

## Is it Time to Abandon Spiegelberg's Diagnostic Criteria and Radical Surgical Management of Ovarian Ectopic Pregnancies?

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### Abstract

Ovarian ectopic pregnancies are rare, but some reports suggested an increasing incidence relative to both tubal and term pregnancies. Moreover, the incidence was also higher when alternative criteria to those set by Spiegelberg in 1878 were used. Women with endometriosis, pelvic inflammatory disease and those using intrauterine contraceptive devices or had assisted reproduction treatment may be more at risk. Presurgical diagnosis can be difficult though early intervention is necessary as early rupture may occur in most cases. Accordingly, medical treatment might not be a viable option. The classical treatment is usually surgical either radical or conservative with total oophorectomy or wedge resection respectively. Both modalities may be excessive especially in women during their late reproductive years and those with low ovarian reserve because of other reasons. There is a tendency now to use more conservative techniques to preserve ovarian tissues. In this manuscript I will present two cases of ovarian ectopic pregnancies, one ruptured and one intact. They were both treated successfully laparoscopically with strong suction, saline jetting and curettage of the ectopic sacs. This was followed by bipolar electrocoagulation of the bleeding residual trophoblastic tissue. No ovarian tissue was removed, and the patients resumed menstruating after 4 and 6 weeks respectively. A larger series will be needed to test the technique.

**Keywords:** Ovarian ectopic pregnancy; Conservative treatment; Laparoscopic management

### Introduction

The first case of primary ovarian ectopic pregnancy was published by St. Maurice in 1689 [1]. Inevitably, the diagnosis was made postmortem. These are rare entities in reality but may also be underdiagnosed due to the 4 diagnostic criteria set by Spiegelberg in 1878. The reported incidence after natural conception ranges from 1 in 2000 to 1 in 60 000 deliveries and accounts for 3% of all ectopic pregnancies [2]. However, there are reports of an increasing incidence relative to both tubal and term deliveries [3,4]. The incidence may even be 1 in 1400 deliveries if Spiegelberg's criteria were not used. In fact, 13 patients with proven ectopic pregnancies did not satisfy these criteria, including absent ovarian tissues during histopathological assessment [5]. The new criteria used included serum beta hCG ( $\beta$ -hCG) level  $\geq 1000$  IU/l, empty uterus during transvaginal ultrasound examination, confirmed ovarian involvement during surgical exploration, with bleeding, visualisation of chorionic villi or presence of an atypical cyst on the ovary, normal tubes and decline in serum  $\beta$ hCG level after treatment of the ovary.

Different classifications of ovarian ectopic pregnancies have been reported in the literature. However, in essence and for practical reasons, two types may be described, namely intra- and extra-ovarian. These may follow fertilization of the ovum within the follicle or fertilization immediately after ovulation that implants on the ovarian surface respectively. A plane of division may be found between the ectopic pregnancy and the ovary in the second type [6]. This may facilitate its excision, unlike the intraovarian type which may need wedge resection or even oophorectomy [6]. Surface implantation may also follow reflux of the embryo out of the fallopian tube. However, many authors questioned the clinical validity and therapeutic significance of any sort of ovarian ectopic pregnancy classification [7].

Most ovarian ectopic pregnancies tend to rupture early during the first trimester and are usually diagnosed as corpus luteal haemorrhages [8,9]. Patients may present in collapsed state hence medical treatment may not be feasible and surgery would be the only option. Accordingly, early intervention is indicated but diagnosis might be difficult at times. As for management, surgical excision of the whole ovary or wedge

resection may not be the best lines of treatment to pursue. This is especially so in women with low ovarian reserve because of previous ovarian surgery and those who are keen to conceive during their late reproductive years. Accordingly, surgical treatment of intraovarian ectopic pregnancies sets a challenge as far as ovarian reserve is concerned. The importance of conservative laparoscopic management has been advocated many years ago on the assumption that implantation was usually superficial [10]. To reduce ovarian tissues damage, one author [6] reported using monopolar scissors to extend the ectopic pregnancy opening laterally by approximately 2 cm and removed the products of conception with an endocatch bag. This was followed by cauterization of the bleeding areas. However, extensive use of monopolar energy may have a detrimental effect on the remaining ovarian tissues. In general, the main concern regarding conservative management is the persistence of trophoblastic tissues which may need future surgical intervention, or medical treatment with methotrexate. On the other hand, drastic surgical treatment may compromise future reproductive and endocrine ovarian functions.

In this article I am presenting two cases of ovarian ectopic pregnancies who had different presentations as the basis for this review. The first one had severe internal haemorrhage and was treated as an emergency, and the second was treated electively, both laparoscopically.

There was no conflict of interest or any financial issues to declare. Both patients signed consent forms allowing the use of their non-identifying data for teaching and research. No ethical clearance was deemed necessary.

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## Case Report

### Case 1

The first patient was a 25-years-old para 0+0 female. She presented to the casualty department at night with abdominal pain, slight vaginal bleeding and signs of internal haemorrhage. She gave history of 5 weeks and 5 days amenorrhoea with a positive  $\beta$ hCG level of 2490.7 mIU/ml tested earlier. Departmental abdominal ultrasound scan examination showed an empty uterus with a heterogenous mass in the pelvis and moderate amount of intraperitoneal fluid. The picture was suggestive of left adnexal disturbed pregnancy. She refused to have transvaginal ultrasound scan examination in the first place.

Emergency laparoscopic examination was performed immediately. The pelvis and lower abdomen were full of blood and all pelvic organs were not initially visible. One and a half litres of clotted blood were removed. Fresh blood was seen seeping out of an ectopic pregnancy in the left ovary over the surrounding clotted blood (Figure 1). The site of the ectopic sac was clearly seen in the left ovary after blood evacuation from the pelvis (Figure 2) and the normal left tube (Figure 3) and normal right tube and ovary (Figure 4) were seen respectively. (Figure 5) on the other hand, depicts the cauterised site of the ectopic pregnancy after strong suction, saline jetting and sharp curettage of the raw area. No ovarian tissue was actively removed. Histopathological assessment of the curetted necrotic tissue removed confirmed the presence of trophoblastic tissues, with no ovarian tissues seen.

The patient was kept in hospital overnight and received 3 units of blood. Her  $\beta$ hCG level fell to 1022.8 mIU/ml one day after surgery, and to 147.8 mIU/ml two weeks later. She started menstruating exactly 4 weeks after surgery and conceived naturally few months later. This proved to be a normal intrauterine pregnancy. The 4-weeks recovery period for this conservative treatment corresponded to that reported after surgical excision techniques in the literature [11].

### Case 2

The second patient was 32-years-old and a regular attendant of the clinic. She had laparoscopic left ovarian chocolate cyst removed in the past leaving behind a small ovary. She also had a low antimullerian hormone level during previous investigations. On this occasion, she presented with vague lower abdominal pains, which were sharp at times and dark vaginal bleeding. She gave history of 6 weeks and 3 days amenorrhoea. Her  $\beta$ hCG level was 9699.8 mIU/ml. Transvaginal ultrasound scan examination showed an empty uterus. The endometrium was 13.5 mm thick. The left ovary was small (3 ml volume). A mass with rich vascular markings was seen medial to the right ovary (Figure 6). Initially, it was thought to be a tubal ectopic pregnancy. However, 3D examination showed the mass to be part, or tightly adherent to the right ovary with a sac-like structure in the middle (Figure 7). Ovarian pregnancies usually appear on or within the ovary as a cyst with a wide echogenic outside ring, but yolk sacs and embryos are less commonly seen [12].

Because of the high  $\beta$ hCG level, the large size of the ectopic sac, endometrium  $\geq 12$  mm thick, and the patient's wishes, laparoscopic surgery was done. As for case 1, strong suction, saline jetting and rough curettage of the ovarian ectopic sac were used to preserve ovarian reserve. Suspected remaining trophoblastic tissue was coagulated with bipolar energy. This was discussed and agreed with the patient preoperatively with the risks involved explained. Not to remove any part of her ovary and to preserve what was left of her ovarian reserve was very important to her.



**Figure 1:** Laparoscopic view of the pelvis full of blood with fresh blood seeping off the left ectopic pregnancy on top of the clotted blood. The fimbrial end of the left tube was seen underneath the left ovary. This view was possible after partial evacuation of the blood in case number 1.



**Figure 2:** Shows clearly the left ovarian ectopic pregnancy sac with clotted blood covering the orifice in case number 1.



**Figure 3:** Shows a healthy looking left fallopian tube in case number 1.



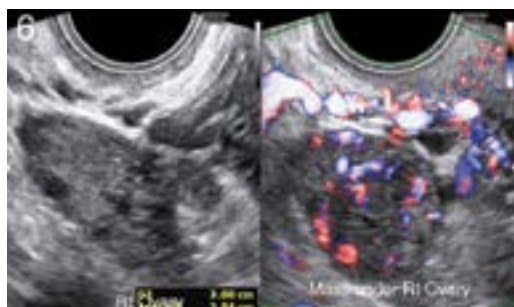
**Figure 4:** Shows normal looking right ovary and tube in case number 1.



**Figure 5:** Shows the site of the left ovary ectopic pregnancy sac after suction, curettage, saline jetting and electrocoagulation with bipolar energy in case number 1.



**Figure 9:** Shows the gap left behind on the underside of the right ovary after removing the ectopic sac in case number 2.



**Figure 6:** Shows the right ovary and a vascular mass initially seen on the underside of the ovary in case number 2.



**Figure 10:** Shows the right ovary after removing the ectopic pregnancy sac and controlling the bleeding with bipolar energy in case number 2.



**Figure 7:** Shows 3D image of the right ovary with the ectopic sac in case number 2.



**Figure 8:** Shows the right ovary with ectopic sac being exposed in case number 2.

Figure 8 depicts the ectopic sac being exposed. At the time of surgery, ovarian pregnancies frequently resemble haemorrhagic cysts [13]. Figure 9 shows the right ovary after suction and curettage. The sac was clearly seen on the medial/underside of the ovary. Figure 10 shows the same ovary after removing the sac and controlling bleeding. Signs of electrocoagulation are clearly evident.

The patient had uneventful recovery and was discharged from hospital on the same day. She was followed by measuring  $\beta$ -hCG levels in the outpatient clinic. It went up to 10893 mIU/ml one day after surgery, before falling to 1075.3 mIU/ml one week later. The level fell further to 76.95 mIU/ml one month after surgery and she started menstruating two weeks later. No methotrexate medication was used. The recovery time was two weeks longer than in the first ruptured case. This might reflect the higher presurgical  $\beta$ hCG level and slightly longer period of amenorrhoea in this case. It might also reflect that most of the trophoblastic tissues had been expelled when the sac ruptured into the abdominal cavity, reducing further the trophoblastic load, in the first case.

## Discussion

There are important differences between the two cases. In case 1 the ovarian ectopic pregnancy was diagnosed intraoperatively, while in case 2 it was provisionally diagnosed after using 3D ultrasound scanning and was confirmed laparoscopically. This conforms with the statement that intact ovarian pregnancies could be detected by ultrasound scanning whereas ruptured ones could not be easily differentiated from ruptured tubal pregnancies [14], as in the first case. Secondly, the ectopic pregnancy ruptured early despite relatively low  $\beta$ -hCG level in case 1. This may be an indication that ovarian ectopic pregnancies should be managed early to avoid severe intraperitoneal

haemorrhage, bearing in mind that the majority of them ruptured early during the first trimester [8,9]. Many cases of ruptured ectopic pregnancies have been reported even with negative urinary pregnancy test levels [15,16].

Surgical management of ovarian ectopic pregnancies usually involves oophorectomy or conservatively wedge resection of the affected ovary. These may be too drastic measures in general but more so in women with compromised ovarian reserve. Both cases presented in this article had endometriosis. The second patient had low antimullerian hormone level as well as a small contralateral ovary. Other reported factors which might increase ovarian ectopic pregnancies risks included pelvic inflammatory disease, assisted reproductive technology and use of intrauterine contraceptive devices.

It is important to stress that no ovarian tissue was removed in both reported cases to preserve future fertility potential. This contravenes with Speigelberg's criteria to establish a diagnosis, but these criteria have already been superseded by other more practical ones [5]. Furthermore, preserving ovarian tissues far outweighs the importance of making a histological diagnosis of the origin of an ectopic pregnancy. This is especially so when both tubes were normal, and the diagnosis was obvious. The technique used in these two cases was easy as there was no brisk active bleeding at the time of intervention. Also plan B for using methotrexate as a backup was in mind, in case of any residual trophoblastic tissue. This should be discussed and agreed with the patients beforehand.

On a different issue, historically most Gynaecologists prefer to perform laparotomy for acute cases with haemoperitoneum. However, managing case 1 safely laparoscopically gave support to the opinion that laparoscopic management is not always contraindicated when dealing with ectopic pregnancies with haemoperitoneum. In fact, it was described as the mainstay method for managing even haemodynamically unstable patients with ectopic pregnancy [17].

## Conclusion

Early intervention in suspected ovarian ectopic pregnancies may be necessary despite low  $\beta$ hCG levels to prevent catastrophic rupture with massive intraperitoneal haemorrhage. This is especially so in women with known risk factors as mentioned before. 3D ultrasound scanning may help in this respect to distinguish an ovarian pregnancy from a corpus luteum [18] and helps with early diagnosis. It may be useful to use strong suction, saline jetting to dislodge trophoblastic tissues and sharp curettage of the ovarian ectopic sac, together with bipolar electrocoagulation of the raw area whenever possible. This could be the primary step and may negate the need for the more drastic procedures including wedge resection. This technique proved to be safe and effective in the two cases presented, but more work is needed to verify its efficacy and safety in the future. This is important as more women are getting pregnant at an older age with the inevitable reduced ovarian reserve. This in addition to the reported increased incidence of ovarian ectopic pregnancies with assisted reproduction treatment which is frequently needed by women in this age group. However, patients should be counselled that they might need further medical treatment in case of residual trophoblastic tissues. In my view, combining this technique with medical treatment, if and when necessary, is superior to the other more drastic surgical techniques as far as ovarian reserve is concerned.

Finally, it is high time to abandon Speigelberg's four strict criteria for diagnosing ovarian ectopic pregnancies. These were set in 1878 when ultrasound scanning, and MRI were not yet developed, and laparotomy was the only means for making a diagnosis. This is especially so regarding the need to have ovarian tissues mixed with trophoblasts in the histopathology specimen to confirm the origin of the ectopic pregnancy. This last point has been addressed and nullified in the past (5, 7). The two cases presented in this manuscript supported this view as well and complied with the diagnostic criteria described by Sergent et al., in 2002 (5). It is high time for these new criteria to be used at this day and age.

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