

## Main Article

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## Abstract

**Objective.** To describe the use of balloon dilation with non-invasive ventilation in the treatment of pregnant patients with idiopathic subglottic stenosis.

**Methods.** The medical charts of four consecutive patients who underwent jet ventilation or high-flow nasal cannula oxygenation with balloon dilation for the treatment of idiopathic subglottic stenosis during pregnancy were reviewed.

**Results.** Objective improvement of subglottic stenosis was seen in all four cases, with end-result Myer–Cotton grade 1 lesions down from pre-procedure grade 3 lesions. Patients also reported subjective improvements in symptomatology, with no further airway issues. All patients delivered normally, at term.

**Conclusion.** Laryngeal dilation with continuous radial expansion pulmonary balloons using non-invasive ventilation for the treatment of idiopathic subglottic stenosis in pregnant patients is safe and efficacious, and should be the first line treatment option for this patient population. The improvement in symptoms, and lack of labour and pregnancy complications, distinguish this method of treatment from others reported in the literature.

## Introduction

Idiopathic subglottic stenosis is a rare disorder, principally affecting females aged 20–40 years. It is characterised by progressive fibrosis with annular stenosis of the subglottic larynx (Figure 1). Various theories have been proposed as to the pathogenesis of the disorder.<sup>1–4</sup> Symptoms usually include progressive dyspnoea, hoarseness and cough. The disorder is frequently mistaken for asthma, and diagnostic delay is common. Diagnosis is made by endoscopic evaluation and identification of the stenosis. Biopsy typically reveals fibrosis and/or chronic inflammation.

Treatment options for idiopathic subglottic stenosis include periodic dilation, cricotracheal resection and tracheostomy. A variety of endoscopic techniques have been described, including: dilation (rigid or balloon), with or without radial incisions; cryotherapy; mitomycin C application; and local steroid application.<sup>5–10</sup> Because of the recurrent nature of the disease, periodic retreatment is common.

There is no clear consensus on the optimal management of the disorder. Because idiopathic subglottic stenosis predominately affects females, often of child-bearing age, management of the disorder in pregnancy may be necessary. However, no clear treatment paradigm for these patients currently exists. Patients may become progressively symptomatic during the course of pregnancy, whether from progression of idiopathic subglottic stenosis, increased weight gain or both.

This study aimed to describe the use of balloon dilation, coupled with jet ventilation or high-flow nasal oxygenation, in the treatment of pregnant patients with idiopathic subglottic stenosis. We present four cases of idiopathic subglottic stenosis during pregnancy. In addition, we review the current literature, and provide recommendations for the evaluation, treatment and follow up of pregnant patients with idiopathic subglottic stenosis.

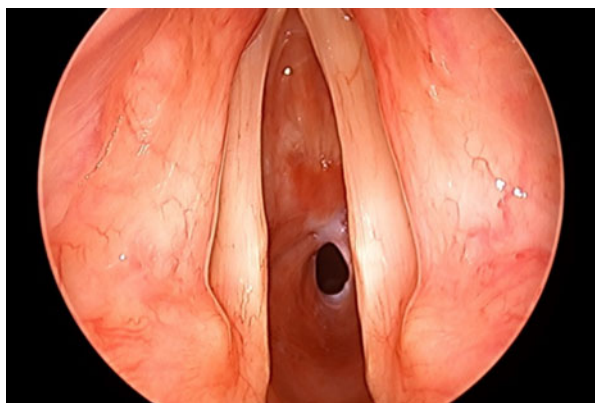
## Materials and methods

### Study design

This study was approved by the Institutional Review Board of the Stanford University Medical Center. The computer database of Stanford Hospital and Clinics, between 1 October 2003 and 30 September 2017, was used to identify those pregnant patients with subglottic stenosis who underwent airway dilation by the senior author. Four patients were identified, all of whom were treated in the manner described below.

### Surgical technique

All of the airway dilations were performed in a standardised manner, with the patients under general anaesthesia. Patients were placed in a semi-recumbent position,



**Fig. 1.** Myer–Cotton grade 3 subglottic stenosis.

and fetal heart rate monitoring was instituted as indicated. For each procedure, intravenous sedation with midazolam and fentanyl or remifentanyl was used. Anaesthesia induction and maintenance were achieved with propofol. Rocuronium or succinylcholine allowed for paralysis to facilitate mask ventilation.

After the induction of anaesthesia, an adult Dedo–Pilling laser laryngoscope (Teleflex, Morrisville, North Carolina, USA) was introduced transorally, and suspended from a Mayo stand to visualize the glottis (Figure 2). Supraglottic jet ventilation was initiated in three cases; in one case, ventilation was performed using transnasal humidified rapid-insufflation ventilatory exchange (Optiflow™, Fisher and Paykel, Irvine, California, USA). A 0-degree 4 mm endoscope was advanced through the laryngoscope to visualise and grade stenosis severity using the Myer–Cotton grading scale (1–4).

Dilation was performed with a 16 mm airway balloon (Inspira Air; Acclarent, Irvine, California, USA) (Figure 3). The balloon was inflated to 4 atmospheres of pressure for 30 seconds. Repeat dilation was performed if needed depending on the stenosis response.

One percent lidocaine with 1:100 000 adrenaline-soaked pledgets were placed into the trachea and glottic area for haemostasis. In order to reduce fibrosis, triamcinolone-soaked (40 mg/ml) pledgets were applied for 1 minute at the level of the stenosis following dilation.

Mask ventilation was utilised if needed to assist emergence from anaesthesia. Fetal heart tones were reassessed in the post-anaesthesia care unit. Patients were discharged home the same day following obstetrical reassessment.

## Results

Four patients required dilation during the course of their pregnancies. In three cases, the patient had a history of prior dilation for known subglottic stenosis, and in one case, the diagnosis was made during the course of pregnancy. Outcomes are shown in Table 1.

All patients had regular obstetrical and pre-natal care. An obstetrical consult was obtained prior to surgery for clearance of the patients. External fetal heart rate monitoring was performed for confirmation of fetal wellbeing before and after surgery in two of the cases, and continuous fetal heart monitoring was performed in the other two cases. No patient required further dilation through the completion of pregnancy. All four patients delivered full-term and fully viable infants via normal spontaneous vaginal delivery at a later date.



**Fig. 2.** Dedo–Pilling laryngoscope with jet ventilation side port.

## Discussion

Subglottic stenosis commonly arises secondary to endotracheal intubation, external trauma, or granulomatosis with polyangiitis (Wegener's granulomatosis).<sup>1–5</sup> It may also be idiopathic, as was the case in the series of patients presented here. Treatment options generally include dilation, cricotracheal resection and tracheostomy. Patients with idiopathic subglottic stenosis tend to be females of reproductive age.<sup>4,5</sup>

Given the epidemiology of the disease, subglottic stenosis may present a therapeutic challenge in patients who become pregnant. It is well understood that maternal distress may have potentially devastating effects on the course of the pregnancy.<sup>11,12</sup> Treatments likely to induce premature labour, or cause contractions, maternal hypoxia or acidosis, must be avoided.<sup>13</sup> In order to minimise the risk of these complications, it is important to select a treatment method that is efficacious in maintaining the airway and oxygenation, and yet is minimally invasive, simple, expeditious, and minimises maternal and fetal stress.

Management of subglottic stenosis in pregnancy has only rarely been reported in the literature, and is limited to a small number of case reports (Table 2).<sup>12–26</sup> Management options have included no intervention, intubation, dilation under inhalational anaesthesia, dilation using topical anaesthesia with or without intravenous sedation, and tracheostomy. In one case, the patient expired several weeks after delivery from subsequent airway obstruction.<sup>14</sup> The literature offers, therefore, no clear consensus on the optimal management, both surgical and anaesthesia, in this group of patients.

Pregnancy incurs significant physiological and metabolic demands on the maternal body. The mother's respiratory system is already altered by pregnancy itself,<sup>27</sup> and the added complication of idiopathic subglottic stenosis and potential for hypoxemia can be harmful to both the mother and fetus. If inadequately treated, idiopathic subglottic stenosis in non-pregnant patients can progress, and could result in pulmonary hypertension, oedema and respiratory failure.<sup>9,28</sup> In pregnancy, inadequate oxygenation may be causally associated with ischaemic placental complications, including abruption, pre-eclampsia and intrauterine growth restriction.<sup>29</sup> Although these complications are rarely caused by idiopathic subglottic stenosis, pregnant patients who have pre-existing idiopathic subglottic stenosis, or who develop idiopathic subglottic stenosis during pregnancy, may require treatment in order to prevent their occurrence.

There remains no consensus regarding the optimal treatment for idiopathic subglottic stenosis in non-pregnant patients. Some practitioners employ purely endoscopic techniques, such as radial incisions, balloon dilation, cryotherapy,

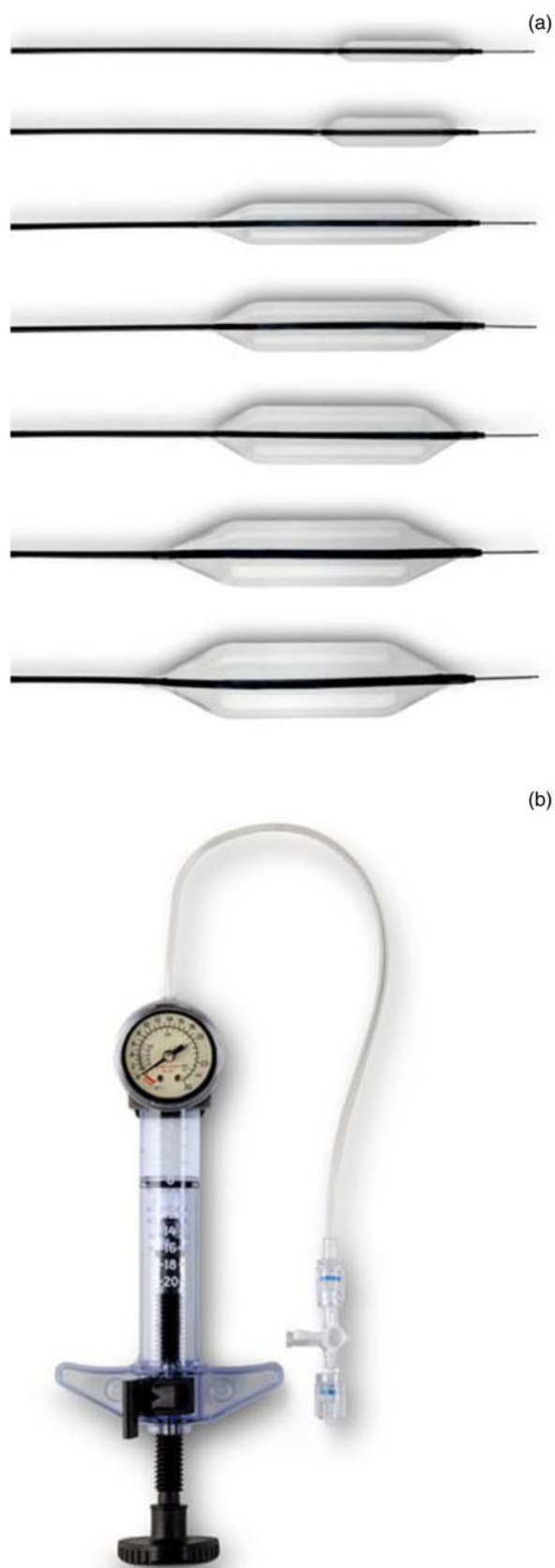


Fig. 3. Acclarent continuous radial expansion balloon (a) and injector (b).

topical mitomycin C or a combination of these. However, long-term re-stenosis rates are between 40 and 95 per cent.<sup>8–10</sup> On the other hand, open techniques such as tracheal resection with re-anastomosis appear to offer higher success rates. Because these techniques are considerably more invasive,

Table 1. Patient pre-operative and treatment characteristics

Characteristic	Case 1	Case 2	Case 3	Case 4
Age (years)	40	30	33	42
Fetal gestational age (weeks)	28	31	17	30
Prior dilations?	No	Yes	Yes	Yes
Operative details				
– Pre-operative Myer–Cotton subglottic stenosis grading	3	3	3	3
– Ventilation technique	Jet	Jet	Jet	High-flow nasal cannula
– Operative time (minutes)	30	6	50	22
Follow-up duration (months)	36	16	19	8

and constitute a much larger undertaking for the patient and physician, it is generally recommended that endoscopic therapy be pursued initially before attempting open procedures.<sup>5,6</sup> Likewise, many different methods exist for achieving anaesthesia and ventilation during both the endoscopic and open surgical management of patients with subglottic stenosis.<sup>30</sup> Regardless of the method chosen, good communication between an experienced anaesthesiologist and surgeon is of paramount importance.

In the case of a pregnant patient with idiopathic subglottic stenosis, the risks of surgical intervention to both mother and fetus must be carefully considered. Surgery during pregnancy is associated with higher rates of premature delivery, intrauterine growth restriction and low birth weight.<sup>11</sup> Although these complications of pregnancy are most commonly encountered in abdominal surgical procedures,<sup>13</sup> surgery for idiopathic subglottic stenosis is still associated with a considerable risk of hypoxia and related injuries. For this reason, the American College of Obstetricians and Gynaecologists recommends involvement of the patient's obstetrician early in the planning process when considering surgical intervention in pregnancy.<sup>31</sup>

Other practitioners' experiences of treating idiopathic subglottic stenosis in pregnancy emphasise the necessity of carefully choosing the best and least invasive procedure for these patients (Table 2). For example, in the case presented by Scholz *et al.*, the authors noted an improvement in symptoms for the first 2 days post-operatively, with a subsequent deterioration in the patient's condition secondary to post-laser laryngeal oedema.<sup>12</sup> Within a few days post-operatively, the patient developed pre-eclampsia with pulmonary oedema, which necessitated emergent upright caesarean section at 32 weeks of gestational age. Two other patients required tracheostomy for acute airway management: one in order to allow for the administration of labour anaesthesia, and the other for caesarean section.<sup>32</sup> The latter patient was initially intubated with a 4.0 microlaryngeal tube, but extubation subsequently failed in the intensive care unit and a tracheostomy was required.<sup>23,24</sup>

Ratner *et al.* elected to use mask induction for the biopsy of a tracheal web, which proved to be an effective form of induction.<sup>19</sup> However, high ventilator pressures were required to maintain oxygenation. Salama and Body used a method of balloon dilation for acute airway management in their full-term patient to allow for normal delivery.<sup>17</sup> This is similar to the method of the current study, and was accomplished with minimal morbidity.

It is clear from these cases that there is both substantial variability in treatment strategies and potential risk related to

**Table 2.** Patient characteristics and outcomes of previous reports

Study authors	Study year	Age (years)	Gestational age (weeks)	Operation	Anaesthesia	Outcomes
Paré <i>et al.</i> <sup>15</sup>	1982	24	36	Tracheostomy	General	Complex tracheal laceration, caesarean section, decannulation
Mallett <i>et al.</i> <sup>16</sup>	1989	17	35	None; heliox used		NSVD
Salama & Body <sup>17</sup>	1994	31	37	Balloon dilation	Local	
Pauzner <i>et al.</i> <sup>18</sup>	1994	30	38	Laser	NA	NSVD
Sutcliffe <i>et al.</i> <sup>14</sup>	1995	33	34	Intubation	General	Caesarean section; patient died several weeks post-operation secondary to airway obstruction
Ratner <i>et al.</i> <sup>19</sup>	2001	33	32	Rigid dilation	General	Caesarean section with spinal
Kuczkowski <sup>13</sup>	2004	37	23	Awake tracheostomy	Local	Caesarean section
Darjani <i>et al.</i> <sup>20</sup>	2007	29	5 months	Rigid dilation, Nd:YAG laser ablation	General	
Scholz <i>et al.</i> <sup>12</sup>	2008	24	29	Laser-assisted dilation	Total IV anaesthetic	Subsequent delivery via caesarean section with spinal
Rumbak <i>et al.</i> <sup>21</sup>	2010					
– Case 1		20	27	Cautery-assisted bronchoscopic balloon dilation	Local	NSVD
– Case 2		21	32	Cautery-assisted bronchoscopic balloon dilation	General	NSVD
Engel <i>et al.</i> <sup>22</sup>	2011	29	37	None	Spinal	NSVD
Nanjegowda & Srinivasan, <sup>23</sup> Karippacheril <i>et al.</i> <sup>24</sup>	2011	28	40	None	General	Caesarean section, tracheostomy after delivery, decannulation
Nash <i>et al.</i> <sup>25</sup>	2014	30	24	None	Epidural	NSVD
Carness & Berman <sup>26</sup>	2014	31	28	KTP laser assisted bronchoscopic balloon dilation	Local	NSVD

NSVD = normal spontaneous vaginal delivery; NA = not available; Nd:YAG = neodymium-doped yttrium aluminium garnet; IV = intravenous; KTP = potassium titanyl phosphate

airway management in the pregnant idiopathic subglottic stenosis patient. One investigator may have induced pre-eclampsia. Some patients were subjected to tracheostomy. Other patients required very high ventilation pressures in order to maintain oxygenation, which could have caused a pneumothorax. For this reason, airway management should be carefully considered and be clear to all providers involved in the care of these patients.

- Idiopathic subglottic stenosis is a rare but potentially serious condition, most common in younger females
- Idiopathic subglottic stenosis may occur, and/or require management, during pregnancy
- There is no consensus regarding optimal management of these patients or ventilation provision during airway surgery
- The technique of balloon dilation, with supraglottic jet or high-flow nasal cannula ventilation, is presented
- Compared to other methods, the technique is safe and effective
- All patients had lasting symptom resolution and normal spontaneous vaginal delivery at term

We have found total intravenous anaesthesia with paralysis, coupled with a non-invasive mode of oxygenation, to be a

successful strategy in this patient population. Two previous studies by Salama and Body,<sup>17</sup> and Lee *et al.*,<sup>8</sup> have demonstrated the efficacy of endoscopic techniques and general anaesthesia, consistent with the findings of the current study. Optiflow has previously been demonstrated to be a safe method of ventilation in the operative management of airway stenosis, and was effective in one case in this study.<sup>33</sup>

## Conclusion

When treating a patient with idiopathic subglottic stenosis, there are many options and variations. However, treatment options are more limited in the pregnant patient given the risks of surgery to mother and fetus. Previous studies have illustrated the necessity of being judicious in one's pursuit of therapy for idiopathic subglottic stenosis. It has been our experience with these four cases that balloon dilation, coupled with supraglottic jet ventilation or humidified high-flow oxygenation via nasal cannula ventilation, is safe, efficacious, and should be the primary mode of treatment for this patient population.

## Competing interests

None declared



## References

- 1 Damrose EJ. On the development of idiopathic subglottic stenosis. *Med Hypotheses* 2008;**71**:122–5
- 2 Mark EJ, Meng F, Kradin RL, Mathisen DJ, Matsubara O. Idiopathic tracheal stenosis: a clinicopathologic study of 63 cases and comparison of the pathology with chondromalacia. *Am J Surg Pathol* 2008;**32**:1138–43
- 3 Poetker DM, Ettema SL, Blumin JH, Toohill RJ, Merati AL. Association of airway abnormalities and risk factors in 37 subglottic stenosis patients. *Otolaryngol Head Neck Surg* 2006;**135**:434–7
- 4 Valdez TA, Shapshay SM. Idiopathic subglottic stenosis revisited. *Ann Otol Rhinol Laryngol* 2002;**111**:690–5
- 5 George M, Lang F, Pasche P, Monnier P. Surgical management of laryngotracheal stenosis in adults. *Eur Arch Otorhinolaryngol* 2005;**262**:609–15
- 6 Giudice M, Piazza C, Foccoli P, Toninelli C, Cavaliere S, Peretti G. Idiopathic subglottic stenosis: management by endoscopic and open-neck surgery in a series of 30 patients. *Eur Arch Otorhinolaryngol* 2003;**260**:235–8
- 7 Krinsky WS, Rodrigues MP, Malayaman N, Sarkar S. Spray cryotherapy for the treatment of glottic and subglottic stenosis. *Laryngoscope* 2010;**120**:473–7
- 8 Lee K-H, Ko G-Y, Song H-Y, Shim TS, Kim WS. Benign tracheobronchial stenoses: long-term clinical experience with balloon dilation. *J Vasc Interv Radiol* 2002;**13**:909–14
- 9 Roediger FC, Orloff LA, Courey MS. Adult subglottic stenosis: management with laser incisions and mitomycin-C. *Laryngoscope* 2008;**118**:1542–6
- 10 Smith ME, Elstad M. Mitomycin C and the endoscopic treatment of laryngotracheal stenosis: are two applications better than one? *Laryngoscope* 2009;**119**:272–83
- 11 Mazze RI, Kallén B. Reproductive outcome after anesthesia and operation during pregnancy: a registry study of 5405 cases. *Am J Obstet Gynecol* 1989;**161**:1178–85
- 12 Scholz A, Srinivas K, Stacey M, Clyburn P. Subglottic stenosis in pregnancy. *Br J Anaesth* 2008;**100**:385–8
- 13 Kuczkowski KM. Nonobstetric surgery during pregnancy: what are the risks of anesthesia? *Obstet Gynecol Surv* 2004;**59**:52–6
- 14 Sutcliffe N, Remington SA, Ramsay TM, Mason C. Severe tracheal stenosis and operative delivery. *Anaesthesia* 1995;**50**:26–9
- 15 Paré P, Donevan R, Nelems J. Clues to unrecognized upper airway obstruction. *Can Med Assoc J* 1982;**127**:39–41
- 16 Mallett V, Bhatia R, Kissner D, Sokol R. Use of an HeO<sub>2</sub> mixture in the management of upper airway obstruction during labor and delivery. *A case report. J Reprod Med* 1989;**34**:429–30
- 17 Salama D, Body S. Management of a term parturient with tracheal stenosis. *Br J Anaesth* 1994;**72**:354–7
- 18 Pazuener R, Mayan H, Hershko E, Alcalay M, Farfel Z. Exacerbation of Wegener's granulomatosis during pregnancy: report of a case with tracheal stenosis and literature review. *J Rheumatol* 1994;**21**:1153–6
- 19 Ratner EF, Cohen SE, El Sayed Y, Druzin M. Mask induction with sevoflurane in a parturient with severe tracheal stenosis. *Anesthesiology* 2001;**95**:553–5
- 20 Darjani HRJ, Parsa T, Pirzeh A, Heydarnazhad H. Idiopathic subglottic stenosis in a pregnant woman: successful treatment with dilatation and Nd: YAG laser ablation. *Tanaffos* 2007;**6**:58–62
- 21 Rumbak M, Dryer J, Padhya T, Camporesi E, Karlinski R, Mangar D. Successful management of subglottic stenosis during the third trimester of pregnancy. *J Bronchology Interv Pulmonol* 2010;**17**:342–4
- 22 Engel NM, Gramke HF, Peeters L, Marcus MA. Combined spinal-epidural anaesthesia for a woman with Wegener's granulomatosis with subglottic stenosis. *Int J Obstet Anesth* 2011;**20**:94–5
- 23 Nanjagowda N, Srinivasan N. Undiagnosed tracheal stenosis complicating airway management in pregnancy. *Sri Lankan J Anaesthesiol* 2011;**19**:86–8
- 24 Karippacheril JG, Goneppanavar U, Prabhu M, Revappa KB. Idiopathic subglottic stenosis in pregnancy: a deceptive laryngoscopic view. *Indian J Anaesth* 2011;**55**:521–3
- 25 Nash Z, Krishna A, Darwish M, Mascarenhas L. Conservative management of subglottic stenosis in pregnancy resulting in vaginal birth. *BMJ Case Rep* 2014;**2014**:bcr2013202137
- 26 Carness JM, Berman JL. Awake microlaryngoscopy and serial balloon dilation in a third trimester multigravida with subglottic tracheal stenosis: anesthetic implications. *A A Case Rep* 2014;**3**:166–8
- 27 Cunningham FG. *Williams Obstetrics*. New York: McGraw-Hill, 2010
- 28 Modgil G, Havas T, Mellis C. Idiopathic subglottic stenosis and the relationship to menses in a 12-year-old girl. *J Paediatr Child Health* 2005;**41**:374–6
- 29 Ananth CV, Vintzileos AM. Medically indicated preterm birth: recognizing the importance of the problem. *Clin Perinatol* 2008;**35**:53–67
- 30 Daumerie G, Su S, Ochroch EA. Anesthesia for the patient with tracheal stenosis. *Anesth Clin* 2010;**28**:157–74
- 31 ACOG Committee on Obstetric Practice. ACOG Committee Opinion No. 474: nonobstetric surgery during pregnancy. *Obstet Gynecol* 2011;**117**(2 Pt 1):420–1
- 32 Kuczkowski K, Benumof J. Subglottic tracheal stenosis in pregnancy: anaesthetic implications. *Anaesth Intensive Care* 2003;**31**:576–7
- 33 Tam K, Jeffery C, Sung CK. Surgical management of supraglottic stenosis using intubationless Optiflow. *Ann Otol Rhinol Laryngol* 2017;**126**:669–72