The most common are: history of EP, STI, pelvic/tubal surgery including bilateral tubal ligation, infertility, IUD, cesarean section and abortions requiring dilation and curettage, as well as many other equally important factors. However, in approximately 50% of cases, no risk factors can be identified [5].

The incidence of ectopic pregnancy has increased in recent years due to the development of Assisted Reproductive Technics (ART). It constitutes the most important risk factor for bilateral ectopic pregnancy [5, 6]. In fact, the overall incidence of ectopic pregnancy has increased from 0.5 to 2% previously to 4.5% today due to ART [7]. The literature reports other significant risk factors, namely: History of ectopic pregnancy, genital infections, pelvic/tubal surgery including bilateral tubal ligation, contraception using IUD and septic abortions [5]. However, in approximately 50% of the cases, no risk factors are found [5], as was the case in this observation. The etiopathogenic mechanism allows to distinguish primary bilateral ectopic pregnancy which occurs spontaneously as in the case and secondary bilateral ectopic pregnancy when it occurs following an in vitro fertilization (IVF) process. Other more specific factors are involved in its pathogenesis, notably ovulation inducers that lead to polyovulation in addition to the technique and the number of embryos transferred [5,6].

Clinically, the difference between the symptoms of ruptured unilateral EP and ruptured bilateral EP is not obvious. Clinical symptoms such as amenorrhea, metrorrhagia, pelvic pain cannot reliably differentiate them, as in this case.

On a biological level, there is no loss of beta-HCG levels that allows discrimination between ongoing intrauterine twin pregnancy, heterotopic pregnancy, single or bilateral ectopic pregnancy. In several cases, beta-hCG levels were lower even for a single EP [5]. We should therefore not rely solely on the beta-hCG level to reach the diagnosis of bilateral EP.

Ultrasound very rarely allows visualization of bilateral EP. Early ultrasound, better endovaginal, is essential for the diagnosis of ectopic pregnancy. However,

ultrasound very rarely allows for visualization of a bilateral ectopic pregnancy as is the case in this observation. In a specific situation where the radiologist identifies an intact ectopic gestational sac with cardiac activity of the embryo, the concomitant presence of hemoperitoneum and a state of maternal shock should suggest multiple localizations of the ectopic pregnancy. This is why the ultrasound examination must explore the entire abdominal cavity looking for other anomalies even when an ectopic pregnancy has already been demonstrated in one of the uterine tubes. In our context, this additional investigation was not carried out after demonstration of the left ectopic pregnancy with cardiac activity. Furthermore, we did not question the lack of concordance between the clinical signs presented by the patient and the unruptured ectopic pregnancy shown on ultrasound.

The diagnosis of bilateral ectopic pregnancy was an intraoperative discovery, as for most authors [1,2,4,6]. Therapeutic management does not differ in any way from a simple unilateral EP. The choice of method generally depends, as for unilateral cases of EP, on the hemodynamic state of the patient, the extent of tubal damage and the desire for future fertility, the size, location and level of beta-hCG [6]. Likewise, the treatment options for cases of spontaneous bilateral GEU are basically those for unilateral EP.

The choice of surgical treatment differs between spontaneous EP and that secondary to ART.

In bilateral EP, there is a tendency to perform bilateral salpingectomy in patients who have used ART even when the contralateral tube appears healthy. On the other hand, in spontaneous bilateral EP, the tendency is to salpingotomy if the contralateral tube appears healthy [8]. Two similar cases were found in the literature with a history of bilateral EP treated by salpingectomy on one tube and salpingotomy on the second, and who would have had normal subsequent pregnancies [9]. As bilateral EP remains in the vast majority of cases an intraoperative diagnosis, bilateral salpingectomy could prevent the future development of bilateral EP when the patient is intended to resort to ART, which itself is known to increase the risks of Secondary bilateral EP. In contrast, for spontaneous bilateral EP, the general principles of management involve conservative surgery whenever possible [10]. When the patient's hemodynamic state is stable with an unruptured bilateral EP, medical treatment with methotrexate could also be instituted. In 2001, Mock et al reported successful use of ultrasound-guided methotrexate injection in the management of bilateral EP secondary to ART [11].

Laparoscopic surgical treatment still remains the gold standard, because the patient recovers more quickly and the subsequent rates of intrauterine pregnancy and EP are similar [12].

The lack of exploration of the contralateral tube, the ovaries and the entire abdominal cavity, even in the presence of dense adhesions, leads to failure to diagnose bilateral EP during surgical intervention in several cases. Which brings these same patients back a few days later in a picture with worsening symptoms, increased beta-hCG levels and sometimes with an acute abdomen due to a rupture of the EP in the unexplored contralateral tube. [13].

The aftermath of EP treatment can negatively affect a woman's subsequent fertility or even worsen pre-existing infertility [14] and bilateral EP is no exception. The recurrence rate of unilateral EP is estimated at 6 to 16%, while recurrence after bilateral EP is difficult to quantify due to the rarity of this pathology, but it is nevertheless significantly increased [15].

The type of surgery, laparotomy or laparoscopy, salpingectomy versus salpingotomy, has no influence on subsequent fertility success. Sommer et al. in 2002 reported that most cases of spontaneous bilateral EP were treated by bilateral salpingectomy thus compromising the prospect of a spontaneous pregnancy [16]. In fact, salpingectomy is usually performed after a thorough assessment of the condition of the contralateral tube, the future fertility plan and the hemodynamic state of the patient.

A full-term pregnancy would have been achieved in a case of spontaneous bilateral EP treated by salpingectomy on one tube and salpingotomy on the other [8]. A similar case was also reported by

Mathelier [9]. Thus, to improve the management of bilateral EP, it is extremely important to inspect the entire abdominal cavity, including the contralateral tube during surgery for unilateral EP, even if this required significant adhesiolysis.

Tubal surgery has long been considered an important risk factor in the occurrence of EP [17]. Likewise, it has also been demonstrated that simple tubal ligation-section using the Pomeroy technique could be complicated by the subsequent occurrence of secondary bilateral EP [18]. This method of sterilization has long been criticized by Levy et al in 1988 [19] after reporting the first case of bilateral EP as a result of this technique. This is why radical salpingectomy is preferable to partial salpingectomy for the surgical treatment of EP or ligation in the context of definitive contraception because tubal remnants present a risk of recurrent EP [20].

Conclusion

Early diagnosis of spontaneous bilateral GEU is crucial to minimize its devastating complications. This requires the education of all women with or without risk factors in pregnancy planning for the purposes of early diagnosis. Furthermore, during early obstetric ultrasound, meticulous exploration of the entire abdominal cavity must be carried out systematically to exclude the possibility of a heterotopic pregnancy or bilateral ectopic pregnancy. This rigor also applies to ectopic pregnancy surgery in order to avoid ignoring a multiple location leading in the best cases to a repeat operation. In addition, complete radical salpingectomy should be performed in cases of bilateral ectopic pregnancy or during tubal ligation as part of definitive contraception.

Acknowledgements

We thank the administration of Regional Hospital of Niamey for the care of the patient. We would also like to express our gratitude to the patient for her cooperation and commitment to contributing to science.

Authors' contributions

OM, YM and AIA managed the patient and wrote the first draft.

SDH, GM, IZ, MCY, RMG, NM helped in editing and reviewing the paper.

All authors read and approved the final version to be published.

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Available online : July 10, 2024

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Conflicts of interest : None

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To cite this article :

M Oumara, M Yahaya, A Issa Abdou, H Soumana Diaouga, M Garba, Z Ide et al. Spontaneous bilateral ectopic pregnancy. A case report from Regional Hospital of Niamey. Jaccr Africa 2024; 8(3): 50-55