Case Series

Atypical presentations of ectopic pregnancies in a rural tertiary care hospital—A case series

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Abstract

In spite of advances in medical sciences, diagnosis of ectopic pregnancy eludes the clinician. In this case series, we describe three variants of life-threatening pregnancies. The successful management of these cases hinged on a strong index of suspicion and early detection. Heterotopic pregnancy is the occurrence of two pregnancies in different implantation sites at the same time, which is uncommon. Heterotopic pregnancy is uncommon in natural conception, despite its prevalence in assisted reproductive technologies. A strong index of suspicion and timely laparotomy was life-saving for the woman. Caesarean scar pregnancy is a form of ectopic pregnancy in which an aberrant pregnancy is implanted at the hysterotomy site of a prior caesarean procedure. Unexplained excessive bleeding at the time of medical termination of pregnancy prompted us to explore and make a timely diagnosis of caesarean scar pregnancy. Finally, tubal choriocarcinoma is a very aggressive, extremely rare trophoblastic tumour that can be either gestational or non-gestational. High serum beta HCG raised the suspicion and timely laparotomy confirmed the diagnosis. An alert clinician who takes timely decisions can avert life-threatening complications in these pregnancies.

Keywords: Caesarean scar pregnancy, choriocarcinoma, ectopic pregnancy, heterotopic pregnancy, tubal pregnancy

Introduction

Ectopic pregnancy diagnosis has long been a mystery. Ectopic and intrauterine pregnancies can occur concurrently most frequently in the ampullary part of the fallopian tube (80%).[1] This condition is called heterotopic pregnancy. About 1 in 30,000 spontaneous pregnancies are heterotopic pregnancies. Caesarean scar pregnancy is the most uncommon type of ectopic pregnancy caused by the implantation of the gestational sac into the fibrous tissue scar from prior caesarean surgery. It affects 1 in every 2000 pregnancies and accounts for 6% of ectopic pregnancies.[2]

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Choriocarcinoma is an uncommon malignant gestational trophoblastic tumour which rarely complicates an ectopic pregnancy. It affects 1 in 1.6 million healthy intrauterine pregnancies and 1 in 5333 tubal pregnancies.[3] Due to its similar presentation, tubal choriocarcinoma might be mistaken for an ectopic pregnancy.[4] It presents with amenorrhea, elevated beta human chorionic gonadotropin (BHCG) levels, vaginal bleeding and pelvic pain.

We are reporting three cases of atypical ectopic pregnancies with unusual clinical presentations. The first case was of an intrauterine pregnancy which turned out to be heterotopic pregnancy. The second case is of a woman who underwent medical abortion but had a severe haemorrhage. On further evaluation, she had a caesarean scar pregnancy. The third case is of a woman suspected of ectopic pregnancy but turned out to be tubal choriocarcinoma. Despite their unusual presentations, timely diagnosis and treatment saved these women. Therefore,

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we are reporting this case series to emphasise the importance of timely diagnosis and management of such clinical entities in pregnant women and practitioners should keep them as a differential.

Case 1

A 28-year-old primigravida who conceived after primary infertility of seven years presented to us with hypovolemic shock with 10+5 weeks of amenorrhea. The patient had received treatment for infertility and was taking Tab Letrozole and Clomiphene. She had a history of laparoscopic ovarian drilling. She had a family history of twin pregnancy. On examination, she was hemodynamically unstable with a pulse rate of 136/min and blood pressure of 90/50 mm Hg. She had pallor, cold and clammy peripheries. On abdominal examination, the lower abdomen was distended and tender. Upon admission, we suspected possible heterotrophic pregnancy or ruptured rudimentary horn or appendicular rupture. She underwent emergency exploratory laparotomy. A hemoperitoneum of approx 1500 ml of volume with clots was present. The uterine size was of 10 weeks with a normal right tube and ovary. There was left-sided ruptured ectopic pregnancy and left-sided partial salpingectomy was done and haemostasis was achieved. Four days later, the patient underwent dilatation and evacuation in view of missed abortion and was transfused with four units of whole blood, two units of fresh frozen plasma and two units of platelets. She was started on Inj. Ceftriaxone 1 gm iv in two doses, Inj. Ondansetron 4 mg IV, Progesterone vaginal pessary two times per day, Zonac suppository bd, Inj. Proluton 500 mg intramuscularly and Inj. β-HCG 5000 IU intramuscularly. Under all aseptic precautions, cleaning and draping were done. The anterior lip of the cervix was held with sponge-holding forceps, cervix dilated with serial numbers of Hegar dilators and the products of conception were removed using ovum forceps. Gentle curettage was done from all the walls of the uterus. She recovered and was asked to follow up after 15 days. She was prescribed Tab Femilon for 21 days [Figure 1].

Case 2

A 26-year-old woman with 7+2 weeks of amenorrhea with left adnexal ectopic pregnancy presented to us with pain in the abdomen. She was gravida 3 with 2 abortions. The urine pregnancy test was positive. She was married for 1.5 years. On examination, she had a pulse rate of 76/min, and a blood pressure of 108/78 mm Hg. Her abdomen was soft with no guarding, rigidity or rebound tenderness. Per vaginal examination revealed cervical motion tenderness and a 4 × 4 cm mass in the left fornix associated with tenderness. Her β-HCG levels were 2,25,000 mIU/ml. An exploratory laparotomy was planned. The abdomen was opened by a transverse incision in layers. A left adnexal mass 4 × 4 cm was found arising from the ampullary region of the left fallopian tube likely to be an ectopic pregnancy. The mass was adherent posteriorly to the sigmoid colon and the omentum. A blunt dissection was done. Left-sided partial salpingectomy was done. Bilateral ovaries were preserved. Her histopathology report was suggestive of left tubal choriocarcinoma likely arising from ectopic pregnancy. Decreasing trends of β-HCG levels (2425 mIU/ml) were reported post-operatively. The patient was started on weekly methotrexate therapy. She was advised of computed tomography of the abdomen, pelvis and head on follow-up [Figure 2].

Case 3

A 32-year-old P1L1A4 with previous LSCS was referred to our hospital for torrential bleeding post-dilatation and evacuation for missed abortion. She was married for 5 years and had a bad obstetric history. On examination, her general condition was poor and had a pulse rate of 120/min, and her blood pressure was 90/58 mm Hg. Her abdomen was soft and non-tender. Upon local examination, there was profuse bleeding through the cervical os. She underwent an exploratory laparotomy. The abdomen was opened in layers, and 100 ml of hemoperitoneum was present. On the right lateral uterine scar site of the previous LSCS, a bluish bulge was present which with oozing. The uterovesical fold was opened, the bladder pushed down, and a

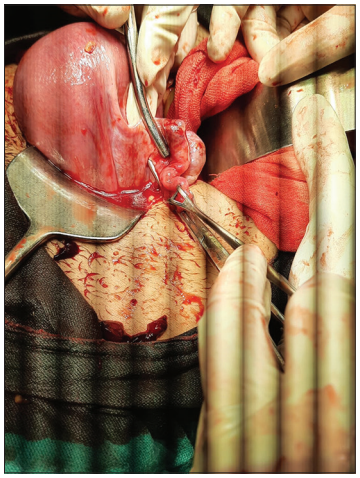

Figure 1: Heterotopic pregnancy

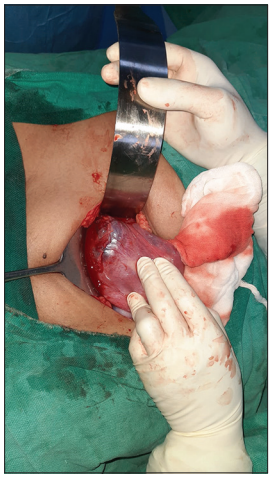

Figure 2: Caesarean scar pregnancy

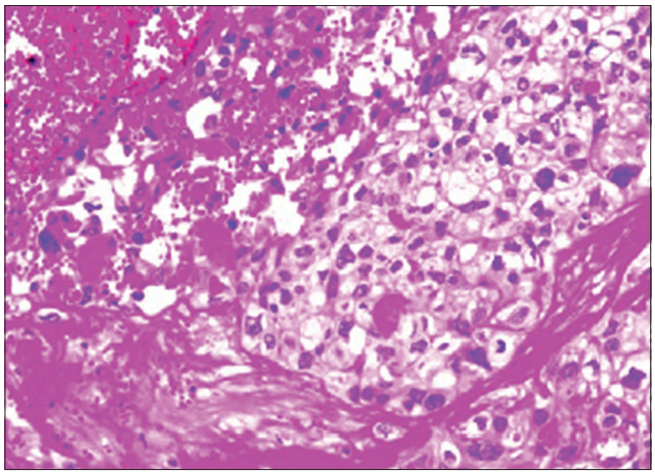

Figure 3: Choriocarcinoma

bluish bulge was incised. Pregnancy was found at the scar site, and retained products of conception were removed. Part of the scar with adherent bits of conception was excised and the sample was sent for histopathological examination. She was transfused with two units of blood. The uterus was sutured with Vicryl 1-0 by interlocking continuous sutures. Haemostasis was achieved, suction drain no 32 was kept in the pouch of Douglas, and the abdomen was closed in layers. The histopathology report was suggestive of haemorrhage and chorionic villi along with uterine myometrium, consistent with rupture of the uterus with products of conception [Figure 3].

Discussion

Choriocarcinoma is a rare aggressive form of the gestational trophoblastic disease, caused by neoplastic changes in the chorionic villi's epithelium. The prevalence of ectopic tubal choriocarcinoma is estimated at around 1 in 1.6 million healthy intrauterine pregnancies, making ectopic pregnancy-related choriocarcinoma extremely rare. A tubal choriocarcinoma might be mistaken for an ectopic pregnancy due to similar presentations like amenorrhea, high beta-human chorionic gonadotropin levels, vaginal bleeding and pelvic pain, as was the case in our instance. Its rarity, similar presentation as of ectopic pregnancy and small tissue size for histopathology examination make the diagnosis of choriocarcinoma difficult.[5] It can develop either from malignant tubal pregnancy or de novo. Histopathology is the gold standard for choriocarcinoma diagnosis. Columns of trophoblastic cells devoid of villous structures, the invasion of arteries and muscle tissue, along with severe necrosis and bleeding, are characteristics of choriocarcinoma. According to a study by Nakayama *et al.*, it is effective to identify the origin of tubal choriocarcinoma utilising p57kp2 immunostaining and DNA polymorphism analysis (gestational or non-gestational).[5] Fallopian tube choriocarcinoma can be suspected in patients whose MRI shows a solid segment with a honeycomb appearance and normal ovaries. Due to the significant potential for metastasis, chemotherapy is both necessary and helpful in its treatment.[6] Patients receiving extrauterine chemotherapy should take reliable birth control for at least a year after completing treatment. β-HCG monitoring is the most useful diagnostic method for tubal choriocarcinoma. When both an extrauterine (ectopic) and an intrauterine pregnancy form at the same time, it is known as a heterotopic pregnancy. In a heterotopic pregnancy, one fertilised ovum implants normally in the uterus and one fertilised ovum implants abnormally outside of the uterus. Similar to ectopic pregnancy, the main risk factors for heterotopic pregnancy are shared by the general population. Women participating in assisted reproductive programmes are at a higher risk of ectopic and heterotopic pregnancy due to recurrent ovulation, a higher rate of tubal malformation and/or damage, and technical difficulties with embryo transfer. The signs and symptoms of a heterotopic pregnancy can be similar to those of a typical intrauterine pregnancy as well as a ruptured ovarian cyst, corpus luteum or appendicitis as in our case. Ultrasonography and blood testing can be used to differentiate between these disorders. A salpingectomy or a salpingostomy is used to treat heterotopic pregnancy. Expectant care for ectopic pregnancies has proven beneficial in a few instances. The particular site of the ectopic pregnancy, together with the pregnant woman's clinical presentation and stability, all affect the treatment for heterotopic pregnancy. It is estimated that 0.6 2.5:10,000 pregnancies will result in heterotopic pregnancy.[7] The incidence of heterotopic pregnancy has significantly increased in women receiving ovulation induction. Despite having infertility, our patient conceived on her own during this round. Pregnancies resulting from assisted reproductive procedures including *in vitro* fertilisation (IVF) and gamete intrafallopian transfer (GIFT) have a greater rate of heterotopic pregnancy. The number of primary and repeat caesarean sections is on the rise and it has resulted in an increased frequency of ectopic pregnancies due to surgical scarring. A hysterotomy scar ectopic pregnancy has also been reported after myomectomy, uterine evacuation, previous abnormally adherent placentation, manual placenta removal, metroplasty, hysteroscopy and *in vitro* fertilisation. There are two categories of ectopic pregnancies brought on by hysterotomies. While type 2 grows in the uterine serosa, type 1 starts in the myometrium and moves toward the uterine cavity. The prognosis for type 2 pregnancies is poor since they can lead to uterine rupture, haemorrhage and maternal death. There is a chance of losing fertility in cases requiring a hysterectomy for significant haemorrhage. Pelvic pain and vaginal bleeding are frequent symptoms in the first trimester. Transvaginal ultrasound (TVUS) combined with transabdominal scans is the ideal test. In uncertain cases, magnetic resonance imaging (MRI) will confirm or exclude the diagnosis. Methotrexate has been used therapeutically or surgically to treat women. Surgical excision using hysteroscopy, laparoscopy or laparotomy along with vacuum aspiration can be used to remove the ectopic scar. It is challenging to identify and treat ectopic pregnancies brought on by a caesarean scar due to their rare occurrence. Early notice enabled the preservation of our women.

We were able to save the lives of these three women because to prompt identification and recognition of their uncommon appearances.

Conclusion

Heterotopic pregnancy is a rare entity and it should be suspected in every pregnant woman presenting with acute abdominal pain and adnexal abnormalities. To timely diagnose and treat heterotopic pregnancy, patients should be extensively checked with ultrasound and, if necessary, MRI. Caesarean scar pregnancies are becoming more common in the literature and increased caesarean section rates around the world will increase its overall incidence. Conservative management is appropriate if the patient requests it and should be delivered under constant supervision. Similarly, tubal choriocarcinoma should be suspected in an ectopic pregnancy with high levels of B-HCG. The importance of histopathological examination of tubal specimens cannot be undermined for timely diagnosis of this extremely rare aggressive but treatable cancer. It is important to consider the possibility of heterotopic pregnancy before inducing abortion.

Authors contribution

Nongrum SML was involved in the concept of the idea, literature search and preparation of manuscript and review.

Bhandari P helped in the literature search, preparation of the manuscript and reviewing of the draft.

Jain S contributed to the concept of the idea, manuscript editing and review for critical scientific facts.

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Conflicts of interest

There are no conflicts of interest.

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