

# UTERINE TORSION WITH PLACENTAL ABRUPTION AND FETAL DEATH AT TERM PREGNANCY: A CASE REPORT AND LITERATURE REVIEW

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## ABSTRACT

**BACKGROUND:** Uterine torsion in pregnancy is a rare event that requires urgent recognition and management. Its presentation is non-specific and ranges from asymptomatic to severe life-threatening conditions such as placental abruption with hemorrhagic shock and maternal and fetal death. Here we describe our case and reviewed similar literature on gravid uterine torsion. To our knowledge, this is the first report of term uterine torsion complicated by placental abruption and fetal death in Africa.

**CASE PRESENTATION:** A 28 year old Ethiopian gravida III, para II woman, presented at 37 weeks of gestation with acute severe abdominal pain, mild vaginal bleeding, absent fetal movement, and shock. Emergency cesarean section was decided with a presumptive diagnosis of placental abruption with hemorrhagic shock and intrauterine fetal demise. Intraoperatively, it was realized that the placenta and the dead fetus were delivered through an inadvertent posterior lower segment hysterotomy made on 1800 levo-rotated uterine torsion. Then the uterus was restored to its anatomic position and the posterior hysterotomy was closed. Despite medical measures, the uterus became atonic and a hysterectomy was required. The patient had an uneventful postoperative and postpartum course.

**CONCLUSION:** Even if gravid uterine torsion is uncommon, it should be kept in mind as one of the differentials in pregnancy-related acute abdominal emergencies due to its non-specific nature and dangerous impact. Furthermore, it can be obscured and associated with other obstetric emergencies, especially placental abruption.

**KEYWORDS:** uterine torsion, placental abruption, pregnancy, Ethiopia, case report

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## INTRODUCTION

Axial rotation of the uterus more than 45° is pathologic and is known as uterine torsion.<sup>1-3</sup> It is a rare entity with an unknown incidence observed in both gravid and non-gravid uterus. It also affects all ages and all parity groups. Uterine torsion for the first time was reported in 1863 by Virchow in a deceased woman. Later Labbe reported it in living women in 1876.<sup>1,2,4,5</sup>

Even if the exact etiology is unknown, conditions affecting the anatomic fixity of the uterus are incriminated in the process. These conditions include myoma, uterine anomalies, adnexal masses, fetal malpresentation, pelvic abnormalities, trauma, and connective tissue disorders.<sup>1,2,3,5,6</sup>

Physiologically, in two third of the cases, the gravid uterus is dextro-rotated less than 45°. The torsion occurs at the level of uterine isthmus and is commonly 180°; however, more severe cases of up to 720° are reported.<sup>2,7</sup> All trimesters of pregnancy are at risk but 67 to 78% of torsions occur in the third trimester of pregnancy.<sup>2,5</sup> The earliest reported gestational age for uterine torsion during pregnancy is in the 6<sup>th</sup> week and the latest is in the 43<sup>rd</sup> week.<sup>2</sup>

In most cases, the diagnosis of gravid uterine torsion is made intraoperatively. This is because the symptoms are either absent or non-specific.<sup>2,4,5,8</sup> Furthermore, it may also be obscured by associated obstetric emergency conditions, especially placental abruption. In general, the symptoms may range from mild abdominal pain to life-threatening conditions such as hemorrhagic shock, and fetal and maternal death.<sup>1-3,5,7,9-18</sup> In suspected cases, imaging such as sonography and MRI may help in the diagnosis.<sup>5,19,20</sup>

Uterine torsion is an obstetric emergency that requires laparotomy to correct the torsion and further procedures may be performed based on the gestational age and associated conditions.<sup>2,3,5</sup> If not treated timely, it is associated with grave outcomes and may result in maternal and perinatal mortality.<sup>2,5,7,8,11,12,21</sup>

In this paper, we report a case of third-trimester (37 weeks) uterine torsion identified intra operatively and complicated by placental abruption and intrauterine fetal death (IUFD). The challenges and difficulties in making the diagnosis are discussed. Moreover, we reviewed similar literature on uterine torsion since 1999. To the best of our knowledge, this is the first report of uterine torsion complicated by placental abruption and fetal death in Africa.

## CASE PRESENTATION

A 28-years old Ethiopian gravida III, para II woman presented to our hospital at the gestational age (GA) of 37+2 weeks from reliable last menstrual period (LMP) with sudden onset of abdominal pain of one-day duration. The pain was initially located in the lower abdomen and later become diffuse and accompanied by absent fetal movement, mild vaginal bleeding, headache, light headedness and syncopal episode. She had no associated history of trauma or previous surgery and no other significant obstetric danger symptoms. She also denied self or family history of chronic medical illnesses.

Her prior obstetric history revealed two vaginal deliveries at term. One is alive and the other was stillbirth after home delivery by a traditional birth attendant with no identifiable cause.

She had regular antenatal care (ANC) follow-ups starting from 16 weeks of GA at the nearby primary healthcare facility and it was uneventful. With the recent complaint, she visited the nearby primary health care and was referred to our hospital.

On arrival to our hospital, she was pale, hypotensive (undetectable blood pressure) and faint pulse. Abdominal examination revealed diffuse tenderness, a large for date tense gravid uterus with a longitudinal fetal lie and breech presentation. There was no uterine contraction and the fetal heart beat (FHB) was negative. On digital pelvic examination, cervix admitted the tip of a finger, uneffaced and posterior, station -1, medium consistency, and mild vaginal bleeding.

Immediately, a double intravenous (IV) line was secured, the patient was catheterized, shock management was initiated and blood was drawn for cross-match and laboratory workup. Emergency obstetric ultrasound showed 37 weeks single intrauterine pregnancy, breech presentation, negative FHB, adequate amniotic fluid, and 12x13cm retroplacental clot. The blood test revealed 8mg/dl of hemoglobin level, 9700 /  $\mu$ L white blood cell count and 148000/ $\mu$ L platelet count. Renal function (creatinine 0.69) and liver enzyme (ALT 23U/L, AST 16U/L) tests were within the normal limits.

There was a preoperative diagnosis of placental abruption with hemorrhagic shock and intrauterine fetal death (IUFD). Informed consent was taken. Cross-matched blood, fresh frozen plasma (FFP), and platelets were prepared for transfusion. Bed in the intensive care unit (ICU) was secured, prophylactic IV antibiotic was given, and the patient was taken to the operating room for surgical exploration (emergency cesarean section).

Under general anesthesia (GA) abdomen was entered through a midline incision. There was an intact gravid uterus with engorged vessels on the wall and then the lower uterine segment was identified

and a transverse incision was made. Upon incision, there was a gush of blood, a freshly dead 2800gm weight male neonate in breech presentation and a completely (100%) detached placenta with a retroplacental clot was found. After delivering the stillbirth baby and the placenta, the uterus was exteriorized. At this point, 180-degree levorotation of the uterus at the uterocervical junction was detected (Figure 1 A) and on further inspection, it was realized that the hysterotomy was made on the posterior aspect of the lower uterine segment (Figures 1 A,B, and C). The uterus was restored manually to its anatomic position. It was soft and succulent but no uterine anomalies, myoma or adnexal mass were noted. Then the uterus was mopped and the incision was closed in a double layer. At the same time, uterotonic agents were administered. Despite all these, the uterus continued to be atonic and the patient became hemodynamically unstable due to bleeding for which a hysterectomy was performed. After hemostasis was secured, the abdomen was closed in layers.

Perioperatively, the patient was transfused with six units of blood, platelets, and FFP. She had an uneventful postoperative course and was discharged on the 5<sup>th</sup> day.

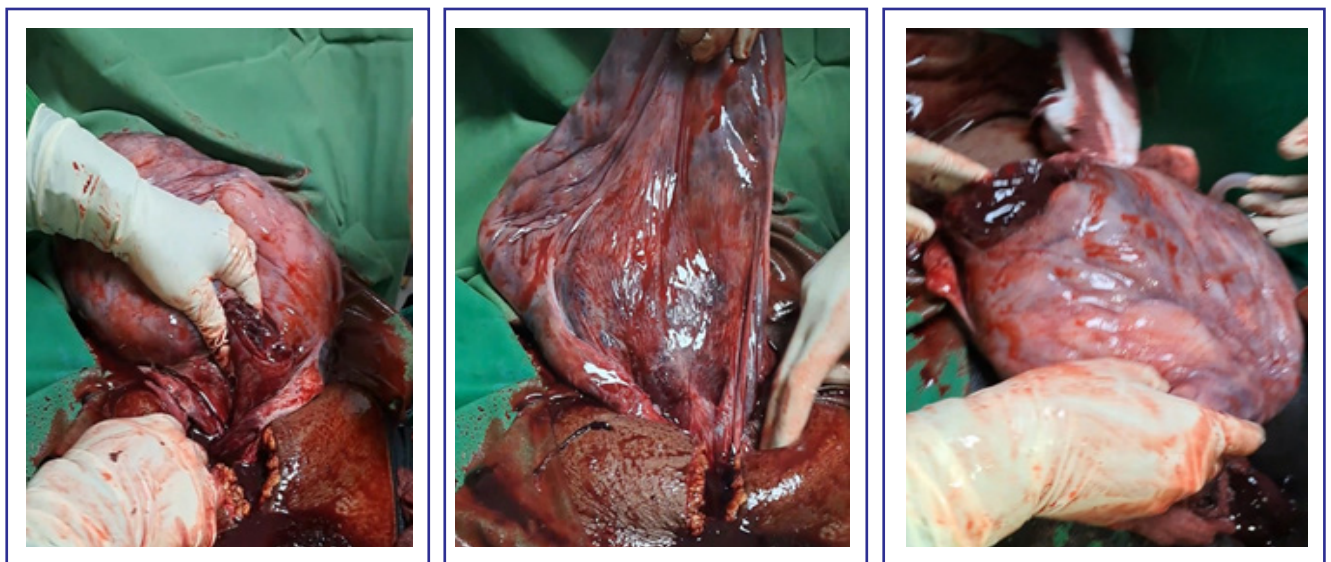


Figure 1: Intraoperative findings after the uterus is exteriorized; (A) 180<sup>o</sup> levorotated uterus at the uterocervical junction (white arrow); (B) no incision is noted on the anterior wall of the uterus after detorsion; (A and C) inadvertently made posterior hysterotomy (red arrow).

## DISCUSSION

Uterine torsion in association with pregnancy was first described by Labbe in 1876.<sup>1</sup> Subsequently, several authors reported their experience with this rare condition. One of the biggest reviews was made in 1992 by Jansen which included 212 cases of gravid uterine torsion.<sup>2</sup> Recently, in 2020, Ramseyer et al. reviewed 41 cases of gravid uterine torsion since 2006.<sup>5</sup> To the best of our knowledge, this paper is the first case report of term uterine torsion complicated by placental abruption and IUFD in Africa. In 1961 Carter reported a similar case from Harare maternity hospital, however, the neonate died after birth despite resuscitation.<sup>9</sup> Furthermore, we reviewed similar literature on uterine torsion in PubMed/Medline using the terms “uterine torsion” and “placental abruption” and found 10 relevant reports since 1999.<sup>9-18</sup>

Compared with a non-gravid uterus, torsion is commonly reported in the gravid uterus. Pregnancy-associated changes occurring on the uterus might exacerbate its physiologic rotation and increase the risk of this rare phenomenon.<sup>1,2,5,22</sup> As it is noted in our case, most uterine torsions occur in the third trimester. Jensen and Ramseyer et al., in their review, found that 67 % and 78% of cases were in the third trimester, respectively.<sup>2,5</sup> This might be explained by the advancement of pregnancy-induced changes in the third trimester. This includes the growth and softening of the lower uterine segment and congestion and loosening of the tissues.<sup>1,22</sup> Although all parity groups are at risk, uterine torsion is observed mostly in multiparous women.<sup>2,5</sup> Among the 41 cases reported since 2006, more than two third (78%) of the cases occurred in parous women.<sup>5</sup> This could be due to the repeated effect of the pregnancy on uterine fixity.

Nesbitt et al. in 1956 concluded that, “no pelvic pathology, torsion unlikely”.<sup>1</sup> Unfortunately, several medical literatures revealed patients with no attributable pelvic pathology. Jansen noted 16% and Ramseyer et al. reported 63.4% of the cases with

no identifiable pelvic pathology.<sup>2,5</sup> So far, various maternal and fetal contributing factors were reported with gravid uterine torsion (Table 1).<sup>1,2,5,6</sup> Myoma and Müllerian anomalies were the most common pelvic pathologies found in association with uterine torsion.<sup>1,2,23</sup> Some authors also attributed 66% of the cases to intrapelvic conditions.<sup>23</sup> All of these conditions are thought to destabilize the anatomic fixity and predispose to uterine mobility and torsion.<sup>2,5</sup> Duplantier et al. reported the first case of gravid uterine torsion with placental abruption following a road traffic accident and suggested to suspect torsion in a pregnant woman involved in blunt abdominal trauma.<sup>10</sup> Others found adhesions following cesarean section as a risk factor and also speculated that it may lead to a weak elongated cervix, especially if it is associated with poor healing at the isthmus.<sup>5,14,24</sup> Uterine torsion can occur iatrogenically following external cephalic version and it is recommended to do ultrasound localization of the placenta prior to and after the procedure for early detection.<sup>25</sup> In our case, the only identified associated factor was the fetal breech presentation. A recent review noted 19.5% of the fetal position are breech.<sup>5</sup> However, the transverse lie is the most common (72%) malpresentation seen in uterine torsion.<sup>2,4</sup> In a woman with multiple predisposing factors, counseling and close follow-up are advised. Authors also advocate limiting or avoiding physical activities in the second and third trimesters of pregnancy.<sup>5,22</sup>

**Table 1: Potential Predisposing/triggering factors for gravid uterine torsion**

<b>Maternal factors</b>	Uterine developmental malformations such as Müllerian anomalies Pelvic mass- myoma and adnexal mass/cysts Pelvic bone or spine (scoliosis) abnormalities Sudden and abnormal maternal movement Abdominal wall laxity /pendulous abdomen Connective tissue disorders such as Ehlers-Danlos syndrome Malpresentation especially-transverse lie Multiple gestations
<b>Fetal factors</b>	Polyhydramnios Hyperactive fetus Placenta previa Fetal anomalies External cephalic version
<b>Iatrogenic and trauma</b>	Previous abdominal surgery including cesarean section Maternal trauma

Considering the rarity and the non-specific clinical features, gravid uterine torsion is easily misdiagnosed and mostly identified intraoperatively.<sup>1,2,5,7,8,22</sup> Kremer et al. described it as a ‘once-in-a-lifetime’ diagnosis for obstetricians.<sup>19</sup> Medical literature reported different patterns of presentation namely acute, sub-acute, chronic and intermittent or recurrent based on duration and degree of torsion.<sup>2,6</sup> If symptomatic, its manifestations range from abdominal pain to life-threatening conditions such as placental abruption with hemorrhagic shock and fetal and maternal death.<sup>2,5,9-18</sup> Other non-specific symptoms include nausea, vomiting, diarrhea, urinary symptoms, pallor, abdominal distension and tenderness, rigidity and obstructed labor.<sup>2,5</sup> Of all these, abdominal pain is the most common, seen in as much as 95% of the cases.<sup>1,5</sup> However, several authors reported asymptomatic cases.<sup>26-29</sup> A recent review revealed nearly one-third (31.7%) of patients had no symptoms.<sup>5</sup> Jansen also reported 11% of asymptomatic gravid uterine torsion in his review.<sup>2</sup> Patient’s symptoms may be obscured by other causes of acute abdominal emergencies in pregnancy like placental abruption, as happened in our patient and others.<sup>9-18</sup> Kopko J et al. described a case of asymptomatic gravid uterine torsion noted accidentally during appendectomy at the 19<sup>th</sup> week of gestation.<sup>30</sup>

Insuspected cases, imaging may assist the preoperative diagnosis. Ultrasound is the first-line imaging for obstetric emergencies and features like a change in placental location, ovarian vessels wrapping around

the uterus (on Doppler ultrasound) and abnormal ovary location may suggest gravid uterine torsion. In setups where magnetic resonance imaging (MRI) is available and if the patient is stable, it may demonstrate a “whirlpool sign” at the uterine isthmus due to a change in the upper vagina from its normal H shape to an X shape.<sup>5,19-21</sup> Since our patient had hemodynamic instability and ANC follow-up at another health facility, it was difficult to retrieve and compare the placental location with the previous ultrasound. The emergency ultrasound examination revealed concealed placental abruption that explained all the manifestations and obscured the uterine torsion.

The patient in our report presented acutely with placental abruption, maternal shock and IUFD. Such presentations are uncommon manifestations of this rare pathology and few cases are reported worldwide (Table 2).<sup>3,9-18</sup> Of the 212 cases reviewed by Jansen only 4% had an abruption.<sup>2</sup> Acute torsion compromises uterine blood flow resulting in increased venous pressure which in turn leads to placental abruption and then fetal death.<sup>3,11-13</sup> As it is noted in our patient, mild vaginal bleeding and concealed abruption may indicate the presence of torsion obstructing the outflow.<sup>11</sup> Shock in a gravid uterine torsion is mostly secondary to hemorrhage from the placental abruption, but in late presentation, it may be caused by infection or bleeding from necrosis of the torsioned uterine tissue.<sup>5</sup>

Table 2-Literature review; Characteristics of reported gravid uterine torsion complicated by placental abruption since 1999

Author, Year (reference)	G A	Degree of Torsion	Hypotension	Fetal Position	Time of Diagnosis	Uterine Incision	Fetal Outcome	Maternal Outcome	Risk factor	Hysterectomy
Carter,1961 <sup>9*</sup>	38	180, left	Borderline	Vertex	intraoperative	Posterior	Died after birth	Alive	None	Not done
Kovavisarach et al, 1999 <sup>3</sup>	36	180, left	Present	---	intraoperative	Anterior	IUFD**	Alive	None	Not done
Duplanteir et al,2002 <sup>10</sup>	38	180, left	---	---	Intraoperative	Posterior	Alive	Alive	Trauma	Not done
Cook et al, 2004 <sup>11</sup>	36	270, right	Present	Vertex	Intraoperative	Not mentioned	IUFD	Alive	None	Done
Munro et al, 2006 <sup>12</sup>	32	180, right	Present	Breech	Intraoperative	Posterior	IUFD	Alive	None	Not done
Gohil et al, 2013 <sup>15</sup>	28	>180, left	Borderline	Breech	Intraoperative	Not done	IUFD	Alive	None	Done (with the fetus in situ)
Sachan et al,2014 <sup>14</sup>	17	380, not described	Present	Transverse	Suspected preoperatively	Anterior	IUFD	Alive	Myoma and previous CS	Uterine artery ligation
Agar et al,2014 <sup>15</sup>	36 <sup>+</sup> <sub>5</sub>	180, right	Absent	Transvers	Intraoperative	Posterior	IUFD	Alive	None	Not done
Zullino et al, 2014 <sup>16</sup>	33	180, right*	Borderline	Transvers	Intraoperative	Posterior	Died after birth	Alive	None	Not done
Ulu et al, 2016 <sup>17</sup>	32	180, not described	Borderline	Vertex	Intraoperative	Posterior (deliberate)	Alive	Alive	Myoma	Not done
Toshniwal,2018 <sup>19</sup>	20	90, right	Present	---	Intraoperative	Posterior (deliberate)	IUFD	Alive	Previous CS	Not done
Our case	37	180, left	Present	Breech	Intraoperative	Posterior	IUFD	Alive	None	Done

+included because it is the only report we found from Africa

\* interpreted as counterclockwise detorsion

\*\*IUFD; intrauterine fetal death

Irrespective of the gestational age and the presence of symptoms, gravid uterine torsion requires immediate surgical exploration to prevent morbidity and mortality to the mother and the fetus.<sup>5</sup> Literature suggested four pathognomonic intraoperative features of uterine torsion such as uterine artery palpable in the vaginal fornix anteriorly or posteriorly, twisting of the vagina and spiraling of the urethra and /or rectum. Furthermore, abnormal location of the ovary, fallopian tube and round ligament, engorged and tortuous vessels, and overlooking of the bladder reflection may also suggest this condition.<sup>2,5,26,31</sup> In the case discussed here, considering the patient's hemodynamic instability and the rarity of uterine torsion, we rushed to stabilize the patient after identifying the 'lower uterine segment' and failed

to detect it before making the uterine incision. Most authors experienced similar challenges intraoperatively especially when complicated by placental abruption and maternal shock (Table 2).<sup>9,10,12,15,16,32-34</sup>

After identifying the torsion, further management depends on the gestational age and status of the uterus (Ischemic or not). Untwisting/derotation should be attempted first, if difficult, as reported by some authors, deliberate incision on the accessible surface (mostly posterior hysterotomy) of the uterus is advised.<sup>2,4,5,17,18,31,35</sup> However, it carries a risk of postoperative adhesion and injury to major vessels, the bladder, and ureters. To avoid injury, it is suggested to make the incision vertical on the fundus or high transverse by observing the bladder plication.<sup>7,9,12,15,28</sup> Others suggested converting

midline incision to facilitate derotation and to prevent the morbidities associated with posterior hysterotomy. However, this was not accepted widely.<sup>5,12</sup> Of the 41 cases reported since 2006, 61 % of them have undergone posterior hysterotomy.<sup>5</sup> In our review of uterine torsion with placental abruption, 7 cases (77.8%) had posterior incisions (Table 2). These patients should be offered elective cesarean deliveries in future pregnancies to avoid labor-induced uterine rupture.<sup>2,4-7,16</sup>

Hysterectomy should be considered in necrotic or ischemic uterus which persisted after detorsion. Furthermore, in situations of severe hemorrhage due to atonicity unresponsive to other means, hysterectomy preserves the patient's life.<sup>5,11</sup> In our case, we performed a hysterectomy for persistent uterine atony causing hemodynamic instability despite medical measures. Gohil et al. described a hysterectomy with the fetus in situ to prevent patient deterioration by avoiding an incision on a hypervascular and congested uterus.<sup>13</sup> As reported by authors, patient deterioration may continue due to amniotic fluid embolism following manual derotation of necrotic uterus.<sup>5,11</sup>

If torsion occurs in earlier periods or prior to fetal viability, detorsion and continuation of pregnancy is suggested.<sup>2,5,22,36</sup> Bukar et al. reported successful delivery at term after manual derotation of a 16-week uterine torsion.<sup>36</sup> Moreover, surgical correction of predisposing conditions can be done simultaneously whenever possible.<sup>1,2,5</sup> In general, when managing this rare condition every effort should be made to save the mother's life as stated by Nesbitt and Corner, "undue regard for the fetus should not deter one from instituting prompt and adequate surgical intervention in an attempt to salvage the mother's life."<sup>1</sup>

Unless timely laparotomy is offered, gravid uterine torsion is associated with maternal and fetal mortality. Moreover, the gestational age and degree of torsion influence both maternal and fetal outcomes. Possible causes of maternal death include shock from bleeding, infection, and embolism.<sup>5</sup> So far there is no report of maternal death before

20 weeks (5<sup>th</sup> month) of gestation and the risk of poor maternal outcome is noted to be high between 5<sup>th</sup> and 8<sup>th</sup> month. A degree of torsion of more than 180° is associated with poor maternal outcomes.<sup>1,2,5</sup> Literature showed a decline in maternal mortality from 15.4% to 2.4% and this is attributed to improvement in timely intervention and resuscitation.<sup>1,2,5</sup> Furthermore, our review of gravid uterine torsion with placental abruption revealed no maternal mortality (Table 2).

Unlike maternal outcomes, perinatal mortality remains a challenge. Nesbit et al., Jensen, Wilson et al., and Ramseyer et al. reported 30.4%, 12%, 18% and 22% respectively.<sup>1,2,4,5</sup> Among the 11 cases investigated in our review, the overall perinatal mortality is 82% (9 cases) (Table 2). This indicates that patients who have associated placental abruption are at high risk of perinatal mortality. In our case, late presentation and degree of rotation contributed to maternal morbidity and fetal death.

## CONCLUSION

In pregnancy-related acute abdominal emergencies, one has to keep in mind the possibility of uterine torsion due to its non-specific nature and dangerous impact. Our case and other reported literature showed that it can be obscured and associated with placental abruption especially when concealed and accompanied by maternal shock and fetal distress/death in the third trimester. Early diagnosis and prompt laparotomy are crucial for favorable maternal and fetal outcomes. Assessing for anatomic landmarks before uterine incision helps in the diagnosis of uterine torsion and avoids inadvertent vascular and ureteral injuries during hysterotomy. Moreover, an attempt to reverse the torsion should be made always before doing a planned posterior hysterotomy.

## ETHICAL APPROVAL

The patient provided informed written consent to use the clinical information and images for publication. Since it is a case report, approval from the institutional review board is not required.

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