

RECURRENT ABDOMINAL PREGNANCY: A RARE OCCURRENCE AND LITERATURE REVIEW***Bello S., Ukwu AE., Burodo AT., Shehu CE., Augie UI., Tori HM., Garba JA. and Nnadi DC.**

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ABSTRACT

Abdominal pregnancy is a very uncommon type of ectopic gestation but is associated with grave maternal and fetal outcomes. Its diagnosis and management presents a challenge to the physician and usually requires a high index of suspicion. We report a rare case of recurrent abdominal pregnancy twice in succession in the same patient. Bleeding per vaginam and persistent abdominal pain were the main presenting symptoms. At laparotomy, there was an intact gestational sac containing a demised fetus and a fistula on the anterior wall of the uterus which was repaired. Ignorance, poverty and poor health seeking behavior were the reasons for late presentation.

KEY WORDS: abdominal pregnancy, recurrence, fistula, Nigeria.**INTRODUCTION**

About 2% of all pregnancies occur outside the uterine cavity and more than 95% of these ectopic gestations occur within the fallopian tubes.^[1] Abdominal pregnancy is a rare form of ectopic gestation where implantation occurs within the peritoneal cavity. It accounts for 1- 4% of all ectopic pregnancies.^[1] The incidence varies from place to place but is more common in the developing countries.^[1,2] The worldwide incidence ranges between 1 in 33,000 and 1 in 10,200 deliveries.^[2] In Sokoto, Nnadi *et al* in a ten-year review of abdominal pregnancy reported an incidence of 3.1/10,000 deliveries.^[3] The incidence appears to be increasing in both the developed and developing countries. In the former, increasing use of assisted reproduction with embryo transfer has been associated with rising number of heterotopic pregnancies. In the developing countries, the high incidence has been attributed to increased risk of pelvic infections, limited diagnostic facilities for early detections of tubal pregnancies before secondary implantation in the peritoneal cavity and poor utilization of medical care by pregnant women.^[1]

Abdominal pregnancy is associated with high maternal mortality (up to 50%), and even higher perinatal mortality (40 – 95%). It is often associated with diagnostic difficulties.^[3]

Recurrence of abdominal pregnancy in the same individual in two consecutive pregnancies is a very rare phenomenon. We present a rare case of recurrent mid-trimester abdominal pregnancies in a 25-year old nulliparous woman who had exploratory laparotomy and repair of a utero-peritoneal fistula.

CASE REPORT

A 25-year old G3 P₀⁺² whose last pregnancy was five years prior to presentation was referred to the gynaecological emergency clinic of UDUTH, Sokoto, on account of bleeding per vaginam, lower abdominal pain and an ultrasound scan that showed an intra-abdominal pregnancy at 25 weeks with no cardiac activity.

The vaginal bleeding started six days before presentation, was initially scanty but later became profuse. The bleeding subsided after two days and thereafter the patient ceased to perceive fetal movements. There was no drainage of liquor. The lower abdominal pain was intermittent, colicky and was aggravated by locomotion. Systemic review was not significant.

She had a spontaneous first trimester miscarriage in her first pregnancy. There was no manual vacuum aspiration performed and the post abortion period was uneventful. In her second gestation, she presented to the hospital at about 26 weeks, with complains of absent fetal movements, fever, foul smelling vaginal discharge and passage of fetal tissues and bones per vaginam. She was managed as a case of abdominal pregnancy and had exploratory laparotomy. At surgery, there was a fistula present on the anterior wall of the uterus. An attempt at repair of the fistula was unsuccessful due to worsening haemorrhage and extensive amount of friable tissues surrounding the area. The patient received 4 units of blood and made good recovery. She was placed on Implanon sub-dermal implant on discharge. The patient defaulted on follow up appointments.

Physical examination revealed a fully conscious but anxious young woman. She was pale anicteric and not dehydrated. There was tachycardia (pulse rate 106 beats per minute) and the blood pressure was 110/70 mmHg. Abdominal examination showed a midline subumbilical incision scar that healed by primary intention. The fundal height was about 18 weeks size and fetal parts were easily palpable. Pelvic examination revealed normal vulva and vagina. The cervix was central, about 3cm long, firm and the cervical Os was closed. The adnexae and Pouch of Douglas were free. The examining gloved fingers were stained with altered blood. A diagnosis of recurrent abdominal ectopic pregnancy with demised foetus at 25 weeks gestation was made. The patient was counselled on her condition and was co-managed with general surgeons. Available investigations included Full blood count and differentials (WBC – $6.2 \times 10^9/L$, PCV – 37.9% and Platelet – $139 \times 10^9/L$), the bed-side clotting time was 4 minutes. The clotting profile revealed a PT of 21 seconds; PTTK was 47seconds and INR of 1.5. The electrolytes, urea and creatinine were within normal limits. The urinalysis showed normal findings. An abdomino-pelvic USS scan showed features suggestive of an intra-abdominal pregnancy with a demised foetus at 25 weeks separate from the uterus. There was no gross congenital abnormality seen. Three units of compatible fresh whole blood were made available for surgery.

She had exploratory laparotomy and repair of a uterine-peritoneal fistula under general anaesthesia. The intra-operative findings included a healed midline subumbilical incision scar, moderate adhesions between the anterior abdominal wall and the rectus sheath. There was an intact gestational sac containing a grossly normal dead female foetus within the peritoneal cavity as depicted in figure 1. The gestational sac weighed 180 grams and was attached to the uterine fundus and bowel. A uterine rent measuring 4 Cm by 3 Cm was present on the anterior wall of the uterus with remnants of placental tissues protruding from the uterine cavity as shown in figure 2. The left uterine tube and both ovaries were grossly normal but the right tube was oedematous. The estimated blood loss was about 300 mls. The post operative period was uneventful and she was placed on Implanon sub dermal implant on discharge.

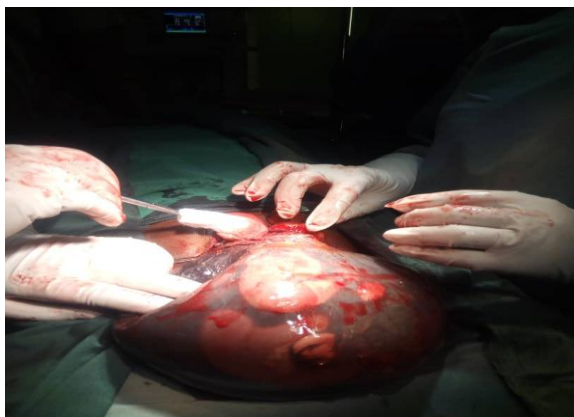


Figure 1: Intact gestational sac.

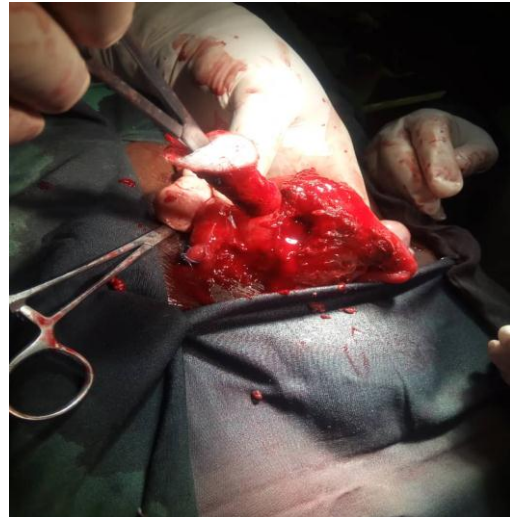


Figure 2: The uterus with a hole on the anterior wall.

DISCUSSION

Abdominal pregnancy can also be classified as primary or secondary depending on its implantation site. If it implants directly in the abdominal cavity and its organs (excluding the tubes and ovaries), it is referred to as a primary abdominal pregnancy. Secondary abdominal pregnancy occurs when the conception is extruded from the uterine tube and becomes secondarily implanted in the abdominal cavity. This occurs in 1 in 10 000 live births.^[4,5] In 1942, Studdiford described the first case of an early ruptured primary abdominal pregnancy and set the criteria for diagnosis of primary abdominal pregnancy.^[6] In 1968 Friedrich and Rankin^[7] modified the stipulated criteria to limit acceptable cases. They proposed that to be a true primary abdominal pregnancy, the pregnancy should be less than 12 weeks of gestation and the trophoblastic attachments should be related solely to the peritoneal surface.

The case presented was most likely a secondary abdominal pregnancy following extrusion of the gestational sac through the fistula on the anterior wall of the uterus. The fistula may have been congenital or acquired from perforation of the uterus by fetal bones following a late intrauterine fetal death in the second pregnancy. She was planned for repair of the fistula but she defaulted on follow up appointment, thus the recurrence would have been prevented. If an abdominal pregnancy is neither diagnosed nor treated and if the fetus dies intra-abdominally, the end result may be a calcified product of conception known as lithopedion. In 1971, Ghatak DP in Sokoto, Nigeria, reported a case of spontaneous rupture of a gravid uterus resulting in a secondary abdominal pregnancy and lithokelyphos.^[8] Intra-abdominal calcification instead of spontaneous re-absorption of a dead mummified fetus is an unusual complication of abdominal pregnancy. Fistula formation is common in situations where the pregnancy is advanced, demised, undiagnosed, and retained for a very long time. This was the situation in a case reported by Nnadi *et al* where the pregnancy was retained for 26

years.^[9] There was also a reported case of lithopedion that had been retained for 29 years.^[10,11] Fistula formation may result from penetration of fetal bones into maternal bowel, rectum, vagina, bladder, abdominal wall or the uterus as was observed in this patient. Undiagnosed abdominal pregnancy presenting as faecal fistula has been reported from Ile-Ife, Nigeria.^[12]

The incidence of recurrence of ectopic pregnancy is approximately 15 percent^[13] and any woman with a previous history of ectopic pregnancy is at high risk for recurrence. Thus a high index of suspicion is necessary. Due to its rarity, the incidence of recurrent abdominal pregnancies is not fully known however, a case has been reported by Simon Pereira *et al* in Mexico City in 1995.^[13] A similar case of recurrent abdominal pregnancy in the same individual has been reported from Amritsar in India.^[14] This followed a previous laparotomy for secondary abdominal pregnancy where the placenta was left undelivered. A case of abdominal pregnancy occurring on two consecutive occasions apparently as a result of a rent in the uterine fundus has also been reported by Tan *et al* in 1971.^[15] This is similar to the index case where a rent in the anterior wall of the uterus observed during a previous laparotomy which was left unrepaired led to recurrence.

When abdominal pregnancy is diagnosed, the widely accepted treatment is immediate laparotomy, for termination of pregnancy because of risk of maternal mortality due to massive intra-peritoneal haemorrhage from placental separation.^[16] One of the challenging problems during laparotomy for abdominal pregnancy is risk of massive haemorrhage when attempts are made to remove the placenta. It is advised that except the entire blood supply of the placenta can be secured with minimal risk to the patient, the placenta is best left in-situ. If left in-situ, there is the need to follow-up of the patient with serial monitoring of serum human chorionic gonadotropin (β -hCG) levels and sonograms (preferably colour Doppler) for placental involution. The use of Methotrexate to hasten placental involution and reabsorption has been reported.^[17]

In conclusion, we emphasize the need for early ultrasound scan for diagnosis and localization of the pregnancy, antenatal care, adequate management including hospital delivery to ensure a good outcome.

REFERENCES

1. Nwobodo EI. Abdominal pregnancy: a case report. *Ann Afr Med*, 2004; 3: 195-6.
2. Isah AY, Ahmed Y, Nwobodo EI, Ekele BA. Abdominal pregnancy with a full term live fetus: case report. *Ann Afr Med*, 2008; 7: 198-9.
3. D. Nnadi, E. Nwobodo, B. Ekele, Abdominal pregnancy in Usmanu Dan-Fodio University Teaching hospital, Sokoto: a 10-year review. *J Basic Clin Reprod Sci*, 2011; 1(1): 34-7.
4. Agarwal N, Odejinmi F. Early abdominal ectopic pregnancy: challenges update and review of current management. *TOG*, 2014; 3: 193-8.
5. Martin JN, McCaul JF. Emergent management of abdominal pregnancy. *Clin Obstet Gynecol*, 1990; 33: 438-47.
6. Studdiford W. Primary peritoneal pregnancy. *Am J Obstet Gynecol*, 1942; 44: 487-91.
7. Friedrich, EG, Rankin, CA. Primary pelvic peritoneal pregnancy. *Obstet Gynecol*, 1968; 31: 649-53.
8. Ghatak DP. Spontaneous rupture of a gravid uterus resulting in abdominal pregnancy and lithokelyphos formation. *J Indian Med Assoc*, 1991; 89(6): 172-3.
9. Nnadi D, Bello B, Ango I, Singh S. A 26-year-old retained demised abdominal pregnancy presenting with umbilical fistula. Case reports in Obstetrics and Gynaecology 2014; 1-3. Accessed on 31st July, 2019 available at <https://www.hindawi.com>
10. S. D. Costa, J. Presley, G. Bastert. Advanced abdominal pregnancy. *Obstet Gynecol Surv*, 1991; 46(8): 515-25.
11. N. M. Spirtos, S. M. Eisenkop, D. R. Mishell Jr. Lithokelyphos. A case report and literature review. *J Reprod Med Obstet Gynecol*, 1987; 32(1): 43-6.
12. A. R. K. Adesunkanmi, O. B. Fasubaa, O. Adeosun, C. O. Orji. Undiagnosed abdominal pregnancy masquerading as faecal fistula in simultaneous intra uterine and abdominal pregnancies—a case report. *West Afr J Med*, 2000; 19(2): 156-9.
13. Pereira S, Karchmer S, Medellin LJ, Villalobos AS, Henales JG. Recurrent abdominal pregnancy: a case report. *Ginecol Obstet Mex*, 1995; 63: 474-7.
14. Phillips C, Gurcharan K. Recurrent advanced abdominal pregnancy: a case report. *J Obstet Gynaecol India*, 1968; 109-111.
15. Tan KL, Vengadasalam D, Lean T. H. Recurrent abdominal pregnancy. *BJOG* 1971; 78(11): 1044-6.
16. Martin JN, Sessums JK, Martin RW. Abdominal pregnancy: current concepts of management. *Obstet Gynaecol*, 1988; 71: 549-57.
17. Ifenne DI, Shittu SO, Mandara MU. Advanced abdominal pregnancy with a live baby. *Trop J Obstet Gynaecol*, 1999; 16: 63-5.