CASE REPORT

Conservative management of subglottic stenosis in pregnancy resulting in vaginal birth

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SUMMARY

We present a case of subglottic stenosis complicating pregnancy. The patient was born prematurely at 24 weeks gestation and required a twelve-month period of intubation. Airway trauma from prolonged intubation resulted in acquired subglottic stenosis. As an adult the patient had a longstanding audible stridor; however, was not breathless during activity before or during pregnancy. The patient went into spontaneous labour at 37 +4 weeks and delivered vaginally with epidural analgesia. This case is significant, as no cases of a patient with such a degree of stenosis delivering vaginally without airway treatment have been reported.

BACKGROUND

Subglottic stenosis (SGS) is more common in women¹ and can be acquired after trauma. Up to 90% of cases occur after prolonged intubation. More rarely SGS is seen as a result of granulomatosis with polyangiitis (previously Wegener's granulomatosis) and in as many as 20% of cases, a cause cannot be found.²

The most common presenting symptom is exertional dyspnoea and in more severe stenosis, stridor is seen. These symptoms are frequently misdiagnosed as asthma.³ Pregnancy can result in a dramatic worsening of symptoms, and cases where life saving airway intervention with balloon dilation, laser treatment or tracheostomy became necessary, intra-partum have been reported.^{3 4} An alternative presentation of disease can though result in SGS going unnoticed until difficulty intubating at the time of administering a general anaesthetic (GA).^{1 5 6}

Although rare, the importance of this case rests in the dramatic and potentially fatal complications that can arise from worsening obstruction in pregnancy or difficulty in intubating and ventilating should a GA be required.

CASE PRESENTATION

A primigravida in her early 30s, presented for booking in the first trimester of pregnancy at a large tertiary hospital. She herself was born at 24 weeks gestation and spent the first year of life intubated and ventilated. The patient recalled of having always an audible stridor and hoarse voice; however, she had been able to run 10 km prior to pregnancy. Her breathing worsened periodically when suffering from chest infections and these occurred on average three times a year.

No procedures had been carried out on her airway and she had undergone an appendicectomy under GA at the age of 15 without complication. Details of

the management of her airway were not recalled by the patient and could not be obtained. She did not though remember any complications arising.

Additional medical history included bilateral retinal detachment, treated under local anaesthetic, and hypothyroidism. She was clinically euthyroid on thyroxine and her booking body mass index was 23.

Pregnancy was initially complicated by several episodes of painless bright red antepartum haemorrhage secondary to a low-lying placenta. These episodes were sufficient to require admission to hospital although her haemoglobin remained above 10.5 g/dL and no transfusions were required. Towards the end of pregnancy, it was no longer considered low lying by ultrasound assessment.

At 35 weeks gestation, mild pre-eclampsia developed and was managed successfully with labetalol 100 mg twice daily.

The diagnosis of SGS prompted significant input from anaesthetic colleagues and referral to an ear nose and throat (ENT) surgeon to make a surgical airway assessment.

INVESTIGATIONS

ENT review at 34 weeks with flexible nasendoscopy revealed that the arytenoids were overlapping and that it was impossible to pass the scope beyond the vocal cords.

Subsequent MRI showed narrowing at the glottis and subglottic region. At the level of the false cords, the airway appeared narrowed to 2.5 mm. SGS was seen for 2–4 mm below the true cords with a maximum diameter of 3 mm at this level. Superior and inferior to the stenosis the lumen was adequate, being 10 mm at the cricoid.

OUTCOME AND FOLLOW-UP

At 37+4 weeks the patient experienced spontaneous rupture of membranes. A normal cardiotocography was seen and labour established within 3 h. An early epidural was sited and progress was quick to full dilation in 3 h. After 2 h to allow descent active pushing began. The second stage became prolonged and delivery was expedited with low cavity forceps.

The third stage was complicated by retained placental tissue and second-degree perineal tear, resulting in a 2.4-litre postpartum haemorrhage. Two units of blood were transfused.

A healthy neonate was delivered with APGAR 9 at 1 min and 10 at 5 min. The remainder of the postnatal period was uneventful with satisfactory review at 6 weeks. The patient's airway remained stable in the puerperium.

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DISCUSSION

When the patient was experiencing episodes of antepartum haemorrhage an elective caesarean section was planned for obstetric reasons. With the resolution of the low-lying placenta and maternal desire for vaginal delivery, debate arose around the safest method of delivery.

Literature search showed that the existing published material is limited to case reports. Ten case reports were found, of which two cases of stenosis were discovered at the time of failed intubation for a category 1 caesarian section. Anaesthetic colleagues fear this situation as it can lead to difficulty intubating and ventilating.^{1–5} This fear was the motivation behind an ENT review recommending a planned caesarean section.

In eight cases SGS was recognised in the antenatal period and management was considered.^{3–6–10}

Worsening respiratory symptoms prompted treatment with balloon dilation or laser in four cases. Of these women three underwent spontaneous vaginal delivery and one underwent elective caesarean section with a spinal anaesthetic.^{6–7} All were uncomplicated. Additionally a case was reported of a patient requiring antenatal tracheostomy.⁸

One case described an emergency caesarean section in a patient with SGS for worsening pre-eclampsia that was carried out successfully with a spinal anaesthetic.³ Obstetrically indicated elective caesarean section for breech presentation with a spinal anaesthetic was also reported.¹⁰

Caesarean section though brings the additional possibility of conversion to GA for operative reasons as reported in one SGS patient where conversion was required intraoperatively because of profuse maternal bleeding.⁹

Some cases have reported the use of respiratory function tests, and the role of this has been to show changes from a baseline to after surgical airway intervention.³ Although with hindsight it would have been interesting to measure this in the case reported, it was not undertaken as our patient's airway was clinically stable during pregnancy with no worsening of stridor or new onset of breathlessness. It would also not have changed the management of the patient, as surgical intervention was not being considered.

The primary concern of the clinical team was avoiding the possibility, however small, of needing to intubate the patient for a GA. This was felt most likely with a category 1 emergency caesarean section where no epidural was in place or for a failed regional anaesthetic.

Evidence available from an audit of regional anaesthetic failure in 5080 caesarean sections gave the likelihood of conversion to GA during caesarean as 1.2% with an epidural top up and 0.8% with a spinal anaesthetic.¹¹ Although epidural is more likely to fail than spinal, the authors felt this was not significant enough to preclude epidural and attempted vaginal delivery as desired by the patient, especially when factoring in the potential for operative complications from a caesarean section.

A plan was therefore made to induce labour with careful timing to give the best chance of delivery during the day when a full clinical team with knowledge of the patient was in hospital. This included a consultant obstetrician and a consultant anaesthetist with the experience of difficult airway management and fibre optic intubation. When the patient went into spontaneous labour the skills of the available personnel were considered and felt to be appropriate. It was ensured that the team and the fibre optic equipment were available at all times. The on call ENT registrar was also informed in case of the very unlikely event that a surgical airway was required.

Adequate analgesia with early epidural placement reduced pain and the associated increase in respiratory effort.¹² Epidural

also provided the ability to top up quickly if a category 1 caesarean was required. The second stage of labour resulted in increased cardiorespiratory effort however, the patient tolerated this.

In reporting successful conservative management of severe SGS in pregnancy, we suggest that vaginal delivery is not significantly more likely to result in airway complication, than elective caesarean section, and should therefore be considered as per maternal choice. The key factors for success in this case were the patient's stable airway, high level of motivation and close cooperation between the patient and the medical team.

We also suggest that radiological evidence alone is not a strong predictor of outcome with less severe obstructions reporting complication.⁴ Clinical assessment of preconception and intrapartum exercise tolerance is likely to provide the strongest predictor of outcome.

Learning points

- ▶ The primary concern in managing subglottic stenosis in pregnancy is avoiding the need for intubation.
- ▶ Regional anaesthesia with epidural is more likely to fail and necessitate conversion to general anaesthesia than spinal however, this is not significant enough to necessarily preclude epidural.
- ▶ Early epidural placement limits the requirement for increased maternal respiratory effort.
- ▶ Exercise tolerance pre- and intrapartum is a good indicator of ability to tolerate labour and more reliable indicator than radiological imaging alone.

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Competing interests None.

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