A 26 year old G2P1001 parturient was admitted at 32 weeks of pregnancy for obstetric services. She was a diagnosed case of placenta previa grade III with placenta accreta. Ultrasonography (USG) at 16 weeks of pregnancy showed normal fetal skull, spine and stomach. At 26 weeks of pregnancy USG showed low lying placenta anteriorly covering internal os, moderate to severe fetal hydrocephalus with dilated lateral ventricles (1.9 cm), single upper limb and short other limbs, skeletal dysplasia and over-distended fetal abdomen with large cystic masses. Level II obstetric USG at 28 weeks showed fetal congenital high airway obstruction and free fluid in fetal abdomen. In her last pregnancy a singleton healthy male baby was delivered by classical cesarean incision if the maternal bleeding will be under control. Surgery was allowed to start. Uterus was opened and covering internal os, moderate to severe fetal hydrocephalus with dilated lateral ventricles (1.9 cm), single upper limb and short other limbs, skeletal dysplasia and over-distended fetal abdomen with large cystic masses. Level II obstetric USG at 28 weeks showed fetal congenital high airway obstruction and free fluid in fetal abdomen. In her last pregnancy a singleton healthy male baby was delivered by elective cesarean section for breech presentation. She was hypothyroid since 3 years and was taking tablet thyroxine sodium 100 mcg once a day. No other comorbidities were present. On examination she was 65 kg in weight and 156 cm in height. Cardiovascular and respiratory system examination revealed no abnormality. Her complete haemogram showed haemoglobin of 10.2 gm/100 ml, total leucocyte count of 7400/mm³ and platelet of 202000/mm³. Her thyroid functions, liver functions, renal functions and coagulogram were within normal limits. Blood pressure was 110/80 mmHg and heart rate was 82/minute. Airway examination revealed mouth opening of 3 cm. She was mallowpatti class II with full range of neck movement. Tablet ranitidine 150 mg and metoclopramide 10 mg was given night before surgery and in morning on day of surgery. In the operating room, standard 5 leads ECG, NIBP and pulse oximetry were attached and baseline parameters were noted. Two large bore (16 G) venous cannulae were secured in upper limbs and normal saline infusion was started. ENT and pediatric surgeons were kept ready for surgery and procedures on baby. After preoxygenation with 100% for 5 minute (target endtidal oxygen >90%), rapid sequence anesthesia was induced with IV thiopentone 325 mg and IV suxamethonium 125 mg. Trachea was secured with 7.0 mm ID tracheal tube. IV atracurium 25 mg was given once the effect of suxamethonium was tapered. Anesthesia was maintained with isoflurane in a mixture of oxygen and nitrous oxide (30:70). Right internal jugular central venous catheterization was done and radial artery was cannulated for continuous invasive blood pressure monitoring. Surgical plan was to perform ex utero intrapartum treatment (EXIT) on fetus after classical cesarean incision if the maternal bleeding will be under control. Surgery was allowed to start. Uterus was opened and baby was delivered out. As soon as the baby was delivered profuse bleeding was started from uterus and placenta. It was decided not to perform EXIT procedure and cord was clamped and cut. Bleeding was continued from uterus and placenta. Three units of packed red blood cells and three units of fresh frozen plasma were transfused. Meanwhile ENT surgeon performed tracheostomy on baby and airway was secured but baby died after half an hour. Emergency cesarean hysterectomy was done after all measures to control the bleeding were failed.
Patient was electively mechanically ventilated for 12 hours in post anaesthesia care unit with close monitoring. Trachea was extubated once the patient becomes conscious and her arterial blood gases were within normal limits. Post-operative course of mother in PACU and ward was uneventful and she was discharged from hospital on 7th post-operative day.

Discussion

Placenta praevia is a major cause of maternal obstetric haemorrhage. If placenta praevia is complicated by accreta and percreta the risk is even higher. As a result of the increasing number of surgical deliveries incidence of these conditions are rising [1,2]. Placenta accreta occurs most frequently in women with one or more prior cesarean deliveries who have a placenta previa in the current pregnancy. According to Clarke et al. [3], in the presence of a placenta previa, the risk of having placenta accreta increased from 24% in women with one previous cesarean section to 67% in women having 3 or more cesarean sections. Hysterecogy is still main treatment to control the bleeding in these conditions with higher maternal morbidity and mortality.

Massive obstetric hemorrhage as a result of placenta previa and accreta may result into complications as injury to the ureters, urinary bladder, and other abdominal viscera, disseminated intravascular coagulopathy, acute lung injury, adult respiratory distress syndrome, acute renal failure, and even mortality [4]. Placenta accreta is one of the leading reason for cesarean hysterectomy [5]. Minor placenta accreta may lead to slightly heavier postpartum bleeding, but may not require hysterecogy but extensive placenta accreta often require. The standard management of these conditions is to leave the placenta in situ, with no attempt at removal. Hysterecogy can be performed 2 to 6 weeks later on an elective basis. Pelvic arterial embolization or balloon catheter occlusion significantly reduces uterine blood flow and allow reduced blood loss during surgery [6]. Other adjunctive procedures like methotrexate therapy, uterine compression sutures, internal iliac vessels embolization, resection of the affected segment of the uterus and oversewing of the placental bed may help [7-9].

Congenital high airway obstruction syndrome (CHAOS) is an ultrasonographic antenatal diagnosis comprising extremely large echogenic lungs, pleural or pericardial effusions, a dilated tracheobronchial tree, inverted or flattened diaphragms, fetal ascites with nonimmune hydrops and complete or almost complete upper airway obstruction [10-14]. These findings result from increased intratracheal pressure and distention of the tracheobronchial tree secondary to the accumulation of fluid in the lungs. Laryngeal atresia, laryngeal web, laryngeal cyst or tracheal atresia may be the causes of airway obstruction. The incidence of CHAOS is rare [10]. Skeletal and vertebral malformations, tracheoesophageal fistula, esophageal atresia, genitourinary malformations, limb deformities and cardiac malformations are usually associated with CHAOS.

The EXIT procedure during cesarean section requires specific anesthesia considerations and techniques and ultimate goal is a healthy mother and infant. The success mainly depends on a well-planned anesthesia technique. The EXIT procedure, in contrast to a routine cesarean section, mandates proper uterine relaxation before uterine incision with the use of high concentration of inhaled anesthetic agents [15]. Complete uterine relaxation is required for delivering the fetal head, shoulders, and if there is any large neck mass or goiter in fetus which are normally unable to pass through a normal lower segment uterine incision. Uterine perfusion is main determinant of fetal oxygenation and has to be maintained by proper uterine relaxation. Also the procedure on fetus requires time and during this period fetus needs inhalational anesthesia via transplacental transfer. Complete uterine relaxation places the mother at risk for massive intraoperatively bleeding from placenta and uterus. Also high concentration of inhaled anesthetic causes more hypotension intraoperatively from profound vasodilatation and if not treated timely and promptly, causes fetal hypoxia from decreased uterine perfusion. Short-term maternal outcomes do not differ between those patients receiving EXIT procedures and those patients undergoing cesarean sections if not associated with any placental abnormality.

In our case fetal CHAOS was associated with maternal previa and accreta. Placenta was low lying and covering internal os. The surgical plan was to perform the classical cesarean section under deep plane of general anesthesia and EXIT procedure (fetal tracheostomy) was planned by ENT surgeons after delivering the fetus through classical cesarean incision. Unfortunately, after uterine incision, there was profuse bleeding from uterus and the whole surgical field was bloody. EXIT procedure was cancelled and fetal tracheostomy was done on fetal table. Maternal blood pressure also became low and ephedrine boluses were given to maintain the maternal haemodynamics. Immediately inhaled anesthetics concentration was reduced and blood products were transfused. All measures to control the bleeding were failed and emergency cesarean hysterectomy was done. Blood loss was around 2500 ml.

Even after thorough search, we did not encounter any study or reported case presenting EXIT procedure in fetus with maternal placenta previa and accreta. Massive maternal bleeding in our case may be due to the deep plane of inhaled anesthesia in addition to the maternal placental abnormalities. Also by classical cesarean incision, mostly the placental site can be avoided, but in our case even after classical incision, bleeding was profuse and even experienced senior gynaecologist failed to control the bleeding.

The aim of the present case is to highlight the possible risk to maternal health if CHAOS or any fetal airway abnormalities are associated with placental abnormalities. Yet more discussion and modalities require if EXIT procedure has to be performed for saving the infant.

References


