Double trouble: A case of bilateral tubal pregnancy*

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ABSTRACT

Bilateral tubal pregnancy is the rarest form of ectopic pregnancy, and in most cases results from assisted reproductive techniques. The incidence of simultaneous bilateral tubal pregnancies has been reported to range from 1 per 725 to 1 per 1580 ectopic pregnancies or approximately corresponds to 1 per 200,000 pregnancies. To date, this is the only case reported in our institution. Bilateral tubal pregnancies are usually diagnosed intraoperatively, but with the advent of diagnostic tools and more readily available diagnostic modalities, an earlier diagnosis can be made to decrease maternal morbidity and mortality.

This is a case of a 24-year old female, who came in at the emergency room complaining of severe hypogastric pain. She was admitted as a case of ectopic pregnancy, probably ruptured. Subsequently, emergency exploratory laparotomy was done which revealed bilateral tubal masses, which on histopathological examination confirmed bilateral tubal pregnancy.

INTRODUCTION

Ectopic pregnancy is defined as a pregnancy that develops after implantation of the blastocyst anywhere else other than the endometrial lining of the uterine cavity. The incidence of ectopic pregnancy ranges between 1-2% of all pregnancies. Nearly 95% of them occur in the fallopian tubes. Synchronous bilateral ectopic pregnancy is very rare, and in most cases results from assisted reproduction techniques. Bilateral ectopic pregnancy is the rarest form of ectopic pregnancy and one has been reported in our hospital.

GENERAL OBJECTIVE

To present a rare case of bilateral tubal pregnancy in a 24-year-old female with no history of assisted reproductive technology and to recognize its importance as a life threatening condition.

Specific Objectives

To define ectopic pregnancy.
To state the incidence of bilateral tubal pregnancy.
To identify the risk factors in developing ectopic pregnancy especially those seen in the patient.
To discuss the pathophysiology of bilateral tubal pregnancy.
To discuss the diagnosis and management of bilateral tubal pregnancy.

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CASE REPORT

This is a case of K.P., a 24-year-old, Gravida 2 Para 1 (1001), who came in with a chief complaint of severe hypogastric pain and was admitted for the first time in our institution on May 4, 2016.

She has unremarkable past medical history. She has a family history of diabetes mellitus on her paternal side but denies other heredofamilial diseases such as hypertension, malignancies, cardiac and thyroid disorders.

She is the second among three siblings, a high school graduate and is currently unemployed. She has been in a relationship with a 28-year-old fast food crew for a year. She is a previous smoker of 0.25 pack years smoker and an occasional alcoholic beverage drinker.

Her last menstrual period was on February 10, 2016 with a computed age of gestation of 12 weeks, and an expected date of confinement on November 17, 2016. She had her menarche at 10 years of age, with regular intervals, lasting for 5 days, and consumes 2-3 moderately soaked pads per day. Patient claims to experience dysmenorrhea every menstrual cycle. She had her coitarche at 14 years of age and had 4 previous sexual partners. She has no history of contraceptive use and denies history of sexually transmitted infections.

She is a gravida 2 para 1 (1001). Her first pregnancy was delivered alive, term, via spontaneous vaginal delivery at home, assisted by a midwife. No fetomaternal complications were noted.

The history of present illness started a few hours prior to admission, when the patient experienced severe, sharp, stabbing, hypogastric pain radiating to the lower back associated with vaginal spotting. No other signs and symptoms were noted such as passage of meaty materials, fever or dysuria. Persistence of the above symptoms prompted patient to seek consult.

On physical examination, the patient was awake, coherent, and not in cardiorespiratory distress. Her blood pressure was 90/60 mmHg, cardiac rate of 102 beats per minute, respiratory rate of 20 cycles per minute and temperature of 36.5^{o} Celsius. She had pink palpebral conjunctivae, anicteric sclera, no nasoaural discharge, no tonsillopharyngeal congestion, or cervical lymphadenopathies noted.

Her chest expansion was symmetrical and no retractions were noted on the intercostal space. Auscultation of the lung area revealed clear breath sounds.

She had a dynamic precordium, her heartbeat had a normal rate and regular rhythym and no murmurs were appreciated.

Abdominal examination revealed a rigid, tender abdomen with muscle guarding.

Her external genitalia was grossly normal.

Speculum exam revealed a pinkish and smooth vaginal mucosa with no polyps, warts or ulcerations. On internal examination, her cervix was firm, closed, with cervical motion tenderness. Uterus was small with bilateral adnexal tenderness.

Patient had full and equal pulses on all extremities. No gross deformities noted.

At the emergency room, pregnancy test was positive. Laboratories such as complete blood count and urinalysis were done.

She was subsequently admitted as a case of Gravida 2 Para 1 (1001), Ectopic Pregnancy, Probably Ruptured and the plan was for emergency exploratory laparotomy with possible salpingectomy.

COURSE IN THE WARDS

Upon admission, patient was venoclysed. Complete blood count was done which revealed a hemoglobin of 106 g/L, hematocrit of 0.314, and white blood cell count of 24.15 x 10^{9}/L with predominance of neutrophils (Table 1). Urinalysis revealed red blood cell count of 8-10/hpf and white blood cell count of 10-15/hpf (Table 2). The patient underwent emergency exploratory laparotomy under spinal anesthesia. Upon exploration, there was 1.3 liters of hemoperitoneum. The left fallopian tube was converted to a 4x4 centimeter cystic, hemorrhagic mass with a 1.5 centimeter point of rupture at the ampullary area. The right fallopian tube on the other hand was converted into a 6x6 centimeter cystic, hemorrhagic mass with no point of rupture. Both ovaries and uterus were grossly normal. No adhesions were noted (Fig. 1). The surgical team then proceeded with bilateral salpingectomy. Estimated blood loss was 2 liters. The patient tolerated the procedure well.

At the post anesthesia care unit, the patient had stable vital signs and was noted to have adequate and clear urine output.

On her first post-operative day, the patient had no subjective complaints. She had stable vital signs and had already passed flatus. On physical examination, she was noted to have facial pallor, and abdomen was noted to be soft and non-tender. Post-operative complete blood count revealed hemoglobin of 73 g/L, hematocrit of 0.213 and white blood cell count of 19.46 x 10^{9}/L with predominance of neutrophils. She was started on soft diet. Two units of packed RBC were transfused. Wound dressing was done and revealed a well coaptated wound with no discharges.

The rest of the hospital stay was unremarkable. Vital sign were stable.

Histopathologic report revealed findings of a right fallopian tube that measures 5.0 x 4.0 x 2.0 cm with a dilated portion 2.0 cm from the fimbrae measuring 4.0 x 3.0 x 1.0cm and a left fallopian tube which measures 5.0

Table 1: Complete Blood Count

	5/4/16	5/4/16(post-op)	5/5/16	5/7/16
RBC x1012/L	3.23 x 10^{12}/L	2.56 x 10^{12}/L	2.26 x 10^{12}/L	3.20 x 10^{12}/L
Hemoglobin	106 g/L	79 g/L	73 g/L	96 g/L
Hematocrit	0.314	0.247	0.213	0.292
MCV	97.1 fl	96.4 fl	94.1 fl	91.1 fl
MCH	32.8 pg	30.9 pg	32.3 pg	30.0 pg
MCHC	338 g/L	320 g/L	343 g/L	329 g/L
Platelet count	363 x 10^{9} L	198 x 10^{9} L	157 x 10^{9} L	246 x 10^{9} L
WBC	24.15 x 10^{9} L	19.46 x 10^{9} L	6.77 x 10^{9} L	5.75 x 10^{9} L
Diff ct: Neutrophils	97.3%	93.1%	80.3%	60.9%
Lymphocyte	1.6%	3.2%	11.9%	1.0%
Monocyte	1.0%	3.6%	6.2%	11.1%
Eosinophil	0	0.1%	1.4%	6.2%

Table 2: Urinalysis

Color	Dark Yellow
Transparency	Hazy
Protein	Trace
Glucose	Negative
pH	Acidic
Specific Gravity	1.025
Red Blood Cells	8-10
White Blood Cells	10-15

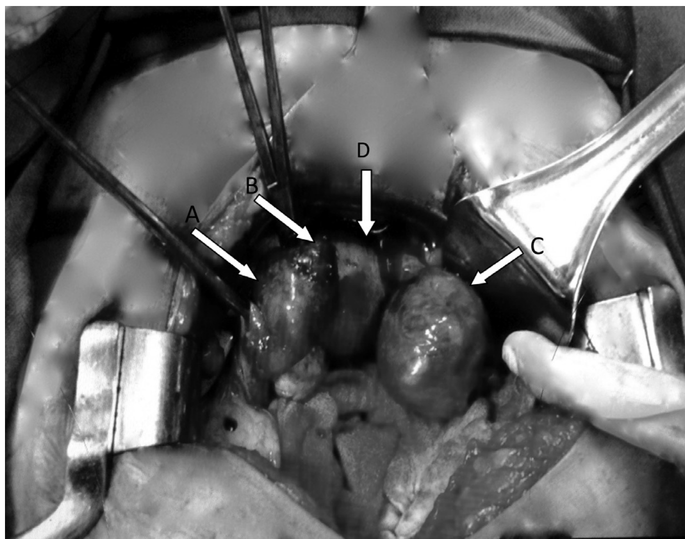

Figure 1. Arrow A shows the 4x4 centimeter cystic, hemorrhagic mass on the left fallopian tube; Arrow B shows the 1.5 centimeter point of rupture at the ampullary area; Arrow C shows the 6x6 cystic hemorrhagic mass on the right fallopian tube. Arrow D shows the uterus.

x 3.0 x 2.0 with a dilated portion 2.0 cm from the fimbrae measuring 4.0 x 3.0 x 1.0 with a 1.5 cm gross rupture at the ampullary area. Both have a dark brown to maroon discoloration, tubular and rubbery. Microscopic sections of both fallopian tubes show luminal projections lined by ciliated cuboidal cells with immature chorionic villi and decidual tissues (Figures 3.1 and 3.2). The final histopathologic diagnosis was Tubal Pregnancy, Right Fallopian Tube and Tubal Pregnancy, Ruptured, Left Fallopian Tube (Fig. 4).

CASE DISCUSSION

Ectopic is derived from the greek word *ektopos* meaning out of place. After successful fertilization, the blastocyst normally implants in the endometrial lining of the uterine cavity. Implantation anywhere else is considered an ectopic pregnancy[1]. About 1-2 percent of

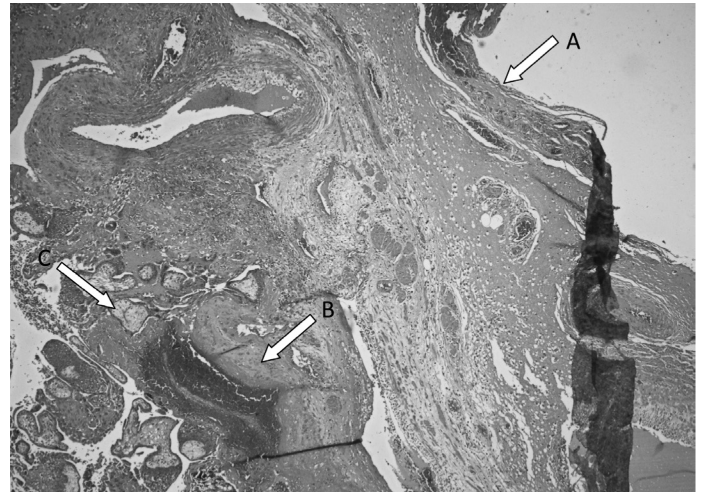

Figure 2. Microsection of the Right Fallopian Tube (showing chorionic villi and decidual tissues). Arrow A shows the lining epithelium (ciliated columnar epithelial cells.) Arrow B shows decidual tissues. Arrow C shows luminal projections lined by ciliated cuboidal cells with immature chorionic villi.

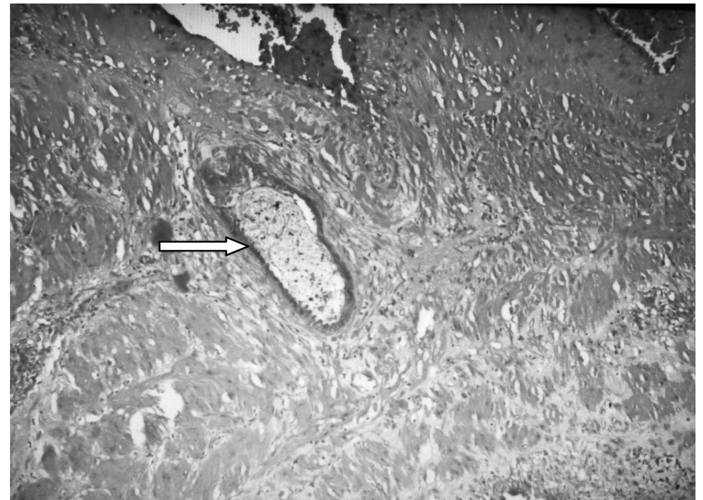

Figure 3.1. Microsection of the Left Fallopian Tube. Arrow shows the luminal projections lined by ciliated cuboidal cells with immature chorionic villi.

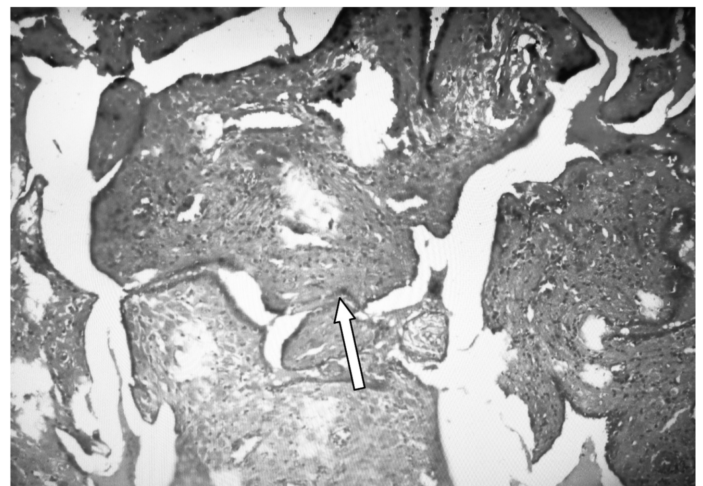

Figure 3.2 Microsection of the Left Fallopian Tube. Arrow shows decidual tissues.

SURGICAL PATHOLOGY REPORT

Name: PA K Specimen No. 1121-16
Date Received: 05-04-16 Date Reported: 05-16-16 Age: 24 Sex: F
Attending Physician: Dr. Specimen: Right Fallopian Tube, Left Fallopian Tube
Clinical Diagnosis: G2P1(1011)Tubal Pregnancy, Ampullary, Ruptured, Left; Ampullary, Unruptured, Right

FINAL HISTOPATHOLOGIC DIAGNOSIS:

TUBAL PREGNANCY, RIGHT FALLOPIAN TUBE
TUBAL PREGNANCY,RUPTURED, LEFT FALLOPIAN TUBE

AMPARO G. BAILON, MD, MD /
RUPALI D. KAKATI, MD
Pathology Resident

DAHLIA TERESA R. ARGAMOSA, MD, DPSP
Pathologist

GROSS DESCRIPTION:

Specimens received and labeled as "Right Fallopian Tube and Left Fallopian Tube" consists of right fallopian tube measuring 5.0 x 4.0 x 2.0 cm. with a dilated portion 2.0 cm. from the fimbrae measuring 4.0 x 3.0 x 1.0 cm, and the left fallopian tube measuring 5.0 x 3.0 x 2.0 cm with a dilated portion 2.0 cm from the fimbrae measuring 4.0 x 3.0 x 1.0 cm with a 1.5 cm gross rupture at the ampullary area. Both have a dark brown to maroon discoloration, tubular and rubbery.

Representative sections as follows:
R. Right Fallopian Tube S1B4
L. Left Fallopian Tube S1B7

MICROSCOPIC DESCRIPTION:

Sections of both fallopian tubes show luminal projections lined by ciliated cuboidal ce
with immature chorionic villi and decidual tissues.

Figure 4: Copy of the final histopathologic report.

all conceptions are ectopic and these account for 6 percent of all pregnancy related deaths[1]. Nearly 95 percent of ectopic pregnancies are implanted in the segments of the fallopian tubes and the remaining 5 percent are implanted in the ovary, peritoneal cavity or cervix[1]. About 81 percent are ampullary implantations and these are the most common, 12 percent are in the isthmus, 5 percent in the fimbrial end and 2 percent in the interstitial region[2]. Multifetal pregnancies may implant simultaneously with either both ectopic or one intrauterine and the other extrauterine[1]. Such usually occurs following assisted reproductive technologies (ART)[12]. The rate of ectopic pregnancies continue to increase in the United States and many European countries due to a number of factors which include increased rates of sexually transmitted infections, failed contraception, use of ARTs and tubal surgery[2].

According to World Health Organization (2007), 5 percent of maternal deaths in developed countries are from ectopic pregnancy. These deaths declined markedly from 1980-1992 due to improved diagnosis and management. In a review of deaths from ectopic pregnancy in Michigan, 44 percent of the women who died were either found dead at home or were dead on arrival at the emergency department[17]. In a review of 206 cases of ectopic pregnancy in the Philippines, the mortality was 7.28 percent. The main causes of death were secondary anemia due to either preoperative hemorrhage or both preoperative and postoperative hemorrhage and infection[18].

Bilateral tubal pregnancy, in the absence of preceding induction of ovulation or assisted reproductive technology, is an extremely unusual occurrence and the rarest form of ectopic pregnancy. Worldwide, the estimated incidence of bilateral tubal pregnancy is 1 in 725 to 1 in 1580 of all ectopic pregnancies[12] which corresponds to an occurrence of 1 in 200,000 pregnancies[3]. This is in contrast with the incidence of the other variants of ectopic pregnancy, with 1 in 9,000 pregnancies for cervical pregnancy, 1 in 2,000 to 1 in 60,000 pregnancies for ovarian pregnancy, 1 in 183,000 pregnancies for abdominal pregnancy and 1 in 16,000 to 1 in 30,000 pregnancies for heterotopic pregnancy[2].

Fishback, in 1953, reported a series of 76 cases of bilateral tubal pregnancy in the Canadian medical associated journal [8,11]. In 1989, Edelstein found 22 more cases[7,11]. Andrews, in 2008, reviewed literature and revealed 45 further case reports, 17 of which are associated with assisted reproductive techniques, and 28 patients are listed as spontaneous [4,11]. De Los Ríos, reviewed and analyzed 42 cases of bilateral tubal pregnancies reported in the last 10 years [9,11]. In total, there are about 200 cases of bilateral tubal pregnancy reported in the literature to date[14]. Most of these cases are usually diagnosed intraoperatively.

In 2007, Llovit reported a case of bilateral tubal pregnancy in a woman with no significant risk factors in the Philippine Council for Health Research and Development Library[11]. At present, there is no reported case of bilateral tubal pregnancy that has been published in the archives of the Philippine Obstetric and Gynecologic Society. There were no cases reported in the hospital census as well. The incidence of bilateral tubal pregnancy in the Philippines is not known.

The major risk factor of ectopic pregnancy is salpingitis[2]. Its morphologic sequelae account for about half of the initial episodes of ectopic pregnancy. The agglutination of the plicae (folds) of the endosalpinx produced by salpingitis can allow passage of sperm, but prevent the normal transport of the larger morula. The morula can be trapped in blind pockets formed by adhesions of the endosalpinx[2]. Though not documented, prior tubal infection, which can distort normal tubal anatomy, is likely present in our patient. Her sexual history revealed that the patient had 4 previous sexual partners which is a risk factor for having tubal infection.

It is also reported that cigarette smoking is associated with increased risk of ectopic pregnancy[2]. The number of cigarettes smoked per day was found to be directly related to the development of ectopic pregnancy, with a fourfold increased risk noted among women who smoked 30 or more cigarettes per day[6]. In the case of our patient, she

is a 0.25 pack-years smoker consuming 2-3 sticks per day for 2 years which probably increased her risk of having an ectopic pregnancy.

There are three possible theories for development of bilateral tubal pregnancy: 1) simultaneous multiple ovulation, 2) sequential impregnation or 3) transperitoneal migration of trophoblastic cells from one extrauterine pregnancy to the other tube with implantation there[16].

Foster stated that bilateral tubal pregnancy requires multiple ovulations to occur with simultaneous fertilization and implantation of both oocytes[5]. Another possible etiology is sequential impregnation, which implies fertilization and development of a second oocyte when a woman is already pregnant[16]. This is considered to be an extremely rare event in humans and is inherently difficult to prove. Tabachnikoff et al stated that transperitoneal migration of trophoblastic cells from one tube to another is a plausible explanation for the finding of fetal tissue in one tube and only villi in the other tube [4,16].

The most possible explanation applicable in our case is the theory of simultaneous multiple ovulation. Both ectopic gestations are of approximately the same age ruling out the theory of sequential impregnation. Transperitoneal migration of trophoblastic cells is also not a likely explanation because histopathology revealed the presence of decidual tissues and chorionic villi in the lumen of both fallopian tubes.

The most common symptoms of ectopic pregnancy which are abdominal pain, absence of menses, and irregular vaginal bleeding often characterized as spotting[1] were all present in our patient. In addition, physical examination of the abdomen revealed a rigid, tender abdomen with muscle guarding. There was exquisite cervical motion tenderness, with bilateral adnexal tenderness on internal examination which gives a high index of suspicion for a ruptured ectopic pregnancy. Rupture of an ectopic pregnancy causes intense pain and exquisite tenderness especially on bimanual examination and cervical motion which are present in nearly all women with an advanced or ruptured ectopic pregnancy. Other symptoms that develop following tubal rupture include syncope, dizziness and an urge to defecate[2].

Diagnostic modalities used to identify ectopic pregnancies include beta HCG level measurements, progesterone, transvaginal ultrasound and diagnostic surgery such as laparoscopy. Identification of an extrauterine yolk sac, embryo, or fetus in the fallopian tubes or ovaries through sonography clearly confirms ectopic pregnancy[2]. This, however, was not done in our case since the patient presented with an acute abdomen thus warranting a direct operation for emergency exploratory laparotomy. Spontaneous bilateral tubal pregnancy is rare, therefore preoperative diagnosis is uncommon indicating limitations in ultrasonography. Martinez, in 2009, made an unusual case report of early diagnosis by ultrasonography of a bilateral tubal pregnancy[10]. The most common method of diagnosing the second ectopic is through direct inspection of contralateral tube in the operating room or laparoscopic examination. In this case, it was diagnosed intraoperatively.

Laparoscopic surgical treatment is preferable to open procedures, because of faster patient recovery[2]. However, because of the acute symptoms of the patient, conservative management, with either laparoscopic surgery or medical management with methotrexate was not considered. Tubal surgery is considered conservative when there is tubal salvage, such as with salpingostomy or salpingotomy. Salpingostomy was attempted on the unruptured side of the fallopian tube. However, it was unsuccessful due to the uncontrolled bleeding from the implantation site. Some have shown conservative surgery may increase the rate of subsequent uterine pregnancy but is associated with higher rates of persistently functioning trophoblast. Radical surgery is defined by salpingectomy. Unfortunately, bilateral salpingectomy was done to our patient due to the size of the mass in both tubes.

Intraoperatively, the left fallopian tube was converted to a 4x4 centimeters hemorrhagic mass with a 1.5 centimeter point of rupture at the ampullary area. The right fallopian tube is converted to a 6x6 centimeters cystic hemorrhagic mass with no point of rupture. Our findings in this case are similar to that of Himangini et al and G.A.AL Quraan et. al who both noted a ruptured right ectopic with unruptured left ectopic pregnancy in which decision to undertake bilateral salpingectomy was also done[13]. With bilateral salpingectomy, the chances of having another natural pregnancy is impossible. The only way for her to have another pregnancy is through assisted reproductive technology such as in vitro fertilization which is cannot be afforded by the patient. Thus, a thorough patient education and counseling regarding the consequences of bilateral salpingectomy was provided for this patient. Advise on methods of assisted reproductive technology that maybe utilized in the event that she desires to have another child, as well as the expenses it would incur, was also explained.

CONCLUSION

This is a rare case of spontaneous bilateral tubal pregnancy. Cases such as this are usually diagnosed intraoperatively, but with the advent of diagnostic tools and more readily available diagnostic modalities, an earlier diagnosis can be made to decrease maternal morbidity and mortality. Although the incidence of a bilateral tubal pregnancy is rare, both adnexae and ovaries as well as

the entire pelvis should be thoroughly examined at the time of exploratory laparotomy when diagnosis of an ectopic pregnancy is made. Even if patients are at a high risk of having another ectopic pregnancy, conservative management is also an important consideration in such cases with attempts to save the tubes. Keeping in mind hopes of having future natural pregnancies, especially in our setting, where not everybody can afford assisted reproductive technologies. ■

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