Recurrent ectopic pregnancy after bilateral salpingectomy: A case report*

By Bernadette MAyumi T. Mortel, MD and Debby F. Pacquing-Songco, MD, FPOGS, FPSRM

Department of Obstetrics and Gynecology, University of the East Ramon Magsaysay Memorial Medical Center

ABSTRACT

Recurrent ectopic pregnancy after bilateral salpingectomy is a very rare condition, with only one previous case reported. This is a case of a 29 year old G4P0(0030) who presented with abdominal pain at the background of missed menses and positive pregnancy test, two years after bilateral salpingectomy. Intraoperatively, a 5x4cm bleeding mass was visualized at the right distal tubal remnant, containing an embryo and decidualized tissues. Excision of the adnexal mass and bilateral tubal remnants were completed. In such cases, total salpingectomy is the gold standard in preventing recurrence of another extrauterine gestation. In the presence of tubal remnants, a hysterosalpingography is recommended to assess tubal patency. Ultimately, the rarity of this case demands prompt recognition of risk factors, clinical presentation and appropriate management. It underscores the importance of maintaining vigilance, with high index of clinical suspicion in all women in the reproductive age group, especially those with risk factors.

Keywords: Amenorrhea, Bilateral salpingectomy, Recurrent ectopic pregnancy, Tubal Patency

INTRODUCTION

Ectopic pregnancy occurs in 1-2% of all pregnancies. It remains to be the leading cause of maternal mortality in the first trimester, accounting for 6% of all pregnancy-related deaths. In recent years, the incidence of ectopic pregnancy has risen due to the prevalence of pelvic inflammatory disease and assisted reproductive techniques. The occurrence is further increased as the surgical management itself for ectopic pregnancy, that is pelvic surgery, is a leading risk factor for having *another* extrauterine gestation. Specifically, pelvic surgery increases the risk of having another ectopic pregnancy fivefold to 10-fold over the general population[1]. However, in spite of the increase in the total incidence of ectopic pregnancy, recurrent ectopic pregnancy *after* salpingectomy is represented by a small population. With mortality rates 10-15 times higher than primary ectopic pregnancies, less than a dozen cases in the last decade have been reported of ipsilateral ectopic pregnancy in tubal remnant following salpingectomy[2,3]. The scarcity is supported by another small sample study in Japan that has stated an incidence rate of 1.16% in an 11-year span[2]. That being said, recurrent ectopic pregnancy following *bilateral* salpingectomy is even rarer with only one previously reported case[4], and the first documented case in the Philippines. Consequently, the rarity of this case necessitates recognition of risk factors, clinical presentation and further studies on possible pathophysiologies leading to repeated ectopic gestations. As such, this is the first reported case in the Philippines of a patient with her fourth ectopic pregnancy preceded by three pelvic surgeries, the last of which being a bilateral salpingectomy.

CASE REPORT

This is a case of a 29-year-old Gravida 4 Para 0, who at 8 1/7 weeks age of gestation, presented with hypogastric pain of two days duration.

All three previous pregnancies were extra-uterine tubal gestations. Her first pregnancy occurred in 2011, for which she underwent an exploratory laparotomy with left salpingectomy for a ruptured tubal ectopic pregnancy. After two years, this was succeeded by an unruptured tubal ectopic pregnancy on the contralateral tube. As the adnexal mass had not yet ruptured, and the patient remained desirous of pregnancy, the conservative surgical option of salpingostomy was performed. Subsequently, after two years, the patient had another ruptured tubal ectopic pregnancy on the right fallopian tube, finally necessitating salpingectomy, that ultimately impaired fertility.

Since the last pregnancy, interval history was unremarkable until the most recent complaint of

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hypogastric pain. It was characterized as dull and intermittent, with no associated vaginal bleeding. Due to a history of similar symptoms, the patient immediately sought consult in a private clinic, wherein a positive pregnancy test was confirmed. Thereafter, a transvaginal ultrasound was done revealing an empty uterus with no gestational sac, and a complex right adnexal mass, with minimal fluid in the cul-de-sac. The initial impression was ectopic pregnancy, probably unruptured, right. However, the patient opted to transfer to our institution for second opinion.

On examination, she had stable vital signs, with no pallor, with unremarkable chest and cardiac exam. Centering on the abdomen, it was soft, with guarding, and direct and rebound tenderness on the right hypogastric area. Speculum examination was remarkable due to yellowish nonfoul smelling vaginal discharge. Furthermore, internal examination revealed a closed cervix, with cervical motion tenderness and right adnexal tenderness, with no adnexal mass appreciated. Lastly, rectovaginal exam confirmed fullness in the cul-de-sac, with tenderness.

Initial assessment was ectopic pregnancy, right, probably ruptured, and the patient was admitted for surgical intervention, particularly, emergency exploratory laparotomy. Intraoperatively, there was hemoperitoneum of 400cc. The uterus was small with filmy pelvic adhesions displacing it to the right (Figure 1). The left fallopian tube was intact from the cornu ending into a proximal tubal remnant which measured 3 cm. The right fallopian tube was also intact from the cornu, with a proximal tubal remnant that measured 2 cms in length. A distal tubal remnant on the right, measuring 4 cms, was also visualized, indicating a previous partial salpingectomy. Thereafter, a 5x4 cm bleeding adnexal mass was noted on the distal tubal remnant, lateral to the corpus, adherent to the right pelvic side wall. The mass was easily removed by blunt dissection with minimal bleeding. Hemostasis was established with suture ligation of bleeders in the mesentery.

Thereafter, total bilateral salpingectomy was completed, with bilateral excision of the remaining fallopian tubes at the cornual area. Finally, other pelvic and abdomen organs were inspected with unremarkable findings. Abdominal wall was closed in layers, and the patient tolerated the procedure well, with an estimated blood loss of 500cc. Postoperative period was uneventful. Due to intraoperative findings of pelvic adhesions consistent with pelvic inflammatory disease, intravenous clindamycin and gentamycin were administered. This was later shifted to oral doxycycline. She was eventually discharged on her 3rd postoperative day. The diagnosis of tubal ectopic pregnancy was confirmed histologically as intraluminal chorionic villi, decidual tissues, and scattered hemorrhage were identified (Figures 3 and 4).

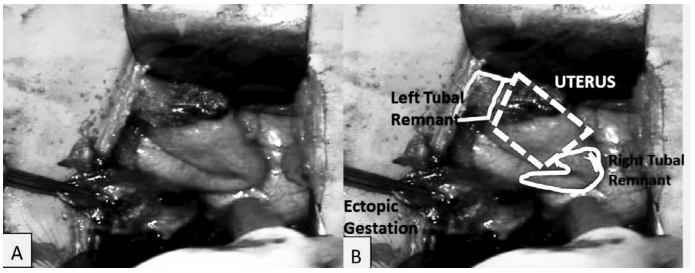

Figure 1. Intraoperative Findings: Dextrorotation of the uterus with dense pelvic adhesions. On the right, a bleeding adnexal mass, approximately 5x4cm is located.

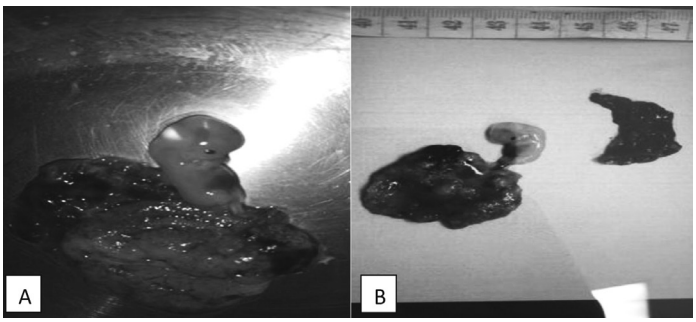

Figure 2. Intraoperative Findings: Ruptured adnexal, measuring 5x4cm containing embryo and decidualized tissues

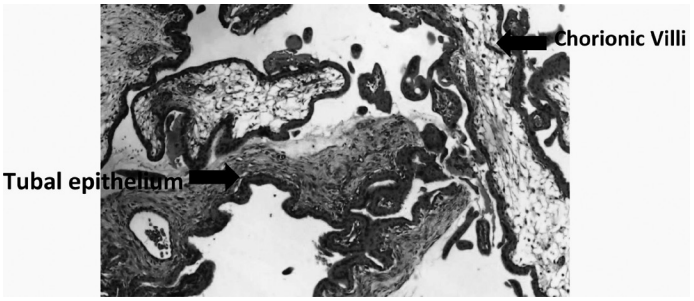

Figure 3. Histopathologic Findings. Tubal epithelium at the lower left with chorionic villi at the upper right

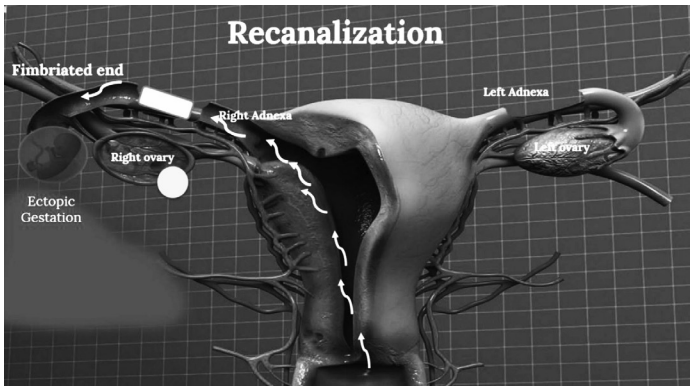

Figure 4. Recanalization of the tubal remnants leading to passage of sperm and fertilization of the egg

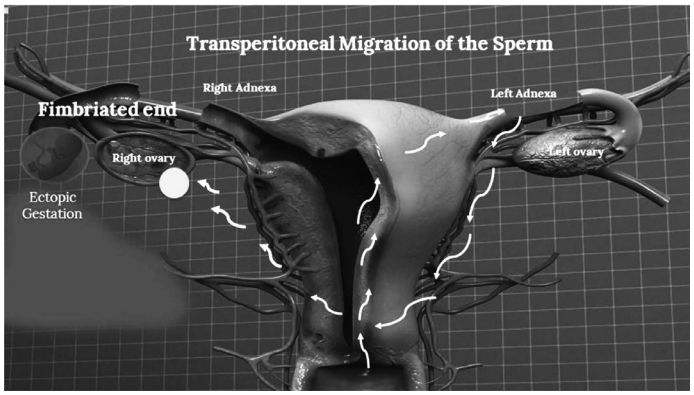

Figure 5. Transperitoneal Migration of the Sperm leading to fertilization of the egg on the contralateral tube

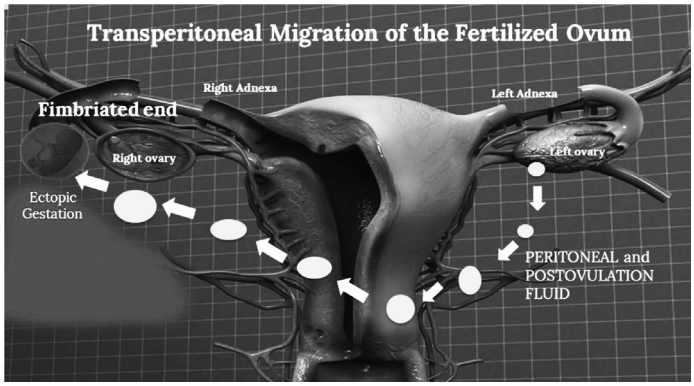

Figure 6. Transperitoneal migration of the fertilized ovum with ectopic gestation on distal tubal remnant

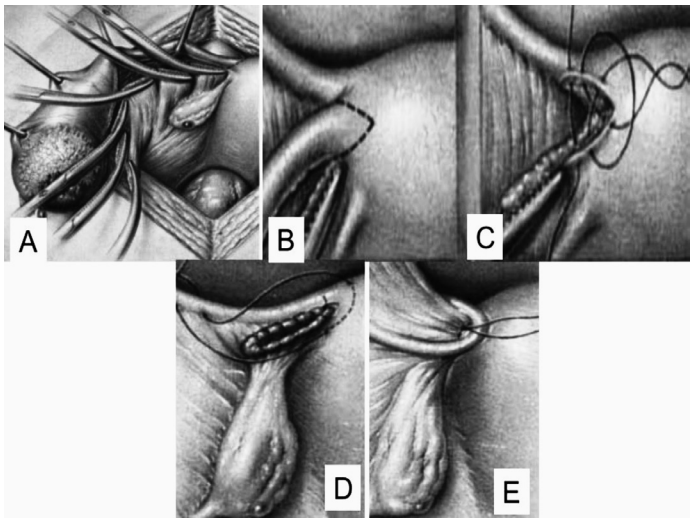

Figure 7. Modified Coffey Suspension as illustrated and described exactly in Te Linde's. A. The mesosalpinx is clamped as close to the tallopian tube as possible. B and C. The tube is removed by creating a small myometrial wedge at the cornu. D and E. The round and broad ligaments are sutured over the cornu

CASE DISCUSSION

As aforementioned, ectopic pregnancy occurs in 1-2% of all pregnancies. Tubal gestations are most common, occurring 95-97% of the time, 70% of which are found in the ampulla. The classic triad of amenorrhea, abdominal pain, and vaginal bleeding present in only 50% of cases[5]. In relation to our patient, she had abdominal pain at the background of missed menses, with no history of vaginal bleeding. This is consistent with a study done on patients with history of recurrent ectopic pregnancy. According to the study, these patients present as a unique clinical subgroup, who consult at a significantly earlier gestation primarily because of their awareness of their condition and early presentation[1].

Most patients with a single ectopic pregnancy rarely have another, as the incidence of the first recurrent ectopic pregnancy is 15%. However, the probability of having a third extrauterine gestation rises to as high as 30% after two previous ectopic pregnancies[5-8]. As observed in this case, one of the leading risk factors of the patient is her history of three previous ectopic gestations. Unfortunately, the exact incidence rate of a fourth recurrent ectopic pregnancy after salpingectomy remains unknown due to the very limited number of cases. Thus, the exact pathophysiology remains unclear. Nevertheless, three theories have been postulated to explain this condition.

The first theory involves the recanalization of the ipsilateral fallopian tube, allowing passage of the sperm, but obstructing the transport of the fertilized ovum to the uterus (Figure 4)[2,3,5]. According to a study on ectopic pregnancy after bilateral tubal ligation, recanalization creates *''an abnormal reconstitution of the tubal lumen with the formation of blind pouches and slit like spaces''*[6]. These physical obstacles hinder the normal passage of the fertilized embryo, thereby leading to the implantation of the ovum in the tubal lumen. In addition, as concluded in several studies, the post reversal tubal length or remnant is the most important determinant factor for recanalization. Post reversal tube length of more than 4 cms resulted in a pregnancy rate of 50%, but nearly zero when the length was less than 4 cm[7]. On top of this, it has been observed that not only was the length of tubal remnant a factor in recanalization, but also it affects time interval between the procedure and another pregnancy. Tubal length longer than 4 cm had a significantly shorter interval to conception (10.2 months) than those with less than 4 cm of tubal remnant (19.1 months)[8]. Similarly seen in our patient, the tubal remnants of the right and left fallopian tubes are significantly lengthened at 4 and 3 cm, respectively. As such in our patient, long tubal remnants in addition to dense pelvic adhesions, there was indeed a high likelihood of developing another ectopic pregnancy. More so, the

hypothesized time interval between each pregnancy given the tubal remnants of 4 cms is consistently seen in our patient, as each of her pregnancy was approximately 2 years apart.

The second theory involves the passage of spermatozoa through the contralateral fallopian tube into the peritoneal cavity to fertilize the ovum on the side of ectopic gestation (Figure 5). In a study published in the British Journal of Obstetrics and Gynecology, transperitoneal migration of sperm occurs routinely in all pregnancies, approximately nearly 50%[9-11]. This has three major implications: (1) spermatozoa freely courses through the peritoneal cavity after intercourse, (2) this theory partly explains the pathophysiology behind abdominal pregnancies, (3) in the presence of a damaged fallopian tube and healthy contralateral tube, the damaged fallopian tube does not necessarily have to be the one to conduct the passage of the sperm in order for it to be the final recipient and localization of the same pregnancy[9]. In relation to our patient, despite having both tubes ligated, the contralateral fallopian may have only been partially obstructed, thereby permitting the free passage of spermatozoa into the peritoneal cavity. This leads to the normal fertilization of the right ovary, ultimately leading to a distal ectopic pregnancy in the right remnant tube.

Finally, the last theory is similar to the previous in that involves the transmigration of a fertilized ovum (Figure 6). In a study reported by the European Society of Human Reproduction and Embryology, it has been postulated that oocytes are extruded into the cul-de-sac, and float in peritoneal and follicular fluid released during ovulation[12]. This fertilized egg is eventually picked up by the fimbriae of the contralateral tube. Due to the longer distance and slower transit, there is a delay in the transport of the fertilized ovum, and the sequential scheduled event of blastocyst implantation. In normal pregnancies, after the disintegration of the zona pellucida and hatching of the blastocyst, the blastocyst begins to invade the uterine lining for implantation. In contrast to ectopic pregnancy, the delay in transport of the fertilized ovum eventually leads to the hatching of the blastocyst in the fallopian tube instead of the uterus. Another contributary factor for the extrauterine gestation is the size of the fertilized ovum. In transit to the contralateral tube, the blastocyst grows and undergoes cellular division. Its enlarged size during contralateral tubal pickup hinders its passage in the tubal lumen, causing implantation in the distal end of the tubal remnant[13].

The similarity amongst the three theories involve the presence of remnant or damaged tubes. Having said this, appropriate surgical measures should be taken to prevent recurrence of ectopic pregnancies. When performing a salpingectomy, it is crucial not to leave a long stump. Partial salpingectomy may be the preferred procedure for some surgeons as it minimizes bleeding at the isthmic portion of the fallopian tube[2]. Nevertheless, total salpingectomy may be the better surgical option. Given that the patient is already high-risk due to probable pelvic inflammatory disease and history of recurrent ectopic pregnancies, proceeding with a total salpingectomy may decrease the recurrence of ectopic gestations better than partial salpingectomy. Proceeding with a partial salpingectomy by leaving tubal stumps nearly has similar risks to salpingostomy in terms of recurrence of ectopic pregnancy[3]. It may be concluded that total salpingectomy is the gold standard surgical management of ectopic pregnancy especially in those who are considered high-risk for recurrence. Though total salpingectomy may be difficult to achieve with endoloops, adequate diathermy may be done instead[2,3]. Furthermore, after total salpingectomy, complete peritonealization of the cornual incision using the broad ligament, also known as Modified Coffey suspension, prevents occurrence of an ectopic gestation in cornu[14] (Figure 7).

CONCLUSION

As the leading cause of maternal mortality in the first trimester, the growing prevalence of ectopic pregnancy instigates us to further explore the few cases of recurrent ectopic pregnancy after bilateral salpingectomy. Similarly to single ectopic pregnancies, history of pelvic surgeries and pelvic inflammatory disease are the leading risk factors for recurrent ectopic pregnancy. It is important to note that as the incidence of repeated ectopic pregnancy increases per individual, so does the risk of having another recurrence. Hence, in eliminating the possibility of recurrent ectopic pregnancies, total salpingectomy with complete peritonealization (Modified Coffey suspension) is superior to partial salpingectomy wherein remnant tubes are left behind. Especially in patients who are considered high-risk in developing another ectopic pregnancy, total salpingectomy is considered gold standard as surgical management of choice. Equally important, this particular case reiterates the importance of clinical awareness and vigilance in considering recurrent ectopic pregnancy in those with previous history of ectopic gestation and surgery. Fertility-impairing surgeries do not absolutely eliminate the possibility of a pregnancy. Finally, in the presence of tubal remnants, it is recommended that tubal stump patency should be assessed via hysterosalpingography, and if needed tubal occlusion devices may be placed to decrease the risk of ectopic gestation. ■

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