

# Cesarean scar pregnancy and uterine artery pseudoaneurysm: preceding or coexisting?

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## Summary

Uterine artery pseudoaneurysm (UAP), an important cause of postabortal or postpartum hemorrhage, usually accompanies a preceding event, which injures the arterial wall, leading to pseudoaneurysm formation. The authors here propose a new concept, UAP without preceding events. Simultaneous occurrence of UAP and cesarean scar pregnancy may well illustrate the situation.

*Key words:* Cesarean scar pregnancy; Postpartum hemorrhage; Preceding event; Pseudoaneurysm; Uterine artery pseudoaneurysm.

## Introduction

Uterine artery pseudoaneurysm (UAP) has recently attracted wide attention as an important cause of postabortal or postpartum hemorrhage. In UAP, there are usually preceding events, in which the uterine arterial wall is injured. If the injured part is surrounded by tissues and has still continuity with the parent artery (the uterine artery), pseudoaneurysm forms. Pseudoaneurysm ruptures and then bleeding occurs [1, 2].

Preceding events have been considered to be “traumatic”, namely, cesarean section, manual placental removal, or dilatation and curettage. The present authors refuted this belief: preceding events can be “non-traumatic”: normal vaginal delivery or uneventful mid-trimester pregnancy termination can be preceding events of UAP [1]. Irrespective whether preceding events are traumatic or not, it is believed that UAP always accompany preceding events [1, 2].

The present authors here speculated that cesarean scar pregnancy (CSP), in which trophoblasts directly invade the shallow myometrium in the site, may injure the arterial wall there, and, thus, causing UAP. In this scenario, UAP and CSP “coexist”. This signifies a new concept of UAP without preceding events. The present authors here propose this new concept by interpreting reported cases with simultaneous occurrence of UAP and CSP as its model.

## Materials and Methods

The present authors searched reported cases of simultaneous occurrence of UAP and CSP via PubMed, using “uterine artery pseudoaneurysm” AND “cesarean scar pregnancy” as index words. They also hand-searched related articles and related conditions.

## Results

There are three reports via PubMed search, of which two reports described patients in whom UAP and CSP occurred simultaneously [3, 4]. Mou *et al.* [3] reported a patient in whom curettage was performed to miscarriage in CSP, leading to UAP formation and its rupture, necessitating hysterectomy. Another patient was described in this Journal by Tan *et al.* [4]. The latter case well illustrates the point: a new concept of “UAP without preceding event” and thus, is discussed in detail.

## Discussion

Tan *et al.* [4] described a patient with the “simultaneous occurrence” of two rare disorders, UAP and CSP. This patient’s course leads to an important clinical question regarding the pathophysiology of UAP. Did curettage for CSP or CSP itself injure the uterine artery? Which was the culprit for UAP, curettage or CSP itself?

As described, a typical UAP has three phases: 1) “preceding event”, in which some traumatic uterine-cavity procedures (curettage, cesarean section, etc.) injure the arterial wall and the pseudoaneurysm is formed, 2) “lag time”, in which the pseudoaneurysm gradually increases, and then 3) “UAP manifestation”, the intra-sac pressure exceeds the sac-wall strength, and then UAP ruptures, usually manifesting as genital bleeding (Figure 1A). Importantly, in phase 2), conditions demanding preceding events (curettage or cesarean section) have already been resolved and UAP symptoms have not yet manifested. Thus, “simultaneous occurrence” of disorders demanding preceding events and UAP is a rare phenomenon, as Tan *et al.* stated [4]. However, rarity is not the point: there may be two sce-

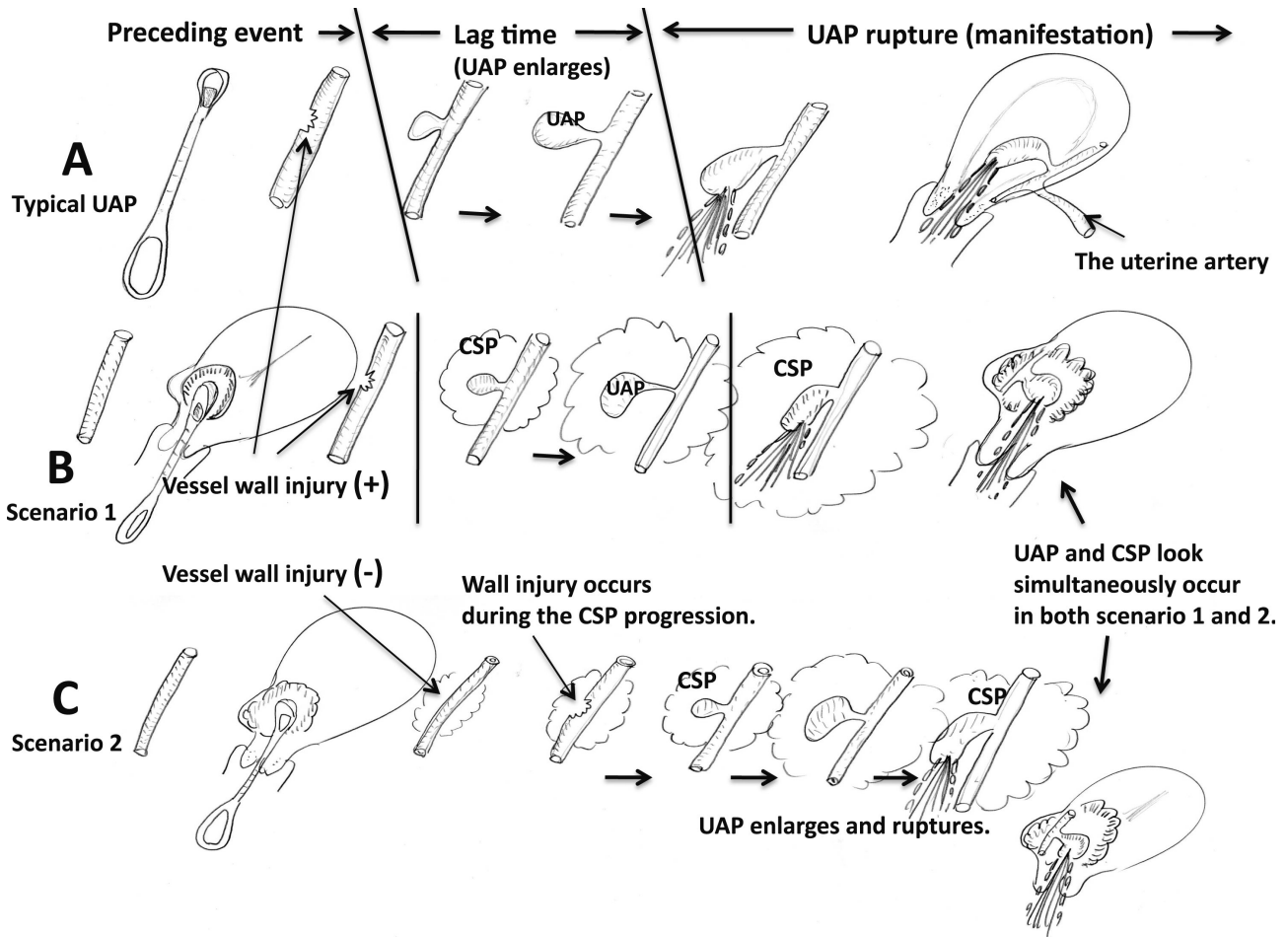


Figure 1. — Schematic presentation of time course of uterine artery pseudoaneurysm (UAP) formation. A typical case (A), scenario 1 (B), and scenario 2 (C) are illustrated (a typical case). There are three phases: a preceding event (curetage), lag time, and UAP rupture. *Scenario 1*: The wall of the uterine artery or its branch, which is within, or adjacent to, cesarean scar pregnancy (CSP), is injured during curetage. UAP is formed and is ruptured, similar to a typical UAP (indicated in A). *Scenario 2*: Curetage did not injure the arterial wall. The arterial wall is injured during the CSP progression.

narios for Tan *et al.*'s case, which is the point that should be learned from this case. The first scenario is as follows (Figure 1B). The authors stated that, "the clinician at the patient's previous hospital may have mistaken the CSP for an abortion. Blind curetage in this case could result in massive hemorrhage, and iatrogenic mechanical injury could lead to the formation of UAP". In this context, curetage can be considered as a preceding event, similar to typical UAP. This scenario is the same as that of a case reported by Mou *et al.* [3]. Although CSP and UAP coexisted, a preceding event, curetage, did exist. Curetage did not ameliorate the CSP, and, thus, CSP was still present at the time of UAP manifestation: a simple scenario. The second scenario is: CSP itself may have caused UAP (Figure 1C). Arterio-venous malformation (AVM) was caused by CSP itself without preceding events [5]. In CSP, there is no decidua and the implantation site is directly surrounded by

the myometrium: vessels in the site may be more likely to be injured while CSP progresses. This can cause arterial wall injury, leading to AVM: there are no preceding events for this type of vessel abnormality. AVM is a different entity from UAP, and, thus, caution should be exercised when considering the pathophysiology of UAP based on that of AVM; however, this mechanism for AVM formation in CSP may also hold true for UAP. Thus, the second scenario highlights "UAP without preceding event", a different concept from the first widely acknowledged scenario.

The present authors, to their knowledge, were the first to describe "UAP without preceding event": it was UAP caused by septic abortion [6]. Infection may have caused vessel injury, leading to UAP. They find a strong similarity between Tan *et al.*'s case and the present: in both, 1) there were no preceding events, and 2) some disorders (CSP or septic abortion) and UAP coexisted.

Which was the culprit for UAP, curettage or CSP itself in Tan *et al.*'s case? Possibly, both were associated with UAP. UAP shows various clinical features and thus the present authors refer to it as a "chameleon in obstetric practice", with its features changing in a patient-by-patient manner [2]. UAP may be hidden everywhere and thus efforts should be made to clarify its concept based on its pathophysiology, which may contribute to revealing a hidden chameleon. As previously reported, UAP is not as rare as previously believed: it occurred in 2-3/1,000 deliveries [1], and thus it resides "everywhere".

## Conclusion

UAP may not always accompany preceding events. Simultaneous occurrence of UAP and CSP may well illustrate a new concept of UAP without preceding events.

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