JOURNAL OF GYNECOLOGIC SURGERY Volume 29, Number 6, 2013 © Mary Ann Liebert, Inc. DOI: 10.1089/gyn.2013.0036

Case Report

Recurrent Distal Ipsilateral Ectopic Pregnancy: A Case Report and Literature Review

Nilesh Agarwal, MRCOG, Ishita Das, MBBS, BSc, Oladimeji Olowu, MRCOG, Anupama Shahid, MRCOG, and Funlayo Odejinmi, FRCOG

Abstract

Background: Ipsilateral recurrence after salpingectomy is rare, but delay in diagnosis can result in rupture and potentially life-threatening hemorrhaging. Because of the paucity of data, there are various methods by which ipsilateral recurrent ectopic pregnancies have been managed. It may be possible to decrease the incidence of such pregnancies further with complete removal of the Fallopian tubes at primary surgery. *Case:* This article presents a rare case of repeat ipsilateral ectopic pregnancy in the distal remnant part of the Fallopian tube following laparoscopic salpingectomy. A complete salpingectomy to remove the ectopic pregnancy and the fimbrial end of the affected Fallopian tube was performed. A brief literature review is provided regarding similar cases. *Results:* This patient's postoperative recovery was uneventful, and she was discharged on the following day. The histopathology results confirmed the presence of products of conception in the fimbrial end of her Fallopian tube. *Conclusions:* Early diagnosis and immediate management of recurrent ipsilateral ectopic pregnancy are imperative for minimizing morbidity and mortality. Every effort should be made to excise the entire affected Fallopian tube during the primary salpingectomy, so that there are no remnants that may become the sites of any possible future ectopic pregnancies. (J GYNECOL SURG 29:XX)

AU2

Introduction

SURGERY REMAINS THE METHOD of management for the majority of women with ectopic pregnancies. The recurrence rate after a first ectopic pregnancy is 10%.¹ The recurrence risk depends on the state of the pelvis and other factors that may have contributed to the first ectopic pregnancy. Is there an iatrogenic reason for some recurrences of ectopic pregnancy? To reduce the risk of recurrence after salpingectomy, meticulous surgery with complete removal of the affected area of the Fallopian tube is essential. If the fimbrial end of the Fallopian tube is left as a remnant, not only does it increase the risk of recurrence of ectopic pregnancy but this situation may also result in added future morbidity.

Between January 2000 and April 2012, from a population of 816 women who presented with ectopic pregnancy to Whipps Cross University Hospital, London, UK, and there were 72 women with repeat ectopic pregnancy. Of these 72 women, 3 of the pregnancies were ipsilateral, of which 1 was found in the distal remnant part of the Fallopian tube.

This article presents a rare case of a repeat ipsilateral ectopic pregnancy in the distal remnant part of the Fallopian tube following laparoscopic salpingectomy.

Case

A 27-year-old, gravida 3, para 0+2, woman presented with 9 weeks of amenorrhea, severe lower abdominal pain, and dizziness. Two years earlier, in another institution, she had undergone laparoscopic salpingectomy for a left ruptured ectopic pregnancy. Her serum human chorionic gonadotropin level was 381.5 U/L and a transvaginal ultrasound scan did not reveal an intrauterine pregnancy, but there was a heterogeneous left adnexal mass measuring 35×40×41 mm with a significant hemoperitoneum. At laparoscopy, it was noted that she actually had a ruptured ectopic pregnancy in the distal remnant of her left Fallopian tube, with a 700-mL ■ F1 hemoperitoneum (Fig. 1). The right Fallopian tube and ovary were normal. A complete salpingectomy to remove the ectopic pregnancy and the fimbrial end of the affected Fallopian tube was performed.

Results

This patient's postoperative recovery was uneventful and she was discharged on the following day. The histopathology result confirmed the presence of products of conception in the fimbrial end of the Fallopian tube.

▲AU1

Department of Obstetrics and Gynaecology, Whipps Cross University Hospital, London, United Kingdom.

2

AGARWAL ET AL.



AU5

FIG. 1. Site of recurrent ectopic pregnancies.

Discussion

In the latest report of the Confidential Enquiry into Maternal Deaths (CEMACE), in the United Kingdom, ectopic pregnancy was rated as the leading cause of early pregnancy deaths and overall the fifth most common cause of maternal deaths.² Between 2006 and 2008, there were 6 maternal deaths caused by ectopic pregnancies out of 11 (55%) early pregnancy deaths. The risk of recurrence of ectopic pregnancy ranges from 10% to 27% which is a 5–10-fold increase of the background risk in the general population.³ The incidence of recurrent ectopic pregnancy is ~15%, and this rises to 30% following two ectopic pregnancies.⁴ This recurrence predominantly occurs in the contralateral tube or in the proximal stump after a partial salpingectomy.

First author/year & ref.	Age (years)	Details	Treatment

TABLE 1. LITERATURE REVIEW OF RECURRENT IPSILATERAL ECTOPIC PREGNANCY (EP) FOLLOWING SALPINGECTOMY

Agarwal/2012 (current case)	27	Left partial salpingectomy for EP 2 years previously Left distal stump EP	Partial salpingectomy
Malhotra/2011 ¹¹	23	Right partial salpingectomy for EP 2 years previously Right distal stump EP	Partial salpingectomy
Sanchez Millan/2010 ¹²	Not mentioned	Right EP treated with methotrexate 8 years previously Right partial salpingectomy for EP 6 years previously Right EP	Methotrexate
Mohiyiddeen/2010 ¹³	38	Partial right salpingectomy for EP 4 years previously Right tubal stump ectopic—resection of stump 6 months later right distal EP	Excised with needle diathermy
Anwar/2010 ¹⁴	35	Right partial salpingectomy for EP 6 months previously Right proximal stump EP	Partial salpingectomy
Liu/2009 ¹⁵	28	Left partial salpingectomy for EP 5 years previously Left distal stump EP	Partial salpingectomy
Fischer/2009 ¹⁶	28	Left partial salpingectomy for EP previously Left proximal stump EP	Partial salpingectomy
Chou 2009 ¹⁷	38	Right partial salpingectomy for EP 2 years previously Proximal right stump EP	Resection of stump
	29	Right partial salpingectomy for EP 3 years previously Right distal stump EP	Partial salpingectomy
Liu/2009 ¹⁸	28	Left partial salpingectomy for EP 5 years previously Left proximal stump EP	Partial salpingectomy
Milingos/2008 ¹⁹	38	Two ipsilateral ectopic pregnancies in right tube Right salpingectomy & removal of tubal stump Right cornual EP	Removal of right uterine cornua
Chou/2008 ²⁰	23	Right partial salpingectomy for EP 2 years previously Right distal stump EP	Partial salpingectomy

(continued)

DISTAL IPSILATERAL ECTOPIC PREGNANCY

	TABLE 1. (CONTINUED)						
First author/year & ref.	Age (years)	Details	Treatment				
Faleyimu/2008 ²¹	22	Left partial salpingectomy for cornual EP 5 years previously Left distal stump EP	Partial salpingectomy & oopherectomy				
Tan/2007 ²²	27	Left partial salpingectomy for EP 8 years previously Left distal stump EP	Partial salpingectomy				
Rizzuto/2007 ²³	26	Right partial salpingectomy for EP previously Right proximal stump EP.	Partial salpingectomy				
Okunlola/2006 ²⁴	30	Right partial salpingectomy for EP previously	Right cornual wedge resection				
	39	Left partial salpingectomy for EP 8 years previously Left distal stump EP	Partial salpingectomy				
	28	Right partial salpingectomy for EP previously Proximal right stump EP Right cornual wedge resection	Right cornual wedge resection				
Zuzarte/2005 ⁹	32	Partial left salpingectomy for EP 3 months previously Distal remnant EP	Partial salpingectomy				
Chien/2005 ²⁵	40	Right partial salpingectomy for EP 10 years previously Right proximal stump EP	Partial salpingectomy				
Rizos/2003 ²⁶	33	Partial left salpingectomy for EP 14 weeks previously Left cornual ectopic pregnancy	Left cornual salpingectomy				
Mathew/2002 ²⁷	25	Partial left ampulary salpingectomy for EP 4 months previously Proximal left stump EP—partial left salpingectomy Proximal left stump EP	Partial salpingectomy				
Adebamowo/2000 ²⁸	Not mentioned	Partial salpingectomy for EP 3 years previously	Total salpingectomy				
Oki/1998 ²⁹	32	Stump ectopic EP Right partial salpingectomy for EP 3 years previously Right proximal stump EP	Resection of corneal portion Partial salpingectomy				
Lema/1995 ³⁰	31	Three consecutive ipsilateral	Milking ectopic pregnancy (first, second) Partial salpingectomy (third)				
Cartwright/1984 ⁵	16	Right partial salpingectomy for EP 4 months previously Proximal right stump EP	Partial salpingectomy				

Ipsilateral EP following partial salpingectomy was first reported in 1984.⁵ Even after bilateral salpingectomy, it is possible to have a recurrence in the interstitial region of the uterus.⁶ In the current authors' unit, of Whipps Cross University Hospital, of the 816 patients who underwent surgical management of ectopic pregnancy, recurrence occurred in 72 (8.8%) patients of which 3 (4.2%) were ipsilateral. The literature review (see section below), shows that, although distal recurrent ipsilateral ectopic pregnancies are rare, they can still occur after salpingectomy. This should always be considered as a differential diagnosis in all patients with a previous history of ectopic pregnancy. The literature review illustrated that, of 26 cases of recurrent ipsilateral ectopic

pregnancies, at least 10 cases occurred in the distal remnant part of the Fallopian tube (38.5% of reported cases).

In current authors' unit, of Whipps Cross University Hospital, 100% of haemodynamically stable women with ectopic pregnancies are managed laparoscopically, with this percentage falling to 85% in hemodynamically unstable women,7 using either electrocautery or an endoloop, according to the most common techniques reported in use for laparoscopic salpingectomy.

Lim et al. conducted a prospective randomized controlled trial comparing the endoloop and electrocautery for surgical treatment of ectopic pregnancy in 102 patients.⁸ These researchers concluded that use of the endoloop appeared to be as effective as electrocautery and was a safe alternative to 4

AGARWAL ET AL.

electrocautery for laparoscopic salpingectomy in tubal pregnancy. However, with either technique the entire Fallopian tube should be removed, leaving neither the proximal stump nor the distal part of the tube in order to minimize the risk of recurrence.

With respect to the etiology of repeat ectopic pregnancy in the distal stump of the Fallopian tube, it has been hypothesized that transperitoneal migration of spermatazoa or of the embryo from the contralateral side may be a reason for recurrent ipsilateral distal tubal ectopic pregnancy.⁹ Again, it is therefore imperitive that, when salpingectomy is performed, all of the fimbrial end of the affected Fallopian tube be removed from its attachment to the ovary, and that the cornual ends are occluded. This should then, theoretically, reduce the risk of recurrent ectopic pregnancy in that tube.

It has been stated that secondary prevention of recurrent ectopic pregnancy is difficult because of the paucity of risk factors that can be modified.³ However, meticulous surgery during primary operation to ensure that the entire Fallopian tube is removed can aid in preventing recurrences.

AU2►

There is also increasing evidence that there is no physiologic reason to keep the fimbrial end of the Fallopian tube separated from ectopic pregnancy and the risks of pyosalpinx and hydrosalpinx. The distal end of the Fallopian tube may also be a source of origin of serous intratubal carcinoma, and the site of development of the majority of serous ovarian and primary peritoneal cancers.¹⁰ For these reasons, leaving a remnant of the Fallopian tube following primary surgery should be a "never event."

Methods for Literature Search

For the literature review three major databases were searched, including Embase, MEDLINE,® and Cochrane databases. References of relevant articles were also hand searched, and a Google Scholar search was performed. The search results were limited to humans, and there were no language restrictions. The search terms used were <u>extrauterine</u>, <u>ectopic</u>, pregnancy, recurrent and ipsilateral (Table 1).^{5,9,11–30}

AU1 ► T1 ►

EQ1►

AU7►

Conclusions

Although recurrent ectopic pregnancy following ipsilateral partial salpingectomy is rare, it is important to maintain a high index of clinical suspicion in a women presenting with symptoms of ectopic pregnancy who has previously had a salpingectomy. Early diagnosis and immediate management are imperative for ensuring a positive outcome. In addition surgeons, should ensure that every effort is made to excise the entire tube at primary salpingectomy so that there is no remnant that may be the location of any possible future ectopic pregnancy.

Disclosure Statement

The authors certify that no actual or potential conflicts of interest in relation to this article exist.

References

1. Job-Spira N, Bouyer J, Pouly JL, Germain E, Coste J, Aublet-Cuvelier B, Fernandez H. Fertility after ectopic pregnancy: First results of a population-based cohort study in France. Hum Reprod 1996;11:99.

- 2. Wilkinson H; Trustees and Medical Advisers. Saving mothers' lives: Reviewing maternal deaths to make motherhood safer. 2006–2008. BJOG 2011;118.
- Butts S, Sammel M, Hummel A, Chittams J, Barnhart K. Risk factors and clinical features of recurrent ectopic pregnancy: A case control study. Fertil Steril 2003;80:1340.
- 4. Tulandi T. Reproductive performance of women after two tubal ectopic pregnancies. Fertil Steril Jul 1988;50:164.
- Cartwright PS, Entman SS. Repeat ipsilateral tubal pregnancy following partial salpingectomy: A case report. Fertil Steril 1984;42:647.
- Al-Sunaidi M, Sylvestre C. Ectopic pregnancy after bilateral salpingectomy. Saudi Med J 2007;28:794.
- Odejinmi F, Sangrithi M, Olowu O. Operative laparoscopy as the mainstay method in management of hemodynamically unstable patients with ectopic pregnancy. J Minim Invasive Gynecol 2011;18:179.
- Lim YH, Ng SP, Ng PH, Tan AE, Jamil MA. Laparoscopic salpingectomy in tubal pregnancy: Prospective randomized trial using endoloop versus electrocautery. J Obstet Gynaecol Res 2007;33:855.
- Zuzarte R, Khong CC. Recurrent ectopic pregnancy following ipsilateral partial salpingectomy. Singapore Med J 2005; 46:476.
- Dietl J, Wischhusen J, Häusler SFM. The post-reproductive Fallopian tube: Better removed? Hum. Repro. 2011; 26:2918.
- Malhotra V, Chauhan M, Ahuja K, Nanda S, Kumar S. Recurrent ectopic pregnancy after ipsilateral salpingectomy: Report of a rare case. J Gynecologic Surg 2011:27:151.
- Sanchez Millan V, de Mingo Romanillos L, Manuel Patiño Maraver V, Redondo Escudero S, de Motta Rodriguez A. Embarazo ectópico recurrente ipsolateral. Prog Obstet Ginecol 2010;53:416.

◀AU4

AU6

AU3

- 13. Mohiyiddeen L, Singh S, El-Gawly R. Three ipsilateral ectopic pregnancies managed surgically. J Obstet Gynaecol 2010;30:216.
- Anwar S, Uppal T. Recurrent viable ectopic pregnancy in the salpingectomy stump. Australasian J Ultrasound Med 2010; 13:37.
- Liu YL, Hwang KS, Chu PW, Ding DC. Recurrent ectopic pregnancy in the ipsilateral oviduct after prior laparoscopic partial salpingectomy. Taiwan J Obstet Gynecol 2009;48:417.
- Fischer S, Keirse MJ. When salpingectomy is not salpingectomy—ipsilateral recurrence of tubal pregnancy. Obstet Gynecol Int 2009;2009:article ID 524864
- 17. Chou SY, Hsu MI, Chow PK, Chiang HK, Su HW, Hsu CS. Recurrent ipsilateral ectopic pregnancy after partial salpingectomy. Taiwan J Obstet Gynecol 2009;48:420.
- Liu YL, Hwang KS, Chu PW, Ding DC. Recurrent ectopic pregnancy in the ipsilateral oviduct after prior laparoscopic partial salpingectomy. Taiwan J Obstet Gynecol. 2009; 48:417.
- Milingos D, Black M, Bain C. Three surgically managed ipsilateral spontaneous ectopic pregnancies. Obstet Gynecol 2008;112:458.
- Chou LL, Huang MC. Recurrent ectopic pregnancy after ipsilateral segmental salpingectomy. Taiwan J Obstet Gynecol 2008;47:203.
- 21. Faleyimu B, Igberase GO, Momoh MO. Ipsilateral ectopic pregnancy occurring in the stump of a previous ectopic site: A case report. Cases J 2008;21: 343.

DISTAL IPSILATERAL ECTOPIC PREGNANCY

- 22. Tan T, Elashry A, Tischner I, Jolaoso A. Lightning does strike twice: Recurrent ipsilateral tubal pregnancy following partial salpingectomy for ectopic pregnancy. J Obstet Gynaecol 2007;27: 534.
- Rizzuto MI, Macrae R, Odejinmi F. Persistent ectopic pregnancy following ipsilateral "salpingectomy." Gynecol Surg 2007;4:305.
- Okunlola MA, Adesina OA, Adekunle AO. Repeat ipsilateral ectopic gestation: A series of 3 cases. Afr J Med Med Sci 2006;35:173.
- Chien W-H, Liang C-C. Repeated ectopic pregnancy on the tubal stump after laparoscopic salpingectomy. Gynecol Surg 2005;2:311.
- Rizos A, Eyong E, Yassin A. Recurrent ectopic pregnancy at the ipsilateral fallpian tube following laparoscopic partial salpinectomy with endo-loop ligation. J Obstet Gynaecol 2003;23:678.
- Mathew M, Kumari R, Gowri V. Three consecutive ipsilateral tubal pregnancies. Int J Gynaecol Obstet 2002;78:163.

- Adebamowo A, Fakolujo O. Second ipsilateral ectopic gestation after total salpingectomy: A case report. Afr J Med Med Sci 2000;29:63.
- 29. Oki T, Douchi T, Nakamura S, Maruta K, Ijuin, H, Nagata Y. A woman with three ectopic pregnancies after *in-vitro* fertilization and embryo transfer Hum Reprod 1998;13:468.
- 30. Lema VM. Three consecutive ipsilateral tubal pregnancies in a nulliparous African woman: The role of conservative treatment. Cent Afr J Med 1995;41:62.

Address correspondence to: Nilesh Agarwal, MRCOG Department of Obstetrics and Gynaecology Whipps Cross University Hospital Whipps Cross Road Leytonstone, London E11 1NR United Kingdom

E-mail: shivnilesh@yahoo.com

AUTHOR QUERY FOR GYN-2013-0036-VER9-AGARWAL_1P

- AU1: Note that Literature Review section was moved to keep the reference citations in order of appearance. AU2: Do you want to remove the entire Fallopian tube or just the fimbral and distal ends? This is not clear
- throughout. Please clarify.
- AU3: Is ref. 2 correct? This reference needed authors.
- AU4: Translate Ref. 12 into English. Is this language Spanish or Portugese?
- AU5: Per e-mail of, do you want this figure in color.
- AU6: Ref. 18 is a duplicate of ref. 15; yet, it is listed two times in Table 1 as both refs. HOWEVER, REF. 15 says "RIGHT PROXIMAL stump" and REF. 18 says "LEFT DISTAL stump." Please supply new ref. 18. If this reference covers two cases, then please remove ref. 18 and renumber all the references in the table from that point on as well as the citations in the text. Alter the table so that the two cases are together as ref. 15.

EDITOR QUERY FOR GYN-2013-0036-VER9-AGARWAL_1P

EQ1: AU7 is cited. But relevant text is not provided. Please check.