

## CASE REPORT

# Diagnosis of unicuspid aortic valve after loss of consciousness during cesarean section: A case report

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## Abstract

Loss of consciousness is a rare but potentially serious complication of delivery by cesarean section under spinal anesthesia. Here, we report the case of a pregnant woman with unicuspid aortic valve that was incidentally diagnosed during aortic valve replacement after transient loss of consciousness during cesarean section.

## KEYWORDS

cesarean section, loss of consciousness, spinal anesthesia, unicuspid aortic valve

## 1 | INTRODUCTION

Loss of consciousness is a rare but potentially serious complication of cesarean delivery under spinal anesthesia, and eclampsia, pulmonary embolism, and amniotic fluid embolism have been reported as causes of loss of consciousness<sup>1-4</sup>. Here, we describe a case of a pregnant woman complicated with unicuspid aortic valve (UAV), which was incidentally diagnosed during aortic valve replacement after transient loss of consciousness during cesarean section.

## 2 | CASE PRESENTATION

A 35-year-old healthy pregnant woman (height: 156 cm, weight: 63 kg, BMI: 25.8 kg/cm<sup>2</sup>) was scheduled to undergo

a repeat cesarean section. A previous cesarean section had been performed 2 years prior without any complications. She was admitted at gestational week 38 for a scheduled cesarean delivery. All preoperative examinations, including electrocardiography, were normal.

In the operating room, blood pressure (BP), heart rate (HR), and arterial oxygen saturation of pulse oximetry (SpO<sub>2</sub>) were 146/80 mmHg, 60 beats/min, and 99%, respectively. Spinal anesthesia, comprising 0.5% hyperbaric bupivacaine hydrochloride (11 mg) combined with fentanyl (10 µg), was injected at the L3-4 interspace using a 25-gauge Quincke needle in the right lateral position. The patient was subsequently placed in the left tilted supine position. Continuous phenylephrine administration (1000 µg/h) was started intravenously just after the intrathecal injection to prevent a decrease in BP due to spinal anesthesia. Her BP (120/70 mmHg) and HR (60 beats/

min) were stable. The operation was initiated after confirming an appropriate sensory block level (Th 4/4).

The fetus was delivered 12 min after the operation began. During the placenta removal, the patient suddenly lost consciousness, with up-rolling of the eyeballs. Before losing consciousness, the patient had no complaints of chest pain, dyspnea, or tingling in the upper limbs. Her HR increased from 79 to 89 beats/min. No arrhythmia was evident on anesthesia monitoring. Spontaneous respiration was temporarily absent and oxygen saturation dropped to 88%; hence, 100% oxygen administration and ventilation with a face mask was initiated. 2 min after loss of consciousness, the patient regained consciousness and spontaneous ventilation resumed. Her BP, which was 130/74 mmHg immediately before losing consciousness, dropped to 105/67 mmHg on regaining consciousness. BP during the period of unconsciousness was not detected as the measurements were intermittent. After regaining consciousness, the patient did not complain of any symptoms. Her pupils were equal and round, and were reactive to light and accommodation; extraocular muscles were intact. She had no paralysis of the arms and no dysarthria. The estimated blood loss, including amniotic fluid, was 740 mL and operation duration was 57 min. Her blood gas analysis was maintained within the normal ranges. The sensory block level was bilateral Th 4 at the end of the surgery.

Several causes of loss of consciousness during cesarean section were considered as differential diagnoses including pulmonary embolism, amniotic fluid embolism, and cerebral ischemia. Whole-body contrast-enhanced computed tomography (CT) revealed a hypoabsorption region near the right coronary cuspid of the aortic valve (Figure 1). The findings of the transthoracic echocardiogram were as follows: mean and peak pressure gradients were 25 mmHg and 44 mmHg, respectively; peak velocity

through the aortic valve was 3.3 m/s; and the valve area was 1.7 cm<sup>2</sup>. Subsequent transesophageal echocardiogram (TEE) suggested a bicuspid aortic valve and a high echogenic mobile mass (1 cm × 1.7 cm in size) near the base of the right coronary cusp (Figure 2).

At this point, a definitive diagnosis was not determined; however, transient cerebral ischemia due to a small embolus could not be ruled out. Further, due to the risk of recurrence of the embolism, surgical resection of the mass near the aortic valve was immediately scheduled. Surgical findings revealed that the echogenic mass was a thickened valve with a single slit-like opening, which was ultimately diagnosed as UAV (Figure 3). The commissure was located between the left coronary apex and the non-coronary apex. There was no obvious tumor or valvular vegetation on the aortic valve or root. The aortic valve replacement was successful, and the postoperative course was uneventful.

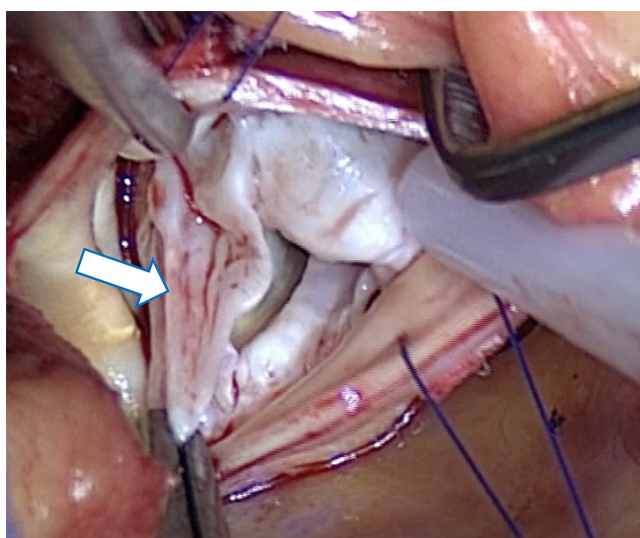
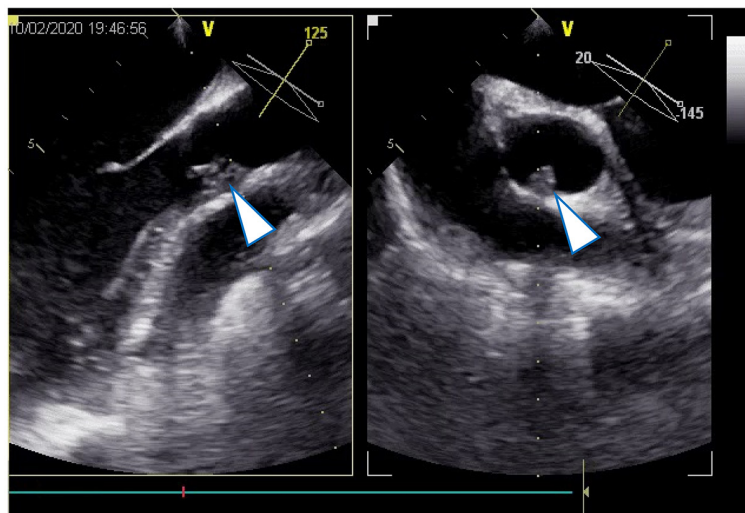
### 3 | DISCUSSION

This is the first case report of a UAV detected following syncope after spinal anesthesia for a cesarean section. UAV is a rare condition accounting for 0.02% of adults worldwide,<sup>5</sup> and the main symptoms include dyspnea, angina, dizziness, and syncope; it is often associated with aortic stenosis (AS).<sup>6</sup> The average age of patients with AS who require surgery is relatively lower than that of patients with bicuspid and tricuspid valves.<sup>7</sup> Similar observations were noted in our case. Echocardiography is the first choice for diagnosing UAV; however, differentiating UAV from bicuspid and tricuspid aortic valves remains challenging. A major concern results from raphes or leaflet calcification. Recently, three-dimensional (3D) echocardiography has become



**FIGURE 1** Contrast enhanced computed tomography at the arterial phase. An axial image indicating a solid mass near the aortic valve and coronary orifice (arrow head). Arrow shows the right coronary artery.

**FIGURE 2** Long axis view (A) and short-axis view (B) of transesophageal echocardiography (TEE) of the unicuspid valve. A high echogenic mass is indicated by arrows heads.



**FIGURE 3** Intraoperative image of the aortic valve. Intraoperative findings of the unicuspid valve, which exhibited a single commissure. The arrow indicates the redundant lesion of the aortic valve, which was recognized as a mobile mass by transesophageal echocardiography (TEE).

increasingly important in their diagnosis.<sup>8</sup> UAV can be further classified into acommisural UAV and unicommissural UAV based on the morphology of the aortic valve<sup>9</sup> This case was categorized as unicommissural UAV, which has a relatively large orifice area compared to acommisural UAV, and is associated with the appearance of symptoms in adults<sup>9</sup> Furthermore, the aortic valve in our patient was an overlong valve with a soft root and stiff tip. Therefore, it was incidentally detected as a mass near the right coronary apex of the aortic valve during the chest CT scan performed during the cesarean section postoperative analysis.

Some large-scale studies have investigated the prevalence of loss of consciousness during cesarean section<sup>10,11</sup> In a previous report based on approximately 25,700 maternal cases, the rate of respiratory arrest similar to the

present case was 1/8455.<sup>10</sup> Another study on 182,600 cases of cesarean sections reported that the rate of loss of consciousness due to a high-level block was 1/6667.<sup>11</sup> Eclampsia, embolism (coronary, pulmonary, or amniotic fluid embolism), massive hemorrhage, vasovagal syncope, arrhythmia, cardiac structural disease, and high-level blocks have been reported as causes of consciousness loss during cesarean section.<sup>1-4,12-14</sup> In our case, the intraoperative course of the patient was negative for massive hemorrhage and a high-level block. Since there was no pre-eclampsia, syncope due to eclampsia was also deemed unlikely. The patient had no history of epilepsy or diabetes causing loss of consciousness, and her postoperative blood glucose levels were within normal range. Vasovagal syncope is the most common cause of transient loss of consciousness, secondary to pain, fear, or medical procedures; however, in this case, there was no bradycardia during syncope suggesting a vagal reflex.

Since the patient's ECG was not recorded during the loss of consciousness, the details are absent. However, the ECG on the anesthesia monitor showed no arrhythmia. Therefore, we initially suspected embolism and performed a full-body CT scan. Although the postpartum condition is a well-established risk factor for pulmonary embolism, it was not evident on contrast-enhanced CT scan. Amniotic fluid embolization is very rare, ranging from 1.9 to 6.1 cases per 100,000 deliveries,<sup>15</sup> but should always be considered in the differential diagnoses because of its high maternal mortality rate.<sup>16</sup> Amniotic fluid embolization is defined by the presence of all of the following criteria<sup>1</sup>: sudden onset of cardiopulmonary arrest or hypotension with evidence of dyspnea,<sup>2</sup> diagnosis of disseminated intravascular coagulation (DIC) modified for pregnancy,<sup>3</sup> clinical onset during labor or within 30 minutes of placental delivery, and<sup>4</sup> absence of fever during labor<sup>17</sup> In our case, the patient's blood sample did not meet DIC criteria. Finally, UAV was diagnosed based on a CT scan. UAV was

associated with syncope during cesarean section in the present case.

There have been no reports of loss of consciousness during cesarean section due to UAV, but there has been a previous case of loss of consciousness due to AS associated with congenital heart disease.<sup>18</sup> Staging of valvular AS includes the valve anatomy, hemodynamic characteristics of the valve, cardiac structural consequences of the stenotic valve, and patient symptoms.<sup>19</sup> Based on the hemodynamics of the valve, this case was classified as stage B (maximal velocity 2.0–3.9 m/s or mean gradient <40 mmHg). However, the valve structure of the UAV in this case could be classified as stage C (severe leaflet calcification/severely reduced leaflet opening), which is defined as asymptomatic severe AS with strong calcification and thickening of the redundant valve.<sup>19</sup> Moreover, hemodynamic changes during pregnancy are known to exacerbate AS<sup>20</sup> and this case was also likely to be more severe at the time of the cesarean section than at the time of the postoperative echocardiographic evaluation. A recent study analyzing a multinational registry found that 12.9% of pregnant women with moderate AS were admitted to the hospital for cardiac reasons.<sup>21</sup> In this paper, the authors warn that even asymptomatic AS patients during pregnancy may present with symptoms at the time of delivery due to the additional hemodynamic stresses.<sup>21</sup> In our case, the patient had no symptoms during pregnancy, but hemodynamic changes at delivery may have caused loss of consciousness.

Two mechanisms for loss of consciousness are possible in this case. The first is a rapid decrease in peripheral vascular resistance. The administration of spinal anesthesia decreases vascular resistance, and the subsequent adoption of the supine position triggers aortic compression. Immediately after the start of surgery, the release of aortic valve compression due to fetal delivery, uterine autotransfusion, bleeding from the placenta removal site, and the influx of fetal components, such as amniotic fluid, into the maternal circulation all occur concurrently. In addition, bolus infusions of oxytocin act as potent vasodilators.<sup>22</sup> Regarding the use of invasive catheterization in normal pregnant women, Ueland et al. reported that the release of intra-abdominal pressure by fetal delivery reduced peripheral resistance by 13% with epidural anesthesia.<sup>23</sup> Furthermore, the release of intra-abdominal pressure by fetal delivery can reduce peripheral resistance by as much as 10% under epidural anesthesia. Brian et al. showed that in pregnant women with AS who underwent cesarean section under epidural anesthesia, the timing of oxytocin infusion reduced the total peripheral vascular resistance by 44%.<sup>24</sup> Several combined factors during the cesarean section could have contributed to the decrease in peripheral vascular resistance, which could have led to syncope. The second mechanism is the unusual valve shape of the UAV.

In the present case, the UAV was overlong and had a hard tip. The hardened tip of the UAV may have temporarily blocked the aortic valve opening, resulting in a decrease in cerebral return pressure. Furthermore, the thickening of the valve located at the right coronary apex and the turbulent flow at the entrance of the right coronary artery may have caused a decrease in blood flow in the coronary artery due to the impaction of the thickened part of the valve into the right coronary artery, leading to loss of consciousness.

This case has several limitations. First, we failed to detect a drop in blood pressure during loss of consciousness because intraoperative blood pressure measurements were conducted intermittently with a cuff. Second, before the cesarean section, we did not auscultate the heart sound; had auscultation been performed, AS may have been detected. However, at the time of this case, COVID-19 was rampant, and our hospital policy was to minimize physical contact with low-risk perioperative patients to the greatest extent possible. Third, 3D-TEE, which is a more accurate method to delineate the exact anatomy of the aortic valve, was not performed. If the 3D-TEE showed the exact morphology of the mass found on the 2D-TEE, probably with an aortic valve area of 1.7 cm<sup>2</sup>, the operative treatment may have been delayed temporarily.

## 4 | CONCLUSION

Overall, we described a case of UAV that was detected after a patient experienced syncope following spinal anesthesia for cesarian section. UAV with AS can lead to syncope, especially during the perinatal period, due to the highly dynamic maternal physiology. Evaluation for valvular heart diseases should be considered in patients with unexplained loss of consciousness during cesarean section after spinal anesthesia.

### AUTHOR CONTRIBUTIONS

**Shun Yukami:** Writing – original draft. **Takashi Juri:** Writing – original draft; writing – review and editing. **Asami Nakajima:** Writing – review and editing. **Tachibana Daisuke:** Writing – review and editing. **Yosuke Takahashi:** Writing – review and editing. **Shinichi Iwata:** Writing – review and editing. **Akira Yamamoto:** Writing – review and editing. **Takashi Mori:** Writing – review and editing.

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Not applicable.

### CONFLICT OF INTEREST STATEMENT

The authors declare that there is no conflict of interest to be reported.

## DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analysed during the current study.

## ETHICS STATEMENT

This study did not include any experiments on animals or human subjects.

## CONSENT

Written informed consent was obtained from the patient to publish this case report.

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