

Airway Management of Caesarean of A Parturient with Severe Idiopathic Subglottic Stenosis: A Case Report

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1. Abstract

We described an infrequent case of parturient suffering from idiopathic subglottic stenosis (iSGS) before pregnancy who was contra-indicated at term to peri medullar analgesia related to a deep thrombocytopenia. A multidisciplinary management was decided with anaesthesiologists, ENT surgeons and obstetricians. General anaesthesia with a small endotracheal tube was immediately completed after delivery by tracheal bougie dilatation. Postoperative extubating was uneventful, and the patient was discharged at day 7 with her baby. This case highlights the importance of anticipated multidisciplinary prepartum strategy about airway management and the central place by the anaesthesiologist to coordinate actors and to ensure maternal safety.

2. Introduction

Tracheal intubation is responsible for most of the subglottic stenosis [1]. Other etiologies are due to tumours, malformations, immune pathologies or without intensified causes, also called idiopathic subglottic stenosis (iSGS). iSGS affects principally young to middle-aged women and estrogens are probably involved in the mechanism of the stenosis [2-3]. The treatment of iSGS mostly relies on endoscopic tracheal dilatation techniques which are less invasive than surgery, but at risk of recurrence

with need of iteratives dilatations [4]. Airway management can be difficult or impossible and concertation with the ENT surgeon is required before any surgery to decide the best strategy [2]. iSGS is a rare situation, poorly described during pregnancy, as the present case about a parturient with iSGS who underwent a caesarean delivery under general anaesthesia (GA). Informed written consent has been obtained from the patient for the purpose of publication, which adheres to applicable CARE guidelines.

3. Case Description

We report the management of a 32 years-old gravida 1 para 1 woman. She only suffered of a circumferential iSGS that was successfully treated five years ago by tracheal dilatation with no residual symptoms. Since the beginning of her pregnancy, the parturient reported an increased inspiratory dyspnea with severe limitation of her daily activities. Recurrence of the iSGS was confirmed during an awake fiberoptic exam on the 6th and 8th month with an indication to perform a dilatation after delivery (Figure 1). To avoid any expulsive efforts with a risk of respiratory distress during the vaginal delivery, a caesarean delivery under spinal anaesthesia was planned for the 39th week of gestation.

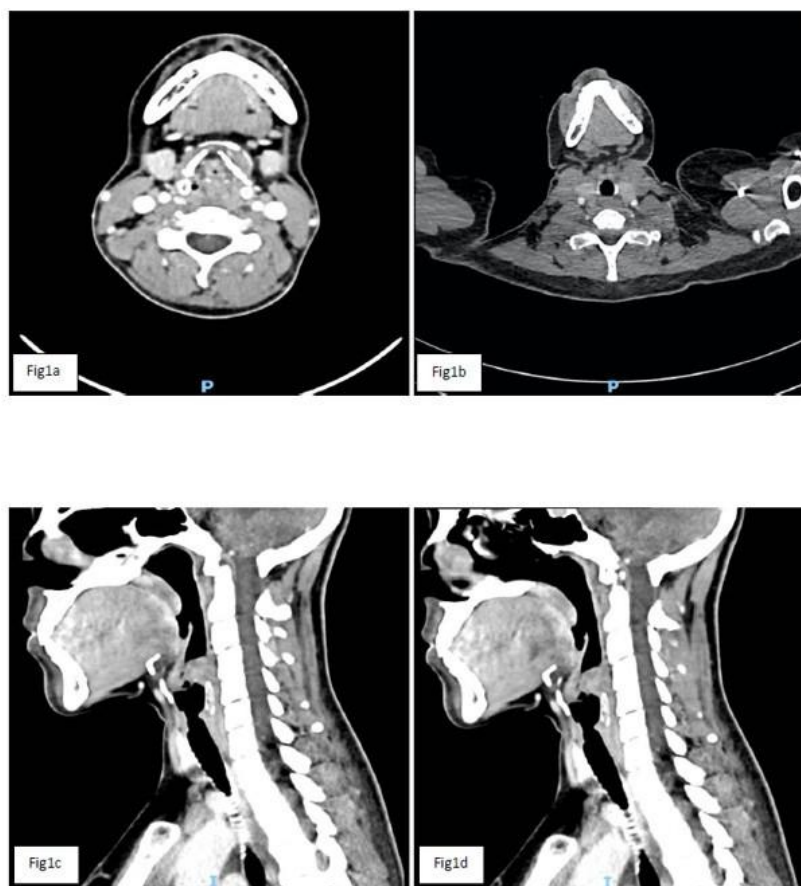


Figure 1: Cervical scanner (axial section) focus on subglottic stenosis (Figure 1a) and trachea at the beginning of his thoracic portion (Figure 1b) recovering normal diameter. Cervical scanner (parasagittal section) focus on subglottic stenosis (Figure 1c & 1d).

However, a severe drop of the platelets count occurred on the 8th month, that was compatible with an idiopathic thrombocytopenic purpura because of the deepness and the fast decrease of platelets. Introduction of corticosteroid therapy (1mg.kg-1 per day), then intravenous immunoglobulins (Privigen 1g.kg-1) failed to increase the platelet number. Consequently, a multidisciplinary concertation with ENT surgeons, obstetricians, internists, and anaesthesiologists decided to continue current treatments and discussed the adjunction of Nplate (thrombopoietin receptor agonist) as the second line of treatment. Anyway, to anticipate a risk of non-response, a caesarean under general anaesthesia at 39 weeks of gestation was planned with the ENT surgeon to perform a tracheal dilatation at the same time or an emergency tracheotomy in case of impossible intubation. But 48 hours before the planned delivery and during the night, the patient spontaneously started the labour with close uterine contractions, and platelets were still low at 40G.L-1 with an absolute contra-indication to spinal anaesthesia. Patient was then conducted into the operating room and before proceeding with GA, a briefing was performed with every physician involved in this situation to check and repeat the timing of the procedure. From anaesthesiologist point of view, we checked that the difficult intubation cartridge including small endotracheal tube, jet ventilation, and cricothyroid kit was immediately available. Two senior anaesthesiologists were present in case of impossible upper airway catheterization. A check list was also done for the neonatal reanimation equipment and

surgical instruments for tracheal dilatation (Figure 2).

After pre-oxygenation in a 30° reverse Trendelenburg position with high-flow nasal cannula oxygen therapy, a rapid sequence intubation including remifentanyl + propofol and rocuronium was administrated and a quick control of the airway succeeded using video laryngoscopy (McGrath) with insertion of a very small tracheal tube (internal diameter of 4 millimetres according to the preoperative imagery) under ENT supervision. A male infant weighing 2850 grams was born through a low transverse uterine incision with Apgar scores of 1, 9 and 10 at one, five and ten minutes respectively, and this baby was quickly transferred to neonatal unit. Umbilical cord was clamped, carbeto-cin was injected and the obstetricians closed the hysterotomy. Uterine atony was rapidly diagnosed during wound closure and intravenous Sulprostone was immediately started combined with 1000mg of tranexamic acid. Total bleeding was estimated around 550mL. Analgesia was ensured by Paracetamol, Nefopam and ultrasound-guided Transversus Abdominal Plane block with a compressive bandage

Once the caesarean was done, and bleeding under control, ENT hesitated to perform immediate dilatation related to the severe thrombocythemia and suggested withholding the parturient from mechanical ventilation. But after a short discussion, the team decided to strictly follow the common multidisciplinary decision, and the surgeon performed rigid bronchoscopy and dilatation of subglottic stenosis along the tracheal tube to keep the airway

safe. At the end, intubation with a medium size endotracheal tube (internal diameter of 6 millimeters) was possible. The patient slept under GA for two hours to optimize the benefice of the tracheal dilatation. A delayed extubating was performed without any complication and systematic aerosolized corticosteroid were introduced to minimize local oedema. Postoperative evolution was simple, and the patient was discharged from hospital seven

days after caesarean.

Despite deep thrombopenia below 50G.L-1, neither haemorrhagic complication occurred, and no transfusion of platelets was required. Platelet count at one month was normal and idiopathic thrombocytopenic purpura was biologically confirmed (Figure 3).

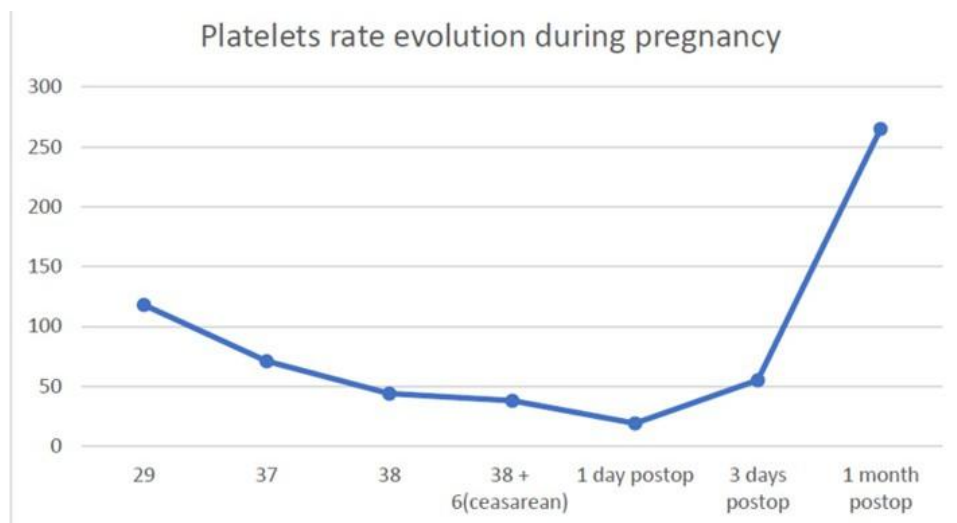


Figure 2: Platelet rate evolution at the end of the pregnancy.

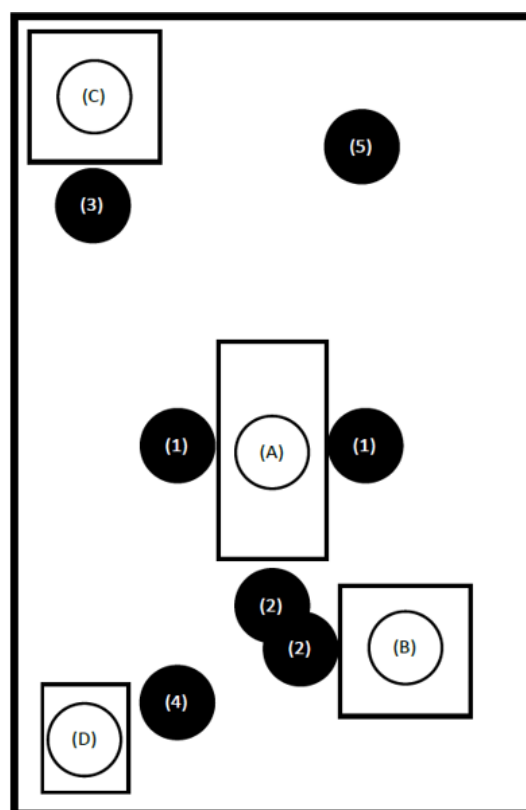


Figure 3: Schematic representation of operating room. (1) Gynecologist surgeon and his aid. (2) Anesthesiologist doctor and anesthesiologist nurse. (3) Pediatrician. (4) Otorhinolaryngologic surgeon. (5) Anesthesiologist team leader of procedure. (A) Operating table. (B) Respiratory and anesthetic equipment. (C) Incubator and neonatal reanimation equipment. (D) Surgical tracheal dilatation equipment.

4. Conclusions

Airways management of a parturient with subglottic stenosis is challenging because it is very infrequent and associated to potential risks. Intubation and/or ventilation difficulties can be harmful for the patient and lead to major injuries such as neurological

damage or death either the mother or her baby [8-9]. Added to them, proper risks due to physiological modifications of pregnancy including upper airways oedema, decreased of pharyngeal space and increased Mallampati score, higher risk of hypoxemia and lower tolerance of apnea ventilation, oesophageal sphinc-

ter incompetence and increased gastric liquid inhalation, higher risk of preoperative memorization [10-11]. Specific preoperative assessment is required for all patients undergoing tracheal intubation to detect potential difficult airway management and to decide appropriate strategy. Most often, balloon dilatation or ENT surgery have been proposed during pregnancy to anticipate the risk of difficult airway during labour and some publications reported success for this strategy.

This case combined idiopathic subglottic stenosis and severe thrombocytopenia. In this situation, GA was the ultimate solution to avoid shortness of breath parturient during labour and it required different experts. Anticipation of this complex obstetrical situation with a multidisciplinary conference, strategy and immediate briefing was important to ensure optimal safety for the mother and the baby whatever the conditions. Aesthetic strategy was discussed and validated collegially to perform caesarean under GA on the first position the Monday morning. But the patient went into labour spontaneously making it impossible to delay surgery. The caesarean delivery occurred at the weekend during the night with restricted staff and with other practitioners than those involved in the strategy except one. This case also highlighted the central place by the anaesthesiologist during the procedure who knew exactly the original plan and who decided prioritization when an unexpected postpartum haemorrhage occurred. The discussion about eventual delayed dilatation illustrates this point and this strategy was probably the best for the parturient. A special mention was the physical presence of ENT surgeon during delivery who reassured the anaesthesiology team during intubation in case of impossible airway management requiring tracheostomy and who performed at the same time tracheal dilatation avoiding postoperative respiratory complications.

Finally, discussion about deep thrombocytopenia is interesting because it was possible to manage this parturient without need of platelets transfusion. Thrombopenia is one of the most frequent biological disorders during pregnancy and its physiopathology associate's dilution by increased plasma volume and intensified platelets destruction during third trimester [5]. Also called "gestational thrombocytopenia", it does not pre-exist before pregnancy, brings no haemorrhagic complication, and corrects by itself spontaneously after delivery. Differential diagnosis between "simple" gestational thrombocytopenia and immune thrombocytopenia, such as thrombocytopenic idiopathic purpura, is not so easy. Thus, diagnostic delays concerning aetiology of end pregnancy thrombopenia may lead to inappropriate prophylactic platelets transfusion [6] and to reach platelets cut-off recommended by guidelines [7]. In this case, we performed caesarean with deep thrombopenia and transfusion rational based on the constataion of haemorrhagic complications.

To conclude, the presence of subglottic stenosis during pregnancy is a singular and challenging situation for the anaesthesiologist without any guidelines. Literature about this situation is

poor with only few cases reporting treatment of subglottic stenosis during pregnancy [12-15]. To perform a GA to control airway was possible under ENT supervision in case of difficulty with an immediate dilatation despite thrombopenia without any harmful consequence.

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