

**Correspondence:**

Delivery or previous cesarean? A comment on “Antepartum hemorrhage from previous-cesarean-sectioned uterus as a potential sign of uterine artery pseudoaneurysm”

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Uterine artery pseudoaneurysm (UAP), an important disorder causing postpartum hemorrhage, has been considered to occur after “traumatic delivery” such as cesarean section (CS). Our recent study (Baba *et al.*, 2016) confirmed that UAP can also occur after “non-traumatic” delivery.

A recent paper entitled “Antepartum hemorrhage from previous-cesarean-sectioned uterus as a potential sign of uterine artery pseudoaneurysm” (Zhang *et al.*, 2017), published in the *Journal of Zhejiang University-SCIENCE B (Biomedicine & Biotechnology)*, described an interesting case with UAP. At 38 weeks, a pregnant woman with previous CS showed “antepartum hemorrhage” and vaginally gave birth to an infant. She had massive postpartum hemorrhage, which was confirmed as due to UAP rupture: uterine artery embolization (UAE) achieved hemostasis. We have some clarifications.

Zhang *et al.* (2017) used the phrase of “prior CS-related UAP”. We wish to make things clearer: it refers to the pathophysiological mechanism of UAP. The time course of this patient was: previous CS→uterine contraction→antepartum hemorrhage→precipitous vaginal delivery→massive postpartum hemorrhage and UAE. A “postpartum hemorrhage” occurred after “vaginal delivery”, and thus, in this sense, their case represented a UAP without “preceding traumatic events”. However, “antepartum hemorrhage” also occurred, highlighting a uniqueness of this patient’s UAP. This poses a question whether UAP was formed during “this vaginal delivery” or “previous CS”.

Two scenarios may be possible. In Scenario 1, this delivery (uterine contractions, hemodynamic changes, or something else) caused arterial wall injury, leading to UAP formation. UAP ruptured before infant delivery, yielding both antepartum- and postpartum-hemorrhage. UAP was formed during this delivery (labor) and manifested even before infant delivery, meaning that the present UAP represented “UAP without preceding traumatic events”. The uterine artery and/or its branches at the site of previous CS scar may be vulnerable to exogenous stimuli during labor contractions, and thus UAP may be more readily formed at this site. Zhang *et al.* (2017)’s phrasing “prior CS-related UAP” may illustrate this condition.

Another scenario (Scenario 2) may be possible. Previous CS caused arterial wall injury, leading to UAP formation but it remained unruptured. UAP continued to be intrauterine. A hyper-dynamic state during labor, i.e. uterine contractions/relaxation and uterine-artery-inflow/outflow, may have caused UAP-sac rupture and resultant antepartum hemorrhage, but

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sac-rupture was transiently sealed. The sac was re-ruptured postpartum, causing postpartum hemorrhage. Thus, in Scenario 2, the present UAP represented “UAP with preceding traumatic events”. The uniqueness of this case is interpreted differently between Scenarios 1 and 2. In Scenario 1, UAP was formed during labor contractions and can manifest as antepartum hemorrhage. In Scenario 2, UAP was caused by the previous CS but hidden intrauterine, and can manifest as antepartum hemorrhage in the next pregnancy. Zhang *et al.* (2017)’s phrasing “prior CS-related UAP” may also hold true to illustrate this Scenario 2. However, the pathophysiological mechanism of UAP is different between Scenarios 1 and 2.

We characterized 50 UAP patients. Focusing on “just preceding (the last) delivery”, 29 had “traumatic delivery/abortion” whereas the remaining 21 did not. Of 21, interestingly, 9 had delivery-/abortion-related “traumatic events” in the “the second last delivery/abortion”, meaning that not only “just before delivery” but also “all prior history of deliveries” may have responsibility for UAP (Baba *et al.*, 2016).

In addition, a lag time between a preceding event and manifestation/detection of UAP may sometimes be very long. Patients in whom UAP was detected 10 years (Johannesson *et al.*, 2017) and even 20 years (Papadakos *et al.*, 2008) after CS have been reported.

UAP is not as uncommon as previously believed. Our study revealed that it occurred in 3–6 per 1000 deliveries (Baba *et al.*, 2016). Zhang *et al.* (2017)’s case is unique and important: it directly showed that UAP may manifest not only as postpartum hemorrhage but also as antepartum hemorrhage. Putting aside the discussion whether their case was traumatic-delivery-related or unrelated, their observation is clinically useful. However, its pathophysiology should be reconsidered and widely discussed. At present, we cannot conclude which scenario was the case; however, looking at UAP from this viewpoint may widen the discussion on this important issue.

Contributors

Shigeki MATSUBARA, Hironori TAKAHASHI, Daisuke MATSUBARA, and Yosuke BABA identified the significance and wrote the manuscript.

Compliance with ethics guidelines

Shigeki MATSUBARA, Hironori TAKAHASHI, Daisuke MATSUBARA, and Yosuke BABA declare that they have no conflict of interest.

This article does not contain any studies with human or animal subjects performed by any of the authors.

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中文概要

题目: 与妊娠相关的子宫动脉瘤形成是源于此次分娩还是前次瘢痕子宫? ——对一篇病例报道“疤痕子宫产前出血可能是子宫动脉瘤的潜在危险信号”的评论

概要: 子宫动脉瘤多被认为与产程中创伤性操作相关,但也可发生于无创伤性操作的自然分娩过程中。最近狄文和张宁等人在《浙江大学学报(英文版)B辑:生物医学与生物技术》中报道了一例临床病例(Zhang *et al.*, 2017): 疤痕子宫妊娠→子宫收缩→产前出血→阴道分娩急产→严重产后出血→子宫动脉栓塞。该病例中的子宫动脉瘤形成的原因可能是:(1)此次分娩过程中,子宫疤痕处的血管分支薄弱,宫缩压力诱发子宫动脉瘤形成并破裂;(2)前次剖宫产术的创伤性操作导致子宫动脉瘤形成但并未破裂,此次妊娠分娩促进动脉瘤的破裂出血。我们汇总分析了50例子宫动脉瘤患者,29例都是前次妊娠(最近第2次妊娠)有创伤性操作,而在另外21例中,其中9例在最近第3次妊娠中都存在创伤性操作。这些提示子宫动脉瘤的形成可能与所有既往的创伤性操作史有关,两者间的时间间隔可能很长,比如10年甚至20年之久。此外,张宁等人的病例报道也提示子宫动脉瘤不仅仅表现为产后出血,产前出血也可能是其危险信号。

关键词: 子宫动脉瘤; 创伤性操作史; 产后出血; 产前出血

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