

## Review Article

# Dangerous Fetal-Heartbeat-Positive Early Abdominal Pregnancies in Japan

Tatsuji Hoshino\*, Mayo Hino, Aya Matsubayashi, Ruriko Oyama, Noriko Ohtake, and Shinya Yoshioka

Department of Obstetrics and Gynecology, Kobe City Medical Center General Hospital, Japan

**\*Corresponding authors**

Tatsuji Hoshino, Department of Obstetrics and Gynecology, Kobe City Medical Center General Hospital, Minatojima-Minamimachi 2-1-1, Chuo-Ward, Kobe, Hyogo 650-0047, Japan, Tel: 81-78-302-4321; Fax: 81-78-302-7537; E-mail: hoshinot@kcho.jp

Submitted: 22 September 2015

Accepted: 23 October 2015

Published: 27 October 2015

ISSN: 2333-6439

**Copyright**

© 2015 Hoshino et al.

**OPEN ACCESS****Keywords**

- Abdominal pregnancy
- Fetal-heartbeat-positive
- Early pregnancy
- Dangerous

**Abstract**

There have been no recent reports of advanced abdominal pregnancies in more economically developed countries such as Japan, but fetal-heartbeat (FHB)-positive early abdominal pregnancies (EAPs) represent problematic ectopic pregnancies. We investigated this problem by performing a review of reported cases in Japan. We searched the literature using the Japanese medical-article research service Japana Centra Revuo Medicina, using the search terms "peritoneal pregnancy" and "abdominal pregnancy". We identified 86 EAPs (before 20 weeks) between 1983 and 2014. Twelve of these were FHB-positive, among which shock developed in six, and blood transfusions were performed in another five. Initial laparoscopy was performed in three patients but converted to laparotomy in one, while initial laparotomy was performed in eight patients. Potassium chloride and methotrexate were administered in one recent case. We conclude that FHB-positive EAP is a potentially dangerous condition. Diagnosis is usually performed late, the subsequent clinical course is usually abrupt, and surgery is often difficult in these cases.

**INTRODUCTION**

Ectopic pregnancies (EPs) comprise 1% of all pregnancies, with abdominal pregnancies (APs) accounting for only 1% of all EPs and 0.01% of all pregnancies. APs are thus considered very rare. Some fetal-heartbeat (FHB)-positive APs have an abrupt clinical course and undergo massive intra-abdominal bleeding with rupture of the pregnancy mass, because the pregnancy contents are not enclosed within the Fallopian tube or uterus. FHB-positive APs also have a rich vascular supply, similar to other EPs, thus increasing the risk of profuse bleeding during surgery. We review the reported cases of these dangerous, FHB-positive APs in Japan from 1983 to 2014 [1].

**METHODS**

The Centra Revuo Medicina Japana [1] review system is similar to PubMed. It is issued by the NPO Japan Medical Abstracts Society and includes all Japanese medical articles and congress abstracts. We searched this system using the terms "peritoneal pregnancy" and "abdominal pregnancy", and identified 95 APs reported in the Japanese literature from 1983 to 2014. These included nine advanced APs (AAPs) reported at 20 weeks' gestation or later, though none after 2002, and 86 early APs (EAPs) (i.e.  $\leq 19$  weeks'

gestation; latest 13 weeks) with sufficient clinical information, including our previously reported case [2].

**RESULTS**

There were 12 (12/86; 14%) FHB-positive EAPs [2-13]. Patient background, complaints, pregnancy method, shock status, gestational weeks, human chorionic gonadotropin (hCG) level, implantation site, surgical method, blood loss, blood transfusion, and other remarks are shown in Table 1.

Imanishi et al., [4] described a recent case of FHB-positive EAP at 8 weeks' of gestation that was managed successfully with potassium chloride injection into the fetal thorax and maternal intramuscular injection of methotrexate (MTX) without surgery, because of the risk of uncontrollable massive bleeding and rectal injury. In that case, magnetic resonance imaging indicated the abdominal pregnancy in the cul-de-sac with a blood supply from the rectal side. Kurihara et al., [5] reported a patient who underwent laparoscopic surgery and was managed by intraoperative autologous blood transfusion. Yamamoto et al., [6] also managed an abdominal pregnancy in the left vesico-uterine pouch by laparoscopy. We previously described a patient with an abdominal pregnancy in the right cul-de-sac case with massive

**Table 1:** Twelve cases of early abdominal pregnancy with fetal heartbeat.

Reference	Maternal age (y.o.) Gravidity (G) Parity(P)	First complaint, Time of first visit	Pregnancy method	Preoperative shock	Gestational age	hCG level (IU/L)	Implantation site	Operation	Blood loss Blood transfusion	Remarks
Imanishi et al. [4]	31 y.o. 1G0P	Positive pregnancy test, 8 wks, introduced	Spontaneous	No	8 weeks	Blood: 62,500	Cul-de-sac	Under transvaginal ultrasound guidance, 15% KCl 8 mL injection into fetal thoracic cavity and 80 mg (50 mg/m <sup>2</sup> ) MTX injection intramuscularly	No	Uneventful postoperative course, spontaneous vaginal delivery of 2810 g male baby, Apgar score 8/10 at 39 weeks
Kurihara et al. [5]	37 y.o. Not described	Lower abdominal pain, 7 wks, walk-in	Spontaneous	Not described	7 weeks	Not described	Cul-de-sac	Laparoscopy	1034 g intraoperative autologous blood transfusion, 400 mL	Multiple myoma
Yamamoto et al. [6]	31 y.o. 0G0P	Amenorrhea and abdominal pain, 8 wks, introduced	Spontaneous	No	8 weeks	Not described	Left vesico-uterine pouch	Laparoscopy	200 mL	
Hoshino et al. [2]	28 y.o. 0G0P	Lower abdominal pain, 9 wks, introduced	Spontaneous	Positive	9 weeks	Not described	Right cul-de-sac	Laparoscopy changed to laparotomy	2448 g Blood transfusion	
Fukuhara et al. [7]	29 y.o. 1G1P	Sudden lower abdominal pain, 10 wks, emergency transfer	Spontaneous	Not described	10 weeks	Blood: 26,875	Left vesico-uterine pouch	Laparotomy	600 g	
Fukuhara et al. [7]	33 y.o. 0G0P	Slight genital bleeding and lower abdominal pain, 13 wks, introduced	Spontaneous	Positive	13 weeks	Not described	Right tube, right broad ligament, right retroperitoneum	Laparotomy	2500 g Blood transfusion	
Kobayashi et al. [8]	33 y.o. 0G0P	Amenorrhea, 9 wks, walk-in	Spontaneous	Positive	9 weeks	Not described	Subserosa between myomas	Laparotomy	1680 g Blood transfusion, RCC 3U	
Kato et al. [9] Taninoue et al. [10]	34 y.o. 1G0P	Slight abdominal pain and lumbago, 11 wks, walk-in	Spontaneous	Positive	11 weeks	Not described	Left tube, left pelvic cavity	Laparotomy	3200 g Blood transfusion, RCC 8U, FFP 4U	
Oguchi et al. [11]	25 y.o. 0G0P	Lower abdominal pain and pre-shock status, 11 wks, emergency transfer	hMG/hCG/AIH pregnancy.	Positive	11 weeks	Not described	Anterior serosa of uterine corpus	Laparotomy	2852 g Blood transfusion	Concomitant intrauterine and extrauterine pregnancies. Continuing intrauterine twin pregnancy
Yoshida et al. [12]	27 y.o. 0G0P	Lower abdominal pain and respiratory distress, 5 wks, emergent transfer	hMG/hCG/AIH Pregnancy.	Not described	11 weeks	Urine: 13,000	Cul-de-sac	Laparotomy		Complicated with polycystic ovary syndrome, 3000 mL serous ascites present during surgery

Kajimura et al. [3]	28 y.o. OGOP	Left lower abdominal pain, 5 wks, admission	Clomid/hCG pregnancy.	Not described	5 weeks	Urine positive pregnancy test	Left posterior broad ligament	Laparotomy		
Takamizawa et al. [13]	25 y.o. 1G1P	Intraperitoneal bleeding, 9 wks, patient transfer	Spontaneous	Not described	9 weeks	Blood: 8,300	Sigmoid colon serosa	Laparotomy	1000 g Blood transfusion	Concomitant intrauterine and extrauterine pregnancies. Intrauterine pregnancy D&C

Y.O.: Years Old; WKS: Weeks of gestation; MTX: Methotrexate; RCC: Red Cell Concentrate mannitol adenine phosphate; FFP: Fresh Frozen Plasma; HMG: Human Menopausal Gonadotropin; hCG: Human Chorionic Gonadotropin; AIH: Artificial Insemination by Husband; D&C: Dilatation and Curettage

preoperative bleeding as a result of detachment of the pregnancy contents [2]. Fukuhara et al., [7] reported an abdominal pregnancy in the left vesico-uterine pouch in which a 600-mL hemorrhage and coagulate including the fetus and placenta was observed at laparotomy. They also described a patient with a secondary abdominal pregnancy near the right tube who developed shock before surgery, and who had 2,500mL blood in the abdominal cavity, while Kobayashi et al.,[8] described an EAP in the anterior uterine serosa between myomas in a patient who developed preoperative shock, who had 1680g blood in the abdominal cavity. Kato [9] and Taninoue et al., [10] reported a patient with a secondary AP in the left pelvic cavity from the left tube who suddenly developed shock after changing from a supine to a right-lateral position for epidural-catheter insertion. Oguchi et al., [11] described a patient with an intrauterine twin pregnancy and an abdominal pregnancy after ovulation induction and artificial insemination with her husband's semen. Yoshida et al., [12] described an EAP complicated by ovarian hyper stimulation syndrome associated with the use of combined human menopausal gonadotropin (hMG) and hCG. Kajimura et al., [3] reported an EAP in the left posterior broad ligament relieved by laparotomy, and Takamizawa et al., [13] described a case of concomitant intrauterine and extra uterine abdominal pregnancy in the Sigmoid colon serosa.

Nine of the reported cases were spontaneous pregnancies and three were conceived after ovulation induction. Eight patients (8/12; 66.7%) presented with abdominal pain as their first complaint. The gestational ages of the fetuses ranged from 5–13 weeks (median: 9 weeks), and the implantation sites varied, but none were far from the pelvic cavity. Most cases (7/12; 58.3%) progressed rapidly to shock or received a blood transfusion before surgery. Six of eight patients with APs later than 9 weeks' gestation (75%) were in shock or received a blood transfusion before surgery. Successful removal was achieved by laparotomy in most cases (8/12), though removal was achieved laparoscopically in two cases and initial laparoscopy was converted to laparotomy in one case [2]. Blood loss ranged from 200–3,200 g except in three cases, and seven of the 12 cases (58.3%) received blood transfusions. Concomitant intrauterine and extra uterine pregnancies were found in two cases [11,13], both of whom had conceived following hMG/hCG and artificial insemination by husband. One case was complicated by polycystic ovary syndrome, and 3000 mL serous as cites was detected during surgery.

## DISCUSSION

AAPs ( $\geq 20$  weeks' gestation) have formed the focus of most discussions of APs, and account for 49%–81% of all APs [14–17]. However, we found no reports of AAPs in Japan since 2002. Masukume et al., [18] described 38 cases of AAPs resulting in live births from 16 countries, including 10 from Nigeria, five from India, four from the United States of America, three each from South Africa, Bangladesh, and Tanzania, and one each from China, Malaysia, Chile, Saudi Arabia, Ghana, Trinidad and Tobago, Pakistan, Thailand, South Korea, and Zimbabwe. Many of these countries are considered to be developing countries. Nunyalulendho et al., [19] described that a total of 163 AAPs have been reported in 22 reports from 13 countries since 1946, according to a literature search and PubMed review. We think that potential AAPs are likely to be diagnosed and terminated during early pregnancy in resource-rich countries, accounting for the apparently higher incidence in non-industrialized countries, which lack medical facilities, medical personnel, and convenient ultrasound facilities for pregnant women [20].

Poole et al., [21] reported 225 early abdominal pregnancies in the English literature between 1965 and 2009, with a mean gestational age at treatment of 10 weeks 0 days, a mean maternal age of 29.7 years, and an average treatment-associated blood loss of 1,450 mL. Fetal cardiac activity was observed in 40 (40/225; 18%) patients, which was similar to the present report based on the Japanese literature, which found FHBs in 12 of 86 cases of EAPs (14%).

FHB-positivity, hCG  $> 10,000$ U/L, and pregnancy mass  $> 4$ cm are considered to be difficult management conditions in EPs. These conditions coincide with rich fetal vascularity, and larger chorion and pregnancy masses. General APs have been described extensively [6,14–18]. As in tubal EPs, FHB-positive APs are highly vascularized, but they are not enclosed by the uterus or Fallopian tube. Detachment of the pregnancy content may occur as a result of only slight movement or pressure, and subsequent bleeding may be hard to control during surgery, making FHB-positive APs a recognized dangerous form of AP.

In our series of 12 cases, abdominal pain was the first complaint in eight patients (66.7%), suggesting an abrupt clinical course. Our series also included nine spontaneous pregnancies, under which circumstances patients tend to only present to a doctor relatively late. Three cases were complicated, including two patients with concomitant intrauterine and extra uterine

pregnancies and one with polycystic ovary hyper stimulation syndrome, both of which may have delayed the diagnosis. The diagnosis of AP is difficult because the pregnancy content is not near the uterus or Fallopian tubes and does not obstruct the intestine resulting in gas echoes. In our series, the median gestational age at diagnosis and treatment was 9 weeks, which is late compared with 5–6 weeks for cesarean scar pregnancy, cervical pregnancy, and tubal pregnancy. Later pregnancies have increased vascularity compared with earlier pregnancies, and feticide was achieved conservatively in a recent FHB-positive case [4] using potassium chloride and intramuscular MTX administration, without laparotomy or laparoscopy, to avoid massive intraoperative bleeding. We think there is some possibility of becoming one of treatment option.

## REFERENCES

1. Japana Centra Revuo Medicina.
2. Hoshino T, Hayashi N, Miyamoto T, et al. A case of fetal heart beat-positive abdominal pregnancy with massive intra-abdominal bleeding and review of literature in Japan. *Obstetrics and Gynecology*. 2013; 80: 117–124.
3. Kajimura H, Ishimaru T, Murakami T, Yamabe T. Two cases of abdominal pregnancy. *Obstetrics and Gynecology*. 1989; 56: 127–132.
4. Imanishi T, Miyamoto T, Furukawa T, et al. A case of abdominal pregnancy diagnosed in the beginnings of pregnancy and managed conservatively without operation. *Kanto Journal of Obstetrics and Gynecology*. 2014; 51: 125–131.
5. Kurihara M, Takayama N, Kamata M, et al. Five cases of emergent laparoscopic surgery with intraoperative autologous blood transfusion without blood transfusion. *Okinawa Journal of Obstetrics and Gynecology*. 2014; 36: 73–79.
6. Yamamoto N, Takeuchi R, Iduchi D, et al. Four cases of abdominal pregnancy managed with laparoscopic operation. *Japanese Journal of Gynecological and Obstetric Endoscopy*. 2013; 1: 141–147.
7. Fukuhara R, Sakamoto T, Tanaka K, et al. Three cases of abdominal pregnancy. *Journal of the Aomori Society of Obstetricians and Gynecologists*. 2005; 19: 99–103.
8. Kobayashi Y, Iemura K, Kanemura Y, Iwaki S, Moritake T, Ogura H. A case of uterine serosal pregnancy. *Tokyo Journal of Obstetrics and Gynecology*. 2003; 52: 31–33.
9. Kato C, Ogino Y, Kato K, Fujita H, Okumura J, Murakami A. One case of abdominal pregnancy. *Medical Journal of Kyoto Second Red Cross Hospital*. 1999; 20: 55–62.
10. Taninoue K, Mochizuki N, Sakai Y, Ishizaka N, Yoshida S. One case of abdominal pregnancy with massive bleeding during epidural tube insertion. *Medical Journal of Kyoto Second Red Cross Hospital*. 1999; 20: 70–73.
11. Oguchi K, Hanatani K, Sakaibara M, Gotou S, Sawaki W, Sakai K. Concomitant heterotopic abdominal pregnancy complicated with intrauterine twin pregnancy after induction of ovulation. *Obstetrics and Gynecology*. 1998; 8: 1089–1092.
12. Yoshida T, Yoshioka S, Mise H, Kozaki S, Yoshioka J, Morishita Y. One case of abdominal pregnancy complicated with ovarian hyperstimulation syndrome. *Medical Journal of Japanese Red Cross Society Wakayama Medical Center*. 1993; 11: 97–102.
13. Takamizawa M, Mitsuya K, Takahashi K, et al. One case of concomitant intrauterine and abdominal pregnancy. *Obstetrical and Gynecological Practice*. 1987; 8: 1169–1173.
14. Molinaro TA and Barnhart KT. Abdominal pregnancy, cesarean pregnancy, and heterotopic pregnancy.
15. Cunningham FG, Leveno KJ, Bloom SL, et al. Ectopic pregnancy. In: Cunningham FG, Bloom SL, Spong CY, et al, editors. *Williams Obstetrics* 24th edition. New York: McGraw Hill Medical; 2014; 377–395.
16. Stovall TG. Early pregnancy loss and ectopic pregnancy. In: Berek JS, editor. *Berek & Novak's Gynecology* 14th edition. Philadelphia: Lippincott Williams & Wilkins. 2007; 601–629.
17. Fylstra DL. Ectopic pregnancy not within the (distal) fallopian tube: etiology, diagnosis, and treatment. *Am J Obstet Gynecol*. 2012; 206: 289–299.
18. Masukume G. Live births resulting from advanced abdominal extrauterine pregnancy, a review of cases reported from 2008 to 2013. *WebmedCentral Obstetrics and Gynaecology*. 2014; 5.
19. Nkusu Nunyalulendho D, Einterz EM. Advanced abdominal pregnancy: case report and review of 163 cases reported since 1946. *Rural Remote Health*. 2008; 8: 1087.
20. Aliyu LD, Ashimi AO. A multicenter study of advanced abdominal pregnancy: a review of six cases in low resource settings. *Gynecol Obstet Invest*. 2012; 74: 249–260.
21. Poole A, Haas D, Magann EF. Early abdominal ectopic pregnancies: a systematic review of the literature. *Gynecol Obstet Invest*. 2012; 74: 249–260.

### Cite this article

Hoshino T, Hino M, Matsubayashi A, Oyama R, Ohtake N, et al. (2015) Dangerous Fetal-Heartbeat-Positive Early Abdominal Pregnancies in Japan. *Med J Obstet Gynecol* 3(5): 1069.