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Research Letter

Acute uterine inversion at cesarean followed by combined “anaphylactoid syndrome” sequence and uterine atony

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Dear Editor,

A 36-year-old healthy G1P1, at 41 weeks, was admitted for induction of labor due to oligohydramnios. Six hours after a prostaglandin induction was initiated, the fetal heart tracing showed sustained bradycardia without uterine “hyperstimulation” or “hypertonus” and an emergency cesarean was performed under spinal anesthesia; single attempt at level L4. After a low transverse uterine segment incision, a 3,112 g male was delivered Apgar 9¹/9⁵. No signs of placental abruption were observed. The cesarean procedure was continued with routine gentle traction of the umbilical cord; during this step a complete inversion of the uterus was witnessed and an immediate facile re-inversion performed within 10 s followed by completion of placenta removal. Within 1 min of the uterine replacement the O₂ saturation dropped from 96%–64%, followed by a drop in blood pressure from 140/75 to 80/40 and tachycardia up to 150 bpm. Immediate crystalloid resuscitation was started. Despite adequate recuperation of heart rate and blood pressure, uterine atony with excessive bleeding and clinical coagulopathy were diagnosed and pharmacologically managed by Oxytocin IV drip 10 IU, Methylergonovine maleate IM 0.2 mcg and prostaglandin F2 alpha IV 5 mg 5 mg/ml; 2 boluses followed by B-Lynch compression suture. Blood workup released to the lab at the initiation of the event showed low fibrinogen (<280 mg/dl) and Platelets 53,000/mm³. The estimated blood loss was a 1000 ml. The parturient was transferred to the ICU for continuous supportive care; 5 h after surgery, due to excessive vaginal bleeding the patient underwent a nonselective bilateral internal iliac artery

embolization procedure. Overall, the patient received 10 units of packed cells, 11 units of fresh frozen plasma, 19 units of platelets and 20 units of cryoprecipitate. She was discharged with no sequelae on day 27. (see Table 1).

We describe a case of acute inversion and immediate reposition where the patient presented an intraoperative respiratory compromise, followed by hemodynamically instability that preceded the clinically observed uterine atony and bleeding. Rapid and facile reposition of the uterus did not lead to improvement of the hemodynamic status, which was achieved only after prolonged respiratory support and coagulation correction. The clinical presentation and the timing of the events recapitulated the sequence described for the “anaphylactoid syndrome of pregnancy”/amniotic fluid embolism (ASP/AFE) in combination with uterine atony. We thus pursued review of the literature in order to evaluate the relation between the acute inversion at cesarean and the “anaphylactoid syndrome of pregnancy” in addition to the expected atony; such a relation would impact on the immediate therapy.

Acute inversion of the uterus following delivery is a rare event and even more rare during Caesarean delivery [1]. In most cases an existent uterine incision and anesthesia allows re-inversion of the uterus to be accomplished immediately.

We identified 29 cases of acute uterine inversion at cesarean delivery; 6 (20.6%), reported a life-threatening event of significant hypotension or cardiac arrest close to the uterine inversion/re-inversion event, not proportional to the reported blood loss [2–6]. Some different pathophysiological mechanisms were suggested. Emmot et al. [3] speculated on the stimulation of the visceral afferent autonomic nerves, by traction and stretching of the peritoneum and broad ligaments at the time of the inversion. Others opposed this option, since a regional, spinal anesthesia used in 4 out of 6 of the cases would blunt this visceral reflex. Another

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Table 1
Cases of uterine inversion during cesarean section.

Estimated Blood loss (ml)	Uterine atony	Cardiovascular collapse	Coagulopathy	Number of cases	Author and year of publication
Not reported	Not reported	Not reported	Reported	1	M. Sivasuriya et al., 1976
1,000 ml	Reported	Reported	Reported	1	J. Broadway, 1988
700 ml; 800 ml	Not reported	Reported	Reported	2	R. S. Emmot et al., 1988
300 ml	Not reported	Not reported	Reported	1	A.P. Masters, 1988
1,000 ml	Not reported	Not reported	Reported	1	D. A. Bayhi et al., 1992
600 ml	Not reported	Not reported	Reported	1	A. Kripkdni et al., 1996
1,500 ml	Reported	Not reported	Reported	1	H. Weiss et al., 1996
Not reported	Reported	Not reported	Reported	1	N. Banerjee et al., 1999
Not reported	Not reported	Not reported	Reported	1	K. Chatzistamatiou et al., 2014
1,600 ml	Reported	Not reported	Reported	1	A.J. Vivanti et al., 2016
45% cases reported massive obstetric hemorrhage	Not available data	Not available data	Not available data	3/24 years	T. F. Baskett et al., 2002
1,500 ml	Reported	Reported	Reported	1	U. Rudloff et al., 2003
600 ml	Not reported	Reported	Not reported	1	A. Khalil et al., 2006
1,500 ml	Not reported	Not reported	Reported	1	D. Vavilis et al., 2008
2,500 ml	Not reported	Not reported	Reported	1	D.Tsivos et al., 2009
600 ml	Reported	Reported	Not reported	1	N.B. Marshall et al., 2010
Average: 1092 (600–2,500) ml	5 (17.2%)	6 (20.1%)	14 (53.8%)	29	Total number of patients reported (%)

*Reported = positive findings; Not reported = negative findings, **All studies reported no maternal death or long term sequelae.

speculation was a sudden release of metabolic waste products, accumulated within the uterus during the inversion due to venous stasis and released after reposition. Contradictory to this assumption is the fact that hemodynamic compromise was seen in several cases, including ours, with an inversion – to reinversion interval of less than a minute, not enough for venous stasis and accumulation of waste products. Finally, some [6] suggested that the hemodynamic instability was a result of vasovagal stimulus and reaction. However, others as well as in our case, reported the hypotension was accompanied by significant tachycardia, a finding that does not comply with vasovagal syndrome.

In view of the latest understandings on the “ASP/AFE” we would like to suggest a different initial pathophysiological cascade. The catastrophic ASP/AFE is a rare obstetric emergency, characterized by cardiovascular collapse, and coagulation abnormalities [7]. Our case and the review of the literature, directed towards similarities between clinical characteristics of the uterine inversion and the “ASP/AFE”; sudden oxygen desaturation [4], hypotension unrelated to the extent of the initial blood loss and coagulopathy only than to be followed by uterine atony. We may theorize that during the process of uterine inversion, minuscule breaks in the maternal–fetal barrier occur, allowing fetal material to rapidly enter the maternal circulation, initiate an immune response and the clinical sequence of events presented above, even before the subsequent uterine atony and its associated excessive bleeding is evident.

We would like to raise the awareness to the possible association between the acute uterine inversion at cesarean and ASP/AFE like sequence of events in parallel with the uterine atony. Emergency protocols with anticipation of maternal respiratory and hemodynamic deterioration; prompt respiratory and hemodynamic support along with administration of coagulation resuscitation products might even prevent the atony event.

Conflicts of interest

All authors have no conflict of interest or benefit that has arisen from the direct applications this research to disclose.

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Patient consent form has been completed and signed by the patient.

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