

CLINICAL CASE REPORT

Advanced abdominal pregnancy: case report and review of 163 cases reported since 1946

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Submitted: 17 September 2008; Resubmitted: 28 October 2008; Published: 1 December 2008

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Rural and Remote Health 8: 1087. (Online), 2008

Available from: <http://www.rrh.org.au>

ABSTRACT

Context: Though relatively rare, advanced abdominal pregnancy (AAP) can have dramatic and catastrophic consequences for the foetus and the mother. Difficult to diagnose pre-operatively, AAP presents special challenges to the physician working in remote areas with limited resources for diagnosis and management.

Issue: Case report: A case of AAP received in our hospital in Kolofata, Cameroon, is presented and followed by a review of 163 other cases reported from 13 countries since 1946.

Lessons learned: A physician working in a remote district with an active maternity service should expect to encounter several cases of AAP during his or her working lifetime. Pre-operative diagnosis of AAP allows time for thoughtful preparation of the patient, family and medical team; however, to be diagnosed, AAP must first be considered. Diagnosis requires a high index of suspicion, and this should be triggered by any of a number of symptoms and signs reported in many cases of AAP. An unusual echographic appearance of the placenta was present in our case and prompted a more thorough investigation that confirmed the diagnosis. This finding has been reported by others and should be added to the list of signs and symptoms that might lead to a diagnosis of AAP in time to save the surgeon from an unpleasant and dangerous surprise.

Key words: abdominal pregnancy, advanced abdominal pregnancy, advanced ectopic pregnancy, advanced extrauterine pregnancy, Cameroon, ectopic pregnancy, extrauterine pregnancy.



Context

Advanced abdominal pregnancy (AAP) is defined as a pregnancy of over 20 weeks' gestation with a foetus living, or showing signs of having once lived and developed, in the mother's abdominal cavity¹⁻⁴. Often undiagnosed prior to operative intervention, and prone to dramatic complications, advanced abdominal pregnancy presents special challenges to the physician working in remote areas with limited resources for diagnosis and management.

Kolofata health district covers an 850² km rural zone in the Far North Province of Cameroon. Its population of 115 274 consists primarily of subsistence level farmers and herders. Illiteracy is very high and fewer than 5% of women can read or write. Health care is provided by six primary care health centers and the 100 bed district hospital. The hospital receives approximately 26 000 patients a year in consultation.

We present a case of AAP received in the Kolofata District Hospital, and follow with a review of 163 others reported throughout the world since 1946.

Issue

Case report

A 30 year-old woman, gravida 5 with four living children and no history of abortion, presented to her local health centre with amenorrhea of 9 months' duration and a chief complaint of abdominal pain and absent foetal movements for 7 days. The patient's third and youngest living child was 12 years old. The birth of her fourth child, who died at 10 months of probable pneumonia, had been followed by a 10 year period of infertility before this fifth pregnancy. There was no history of pain prior to this episode, and she had had no vaginal bleeding during the pregnancy. Three previous antenatal visits were made to the health center. Each visit consisted of an interview and clinical examination

by a nurse or midwife, who noted an apparently normal pregnancy. Ultrasound examination, unavailable in rural health centers in Cameroon, was not performed.

Detecting a transverse lie and no foetal heartbeat, the consulting nurse referred the patient to our hospital.

On examination she was in no evident pain or distress. She weighed 62 kg and had a blood pressure of 110/70 mmHg. Her conjunctivae were pale (hematocrit 26%; normal range 38-46%). Her abdominal circumference was 89 cm. An abdominal mass presumed to be the uterine fundus was palpated at 31 cm above the symphysis pubis (Fig1). The foetal presentation was not clear. No foetal heart sounds were heard. Ultrasound examination revealed a partially decomposed full term male foetus lying obliquely, the head in the mother's right upper quadrant, and a large globular placenta of abnormal echogenicity: multiple fluid-filled spaces in the placenta gave an unusual picture not unlike that of molar tissue. The mother's bladder was empty but a mass suggestive of a possibly empty uterus adjacent to the foetus was detected. A diagnosis of possible mixed-molar or abdominal pregnancy was made. To rule out the latter, the ultrasound examination was repeated several hours later with the mother's urinary bladder full. This time, the non-gravid uterus was clearly visible posterior to the bladder and distal to the externally implanted placenta and foetus. The patient was taken to the operating room.

An exploratory laparotomy under ketamine anaesthesia was performed through a sub-umbilical median incision that extended 3 cm above the umbilicus. The thickened peritoneum was entered and a partially macerated male foetus lying in a prone and slightly oblique position was revealed (Fig2). No putrid odour was noted. The foetal head was in contact with the maternal liver, stomach and transverse colon. The breech was astride the placenta, the maternal surface of which cupped the external wall of the uterine fundus. The placenta, enveloped in membranes and coated with meconium, was friable. Placental tissue



penetrated the mesentery posteriorly and the uterus inferiorly. The foetus was removed without difficulty. Extraction of the placenta incited a profuse haemorrhage which was controlled by prolonged manual pressure, sutures and a pedicle graft of the omentum. Although most of the placenta was removed, placental debris adherent to surrounding structures was left in place. The fallopian tubes, ovaries and uterus were macroscopically normal. Intestinal motility and the mesenteric arterial pulse were monitored throughout the procedure and remained normal. A peritoneal drain was inserted before closure. Blood loss was estimated to be 500 mL. No transfusion was given. The baby weighed 3000 g and had no evident deformity (Fig3). The mother was given heparin for 24 hours and ceftriaxone for 3 days, followed by penicillin and gentamycin for 4 days. She was discharged well on post-operative day 8 and was in excellent health when last seen for routine follow up 3 months later.



Figure 1: Normal external appearance of woman carrying an abdominal pregnancy at term.

Review of published cases

A journal search and PubMed review was completed. The journal search consisted of articles published pre-1950 (not listed in PubMed) compiled by a manual search of old journals for relevant papers, guided by references listed in papers post-1950 found through PubMed. Altogether, 163 cases of AAP described in 22 reports from 13 countries

since 1946 (Table 1 and Table 2) suggested that while patterns emerge, there is no typical case of AAP, and controversies remain concerning optimal management.



Figure 2: Breech of partially macerated male foetus lying prone astride the placenta, outside the intact uterus.



Figure 3: Newborn and placenta.

Table 1: Characteristics of 163 cases of advanced abdominal pregnancy reported from 13 countries since 1946¹⁻²²

Reference	Year of publication	Country	Total hospital deliveries during study period	Total cases of AAP during study period	Diagnosed pre-operatively	Abnormal lie or presentation	Abnormal placenta on ultrasound	Mothers transfused	Mothers with post-operative complications	Foetal or perinatal deaths	Maternal deaths
5 [†]	1946	USA	41 634	13	4	6	NA	8	NA	11	2
6 [‡]	1948	USA	NA	13	NA	NA	NA	NA	NA	8	4
7	1951	USA	41 941	9	NA	NA	NA	NA	NA	7	2
8	1953	Hong Kong	60 045	12	7	8	NA	NA	NA	11	1
9	1954	South Africa	NA	1	1	1	NA	1	1	0	0
10	1956	USA	68 394	8	5	NA	NA	NA	NA	5	0
1	1957	USA	31 616	10	6	8	NA	10	7	7	1
2	1962	USA	177 530	14	NA	NA	NA	NA	NA	12	2
11	1977	USA	87 239	4	NA	NA	NA	NA	NA	3	2
12	1982	USA	70 954	5	2	NA	NA	4	4	3	NA
13	1982	Saudi Arabia	102 000	10	3	7	NA	NA	4	5	2
14	1985	USA	120 000	9	NA	NA	NA	5	NA	8	0
15	1989	Zimbabwe	218 500	23	NA	NA	NA	NA	NA	19	0
16	1989	Papua New Guinea	NA	2	NA	NA	NA	NA	NA	NA	NA
3	2000	Ghana	62 348	13	5	9	NA	12	7	9	2
17	2003	Jordan	NA	1	0	1	1	1	NA	0	0
18	2004	UK	NA	1	0	1	NA	1	1	0	0
19	2005	Nigeria	NA	1	1	1	1	NA	NA	NA	NA
20	2005	Australia	NA	1	1	NA	1	NA	1	NA	NA
21	2007	Tanzania	3000	4	1	NA	NA	NA	0	3	0
22	2007	Taiwan	NA	1	NA	NA	NA	NA	NA	NA	NA
4	2008	USA	NA	8	5	NA	2	7	4	3	NA

AAP, advanced abdominal pregnancy; NA, data not available.

[†]Maternal mortality figure was revised in reference 2 - the corrected figure is reported here; [‡]AAP defined as >28 weeks.

Table 2: Summary of 163 reported cases of advanced abdominal pregnancy^{1-15,17-21}

Indicator	Value	<i>N</i>	References
Number of deliveries per AAP [†]	8099	1085 201/134	1-3,5,7,8,10-15,21
Number of deliveries per AAP, industrialized country	8879	639 308/72	1,2,5,7,10-12,14
Number of deliveries per AAP, non-industrialized country	7192	445 893/62	3,8,13,15,21
AAP diagnosed pre-operatively	45%	36/80	1,3-5,8,9,11,12,17-21
AAP with abnormal lie	68%	42/62	1,3,5,8,9,13,17-19
AAP with echographically unusual placenta	45%	5/11	4,17,19,20
Foetal or perinatal death	72%	114/158	1-15,17,18,21
Maternal death	12%	18/145	1-3,5-11,13-5,17,18,21
Mothers transfused	80%	49/61	1,3-5,9,12,14,17,18
Mothers with post-operative complications	55%	29/53	1,3,4,9,12,13,18,20,21

AAP, advanced abdominal pregnancy.

[†]All data concerns hospital deliveries.

Data: Because complete descriptions are not uniformly available in all reported cases and case series, the denominator varies for each of the indicators listed. Calculations are necessarily based only on data explicitly stated in the article. In articles where foetal lie, for example, is not mentioned, the cases presented in that paper are considered in neither the numerator nor the denominator of the indicator ‘AAP with abnormal lie’.

Incidence: In the series reviewed here, one AAP occurred for every 8099 hospital deliveries. The incidence was 19% higher in non-industrialized countries than in industrialized countries.

Diagnosis: The diagnosis of AAP is difficult and is made on the basis of history, physical examination and imagery. Echographic evidence of a non-gravid uterus alongside a foetus is diagnostic, but this will be seen only if the operator purposefully seeks the uterus on ultrasound examination. A pre-operative diagnosis of AAP is missed more often than it is made: only 45% of cases described here were diagnosed pre-operatively. The insertion of a balloon catheter in the uterus during ultrasound may help clarify an ambiguous image¹⁵. In any case, a high index of suspicion is crucial, and this should be triggered by any of the following clues:

- a history of bleeding or excessive abdominal pain during the first trimester
- a history of previous abortion or pelvic surgery
- a history of infertility
- bleeding or non-labour abdominal pain during the third trimester
- maternal declaration of cessation of foetal movements
- perception on the part of the mother or the physician that ‘something is not right’
- abnormal foetal lie
- displaced cervix or abdominal mass palpated apart from the foetus
- unusual echographic appearance of the placenta
- failed induction.

Management: Because perinatal death may result from either prematurity or prolonged gestation in a compromised environment, the decision about when to intervene in the case of a live baby should be made in consultation with the mother. Once the foetus has reached a viable age, there is little reason to delay delivery. Regardless of timing, the mother’s own safety will be best assured by careful monitoring, foresight and pre-operative preparation.

The principal controversy concerning management of AAP is whether or not to remove the placenta. Because the



abnormally implanted placenta's blood supply is diffuse and often unidentifiable, attempts to remove it can incite catastrophic haemorrhage. Measures taken to control this haemorrhage during surgery risk compromising the blood supply of other organs. A placenta left in situ might resorb spontaneously but if it does not, the risk of infection, necrosis, and the need for a second surgery is considerable. Most authors agree that the placenta should be removed provided its blood supply is identified and can be ligated without damaging other organs. If the blood supply cannot be identified and safely ligated, the placenta should be left in place and the patient followed for possible complications. The use of methotrexate to shrink a placenta left in situ has been largely discredited. Delaying removal of a stillbirth to give time for the placenta's blood supply to shrink remains controversial.

Outcome: Foetal and maternal complications in AAP are the rule. In the series presented here, foetal or perinatal mortality was 72%, and pressure deformities were common among survivors. Maternal post-operative complications, most notably haemorrhage and infection, occurred in more than half of all patients, and over three-quarters required blood transfusion. Maternal mortality was 12%.

Lessons learned

Conclusions

Advanced abdominal pregnancy is rare, but given an overall incidence of one AAP for 8099 hospital deliveries, a medical officer responsible for a district population of 100 000 might expect to encounter an AAP every few years if every pregnant woman near term presented for prenatal care or delivery.

The case reported here has many features of other cases reviewed: the mother, pregnant and at term following a ten-year period of secondary infertility, presented with abdominal pain, absent foetal movement and abnormal foetal lie. The unusual echographic appearance of the placenta was

key to suspecting that the pregnancy was extraordinary, and this suspicion prompted the repeat ultrasound examination, this time with a filled bladder to secure a higher quality result. The second examination confirmed the non-gravid state of the uterus and permitted the diagnosis of AAP. Abnormal echographic appearance of the placenta has not been cited by other authors as diagnostically important in cases of AAP, but it was described in several cases reviewed here and should be considered helpful.

For the physician in a remote area, where capacity to respond to potentially disastrous surprises is limited by lack of personnel and other resources, the ability to diagnose AAP pre-operatively is especially important. Pre-operative diagnosis gives the medical team time to review management and possible complications of AAP. The patient and family can be briefed. Additional personnel, adequate supplies of blood and resuscitation equipment can be made available. For these reasons, whenever a pregnant woman at term presents with any of the diagnostic criteria listed above, a thorough investigation, complete with clinical and, if possible, ultrasonographic assessment of the uterus and the placenta, in addition to the foetus, should ensue to assure that the baby is indeed within the uterine cavity. Regardless of symptoms and signs appreciated retrospectively, if the possibility of abdominal pregnancy is not considered at presentation, abdominal pregnancy will not be diagnosed.

Ultrasound examination, still insufficiently accessible in many remote areas, is a valuable tool and should be part of the minimal standard of care for every pregnant woman.

The variable definition of AAP and the changing accessibility of imaging technology over time and place may introduce error when comparing case studies or drawing statistical conclusions. More than 90% of the cases presented here were reported prior to 1990, when ultrasound was rarely available in rural settings in the developing world. It may be assumed that many potential cases of AAP in resource-rich countries are diagnosed and terminated in early pregnancy, resulting in the higher incidence seen in non-industrialized



countries. Finally, publication bias may have resulted in an underestimation of mortality, as the thrill of success in these unusual cases naturally tends to make one more eager to publish.

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