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Heterotopic Ovarian - Tubal Pregnancies with Possibilities of Superfetation

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Abstract Heterotopic pregnancy is a rare condition found. The incidence is around 1 in 30,000 spontaneous pregnancies. Ovarian pregnancies occur around 0.5-3% and heterotopic pregnancies around 2% of all ectopic pregnancies. The diagnosis is intricate and based on surgical and histopathological observations. The management is, in spite of medical improvement. We presented an ectopic pregnancy suspected of being a right ovarian pregnancy and undergoing surgery, then 1 month later an ectopic pregnancy reappeared in a tubal rupture on the opposite side. We describe based on the literature review, diagnosis criteria and case management.

Keywords: *heterotopic pregnancy, ovarian pregnancy, superfetation*

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1. Introduction

Heterotopic pregnancy is a rare condition found. The incidence is around 1 in 30,000 spontaneous pregnancies, and in the present time there are about 1% of all pregnancies [1].

Generally heterotopic pregnancy can be diagnosed at 5-8 weeks gestation by 70%, at 9-10 weeks gestation by 20% and at 11 weeks gestation and above about 10% [2].

Ovarian pregnancies occur around 0.5-3% of all ectopic pregnancies [3]. In previous studies, 91.8% of primary ovarian pregnancies, 6.1% combined ovarian and tubal pregnancies and heterotopic pregnancies 2.0% [3].

Variations in heterotopic pregnancy that have been reported include a combination of intrauterine pregnancy against the tube and ovary. But also found cases of heterotopic pregnancy where there is a combination of tubal pregnancy and ovarian pregnancy [4].

Superfetation itself is a condition where there is fertilization of the ovum and implantation of the embryo in 1 pregnancy, which results in a pregnancy with 2 different gestational ages. Superfetation is a very rare occurrence in humans because follicular development and ovulation are suppressed during pregnancy [5].

This case will discuss a case that was initially diagnosed with an ectopic pregnancy (ovarian pregnancy with a differential diagnosis of rupture of the luteal corpus), but in development it is estimated to experience superfetation because at intervals a month later it has a tubal pregnancy.

2. Case Report

In this case, the patient was referred from a peripheral hospital with a diagnosis of Ectopic Pregnancy. P0A1 19 years old was admitted to hospital on March 7, 2019 at 14.40 referred from Siti Mariam Hospital with a diagnosis of P0A1 19 years old pelvic peritonitis dd/intraabdominal bleeding The patient complained of lower abdominal pain felt disappear arising one day before hospitalization pain intensified accompanied by bleeding in the birth canal.

From physical examination obtained blood pressure: 90/70 mmHg, pulse: 112 times / minute, respiration: 28x / minute, temperature: 36.8^0 C. Anemic conjunctiva was found +/ +. Examination of the abdomen shows tense palpation with muscular defense throughout the abdominal region with normal bowel sounds.

The supporting examination was carried out by USG and laboratory examination. On ultrasound examination it was found that the uterus appeared slightly enlarged with a complex mass of adnexa (gestasional sac +) suspicious of adnexa with free fluid, suspecting that an ectopic pregnancy was disrupted.

In laboratory tests, it was found that Hb: 8.6 gr / dL, Leukocytes: 13,500 / mm3, Platelets: 258,000/mm^3 and Urine hCG test (+).

Based on these data, it is diagnosed as an ectopic pregnancy disturbed. Then an emergency laparotomy and blood transfusion of up to Hb > 10 mg / dL were decided. During intraoperative bluish peritoneum was found. Suspected ovarian pregnancy was found in the left ovary,

which broke during manipulation. There was bleeding from the left ovary. Further exploration of the left tube, right tube and right ovary in good condition. It was decided to do a partial left oophorectomy. And tissue was sent to the PA laboratory.

A month after surgery, the patient had vaginal bleeding with positive HCG and an acute abdominal condition from the peripheral hospital was referred to Prof. Dr. R.D Kandou Manado, a re-examination of the history of the disease, physical examination, and supporting examinations. On significant physical examination is anemic conjunctiva and there are acute signs of the abdomen. On examination of ultrasound and laboratory support, it was found that there was an impression of a disturbed ectopic pregnancy originating from the right adnexa with laboratory results showing signs of anemia. It was decided to do an emergency laparotomy again. During the surgery found an ectopic pregnancy in the right tube. Surgery on the left ovary showed signs of healing without bleeding. Tissue re-examined to anatomic pathology.

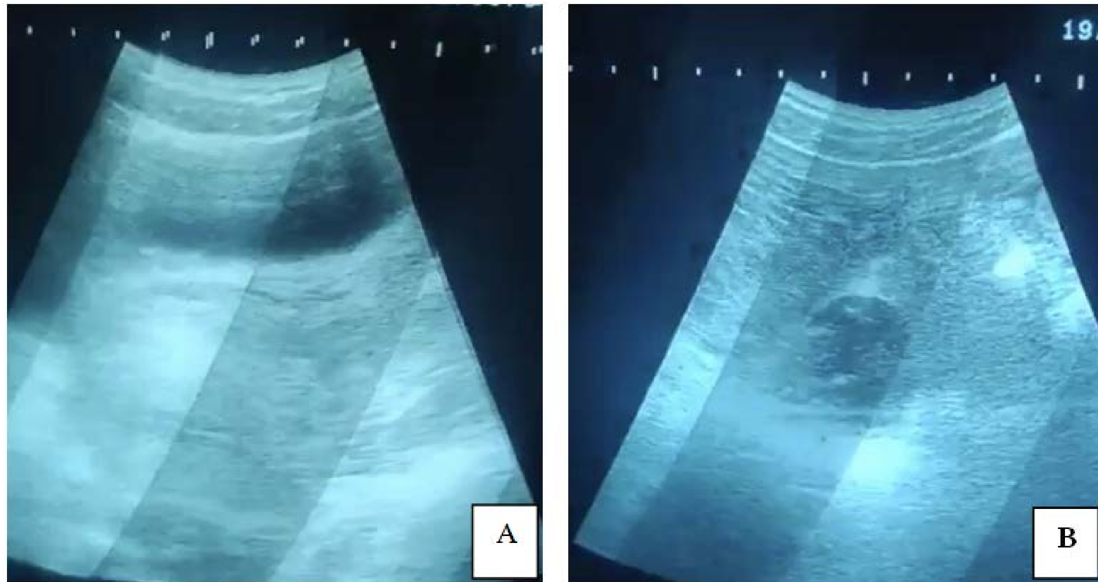

Figure 1. Ultrasonography of Ectopic Pregnancy (A) Empty uterus (B) Ectopic pregnancy

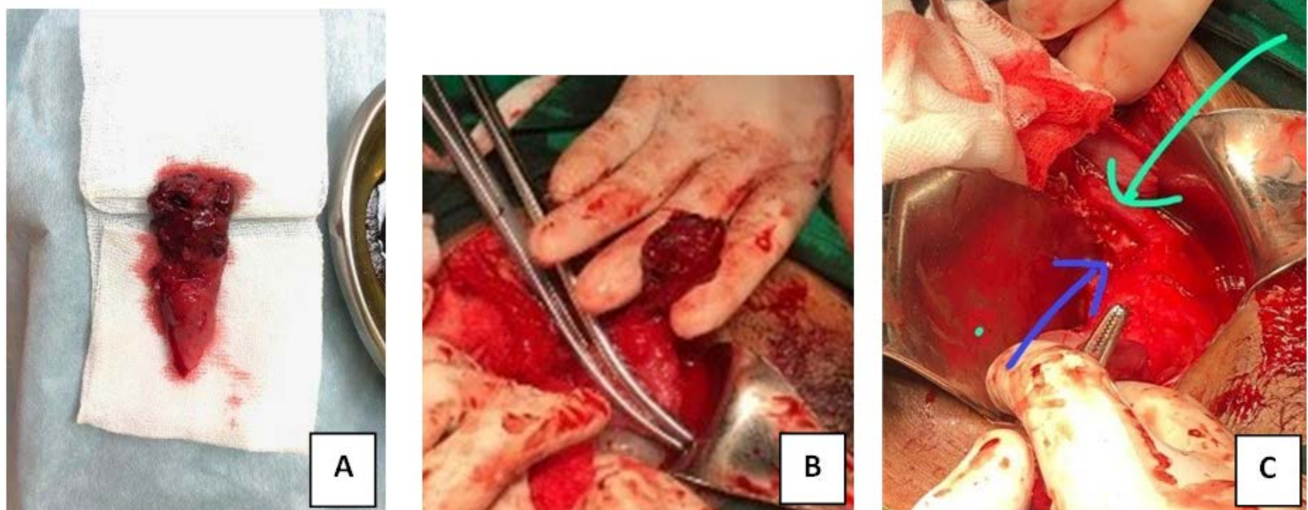

Figure 2. Intraoperative findings (A) The product of conception (B) Product of conception implanted on the left Fallopian tube (C) Normal structure of left fallopian tube and partial left ovary

3. Discussion

Heterotopic pregnancy is an intrauterine and extrauterine pregnancy on 1 occasion. Incidents are rare and very life-threatening where they are quite difficult to diagnose and are often misdiagnosed [2]. The incidence of heterotopic pregnancies is around 1: 30,000 events [2]. The incidence of ovarian pregnancy is increasing nowadays. This occurs due to the increased incidence of pelvic inflammatory disease and an increase in lifting use of Assisted Reproductive Techonologies (ARTs) [4].

Heterotopic pregnancy is an emergency and can be life threatening. The predisposing factors that cause heterotypic pregnancy and ectopic pregnancy are the same. First, ovulation factors whereas multiple ovulation occurs or the occurrence of ovulation hyperstimulation syndrome caused by drugs for ovulation induction. Second, it occurs due to in-vitro fertilization and embryo transfer. Third is tubal factor [1].

Ovarian pregnancy can be intrafollicular and extrafollicular. Intrafollicular ovarian pregnancy can also be said to be a failure of follicular expulsion where the fertilized ovum remains in the ovary but events are very rare. Extrafollicular ovarian pregnancy occurs where the ovum after fertilized migrates and implants in the ovary.

In this case, the patient was referred from a peripheral hospital with a diagnosis of Ectopic Pregnancy. When she was at the hospital referred the patient's condition in severe illness. On history taking, it is found that lower abdominal pain has disappeared since 1 day after was hospitalized, pain has intensified, bleeding from the birth canal, history of late menstruation and a history of vaginal discharge. From physical examination obtained the abdomen shows tense palpation with muscular defans throughout the abdominal region with normal bowel sounds.

The supporting examination was carried out by USG and laboratory examination. On the surgery, there is bleeding from the left ovary. Further exploration of the left tube, right tube and right ovary in good condition. It was decided to do a partial left oophorectomy. In diagnosing the occurrence of ovarian pregnancy, especially when preoperative is very difficult to do, most can be diagnosed at the time of intraoperative [4].

Symptoms experienced such as abdominal pain, vaginal bleeding, amenorrhea, and symptoms are very similar to tubal pregnancy where sometimes patients only feel lower abdominal pain [5].

Ovarian pregnancy can be diagnosed by Transvaginal Ultrasound if there is a thickened wall, an echogenic ring and an anechoic region in the middle located around or in the ovary. The difference with tubal pregnancy is that the echogenic ring is formed thinner than an ovarian pregnancy. We must be able to distinguish between ovarian pregnancy against the luteal corpus, where in ovarian pregnancy there is an increase in echogenicity in the ovarian stromal when compared to the luteal corpus [5].

A possible differential diagnosis in an ovarian pregnancy is rupture of the luteal corpus which can also cause massive bleeding. This is in accordance with existing studies that ovarian pregnancy both clinically and pathologically can be compared with tubal pregnancy, hemorrhagic luteal corpus rupture and brown cyst rupture [4].

Ovarian pregnancies usually rupture on the 40th day of gestational age, but some can last until the third trimester and even until the term pregnancy [1].

Estimated possibility in this case is acute abdomen due to rupture of the luteal corpus accompanied by tubal pregnancy. Superfetation can occur in this patient. Where there is an ovarian pregnancy and tubal pregnancy with different gestational ages, causing an ectopic pregnancy to occur twice as much. Where it has been reported in Japan there was 1 case with superfetation after ovarian stimulation and intrauterine insemination [5]. There was a case in Italy showed a superfetation due to an intrauterine insemination program and at 6 weeks gestation, ruptured tubal pregnancy and after that the patient gave birth to intrauterine pregnancy without the presence of intrauterine insemination without any complications [5].

In this case the patient denied having sexual relations after the first operation, so fertilization was less likely after the first operation.

The traditional treatment for ovarian pregnancy is oophorectomy and salpingo-oophorectomy [3].

In the existing study found 85.7% performed wedge recession, 8.2% performed oophorectomy and 4.1% performed salpingo-oophorectomy. Laparoscopy can increase the rate of conservative surgery [3]. If there are small lesions in the ovary, wedge resection or cystectomy can be performed, where if there are large lesions, then oophorectomy is performed [5].

The use of medical drugs can be given methotrexate or etoposide once administration can be given but is rarely done and is still controversial. But some of which are given to give good results. Although the administration of methotrexate is a treatment that does not use surgery, but the failure rate is very large and the risk of ovarian bleeding is very large [4].

4. Conclusion

In conclusion, ovarian pregnancy is very difficult to diagnose preoperatively, due to non-specific symptoms and unspecified risk factors, Rupture of the luteal corpus can be one of the most frequent differential diagnoses with ovarian pregnancy. Heterotopic pregnancy and superfetation also make the possibility of miss-diagnosed in patients with symptoms of disturbed ectopic pregnancy. The importance of investigations can help to diagnose all of these events. To be able to diagnose the pregnancy clearly tissue is very important to do anatomic pathology examination.

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