

# Proximal airway obstruction caused by adenoid cystic carcinoma in a pregnant woman: A case report

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Respiratory tumours are very rare during pregnancy. Here, we report a case of a primigravida woman diagnosed at 27 weeks' gestation with stridor and dyspnoea, and a primary tracheal tumour. To the best of our knowledge, this is a 4th case report of tracheal adenoid cystic carcinoma during pregnancy. Our staged management of airway obstruction during pregnancy and definite treatment are discussed.

**Keywords.** tracheal tumor; airway obstruction; pregnancy; tracheal surgery; tracheostomy; cryoablation.

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Primary tracheal tumours have an annual incidence of about 0.1 in 100 000 population.<sup>[1]</sup> Adenoid cystic carcinoma (ACC) is the second most common type of tracheal tumour after squamous cell carcinoma.<sup>[1]</sup> Tracheal tumours, especially in their early phase, are usually misdiagnosed as asthma due to similar presentations.<sup>[2]</sup> Primary tracheal tumours are very rare during pregnancy. Nearly 1 in 2 000 pregnant women have tumours in sites other than the airway, i.e. breast, cervical, skin (melanoma), ovarian, and haematological malignancies.<sup>[3,4]</sup> This is a report of the airway management of a young primiparous woman with tracheal ACC and airway obstruction during pregnancy who underwent definitive surgery after delivery.

## Case

A 30-year-old primigravid woman was referred at 27 weeks' gestation in July 2020 with a history of a gradual onset of shortness of breath, coughs, and recent stridor with a background of asthma

diagnosed in the past year. Stroboscopy showed a subglottic tumour and computed tomography (CT) scan confirmed a tumour in the proximal trachea causing more than 95% obstruction of the lumen with extraluminal extension (Figs 1A, 1B and 1C). She was intubated in the operating room by means of a fibre-optic guided small orotracheal tube, and immediate tracheostomy was done only distal to the tumour to maintain a secure open airway. In the same session, a biopsy was taken from the tumour and partial debulking was done using cryoablation. She was discharged with a fenestrated tracheostomy. Subsequently, partial cryoablation was done twice with an interval of two weeks (Fig. 2A). The pathological analysis showed low-grade ACC with a dominant cribriform pattern (Figs 2E and 2F). Two weeks after caesarean section delivery, resection of the proximal trachea (ring 1 - 5) and laryngotracheal anastomosis were done. The proximal posterior margin at the level of the post cricoid cartilage was very close to the tumour (Figs 2B, 2C and 2D).

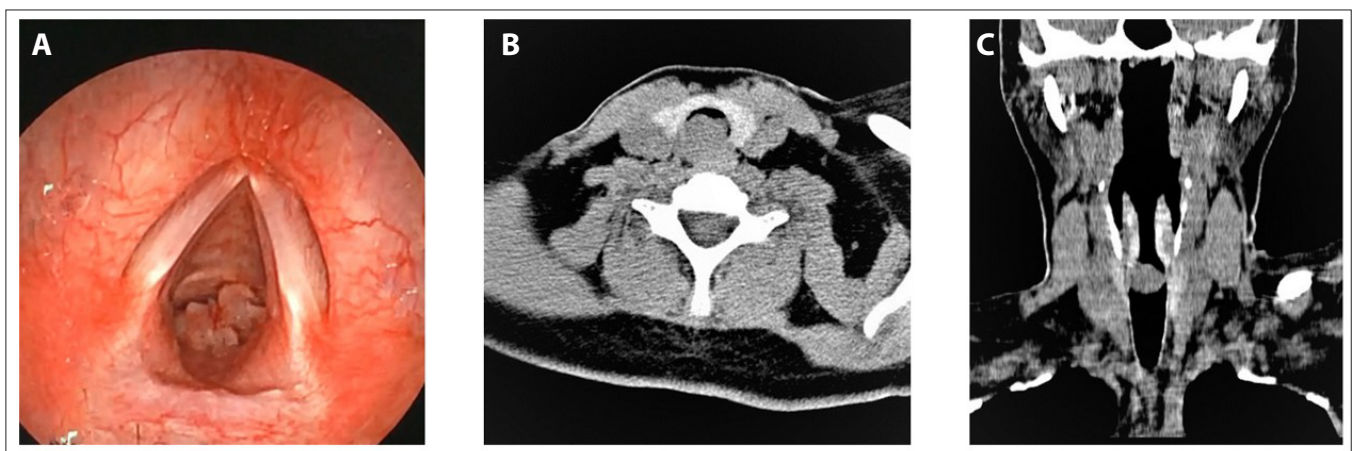


Fig. 1. Diagnostic workup. (A) Stroboscopy photo of the proximal cauliflower tracheal tumour. (B) Axial computed tomography scan of the proximal tracheal tumour. (C) Coronal computed tomography scan of the proximal tracheal tumour.

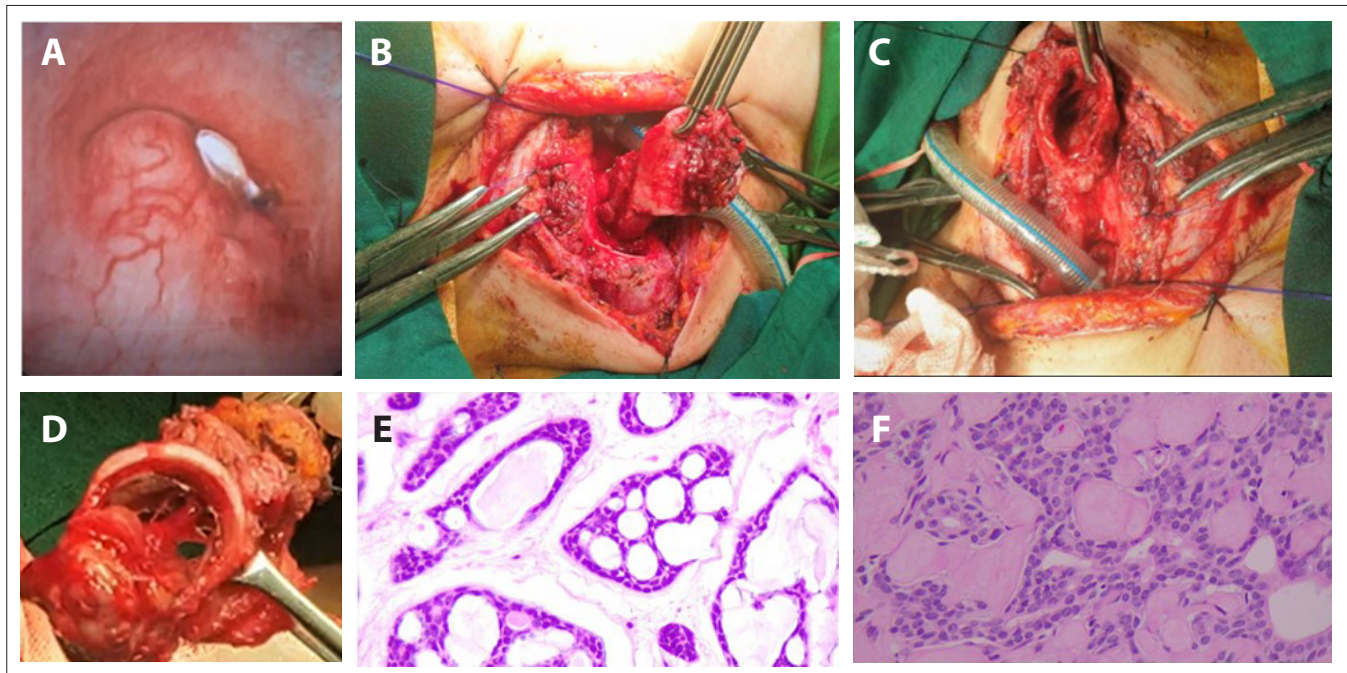


Fig. 2. Cryoablation, intraoperative image, and histology. (A) Proximal tracheal tumour after 3 times cryoablation with a tracheostomy tube in place. (B) Intraoperative image with proximal margin. (C) Intraoperative image with distal margin. (D) Intraoperative image of sleeve tracheal resection. (E) Cribriform and tubular pattern (H&E, 40 $\times$  magnification). (F) Dual population of ductal and myoepithelial cells associated with myxoid globules (40 $\times$  magnification).

Pathological examination showed microscopic involvement of the proximal margin. She received 60 Gray radiation doses in 30 sessions using the 3D conformal technique 6 weeks postoperatively. Re-bronchoscopy done early (Figs 3A and B) and 6 months (Figs 3C and 3D) post radiotherapy showed a normal healing trachea. She is planned to be clinically evaluated every 3 months in the first 2 years, followed by lifelong annual clinical evaluation, CT scan and if required, bronchoscopy. This study was reviewed and approved by the Ethics Review Board of Imam Khomeini Hospital affiliated with Tehran University of Medical Science (ref. no. IR.TUMS.IKHC.REC.1400.20). Informed consent was obtained from the patient.

## Discussion

Malignant tracheal tumour comprises only 0.2% (0.1 - 0.4%) of all respiratory tract malignancies, with an annual incidence of about 0.1 in 100 000 population, accounting for only 0.02 - 0.04% of all reported malignancies.<sup>[1]</sup> The median age is >50 years, indicating its rarity in young patients.<sup>[5]</sup> Our patient, a 30-year-old primiparous woman, was misdiagnosed with mild asthma before referral to our institution. Her symptoms

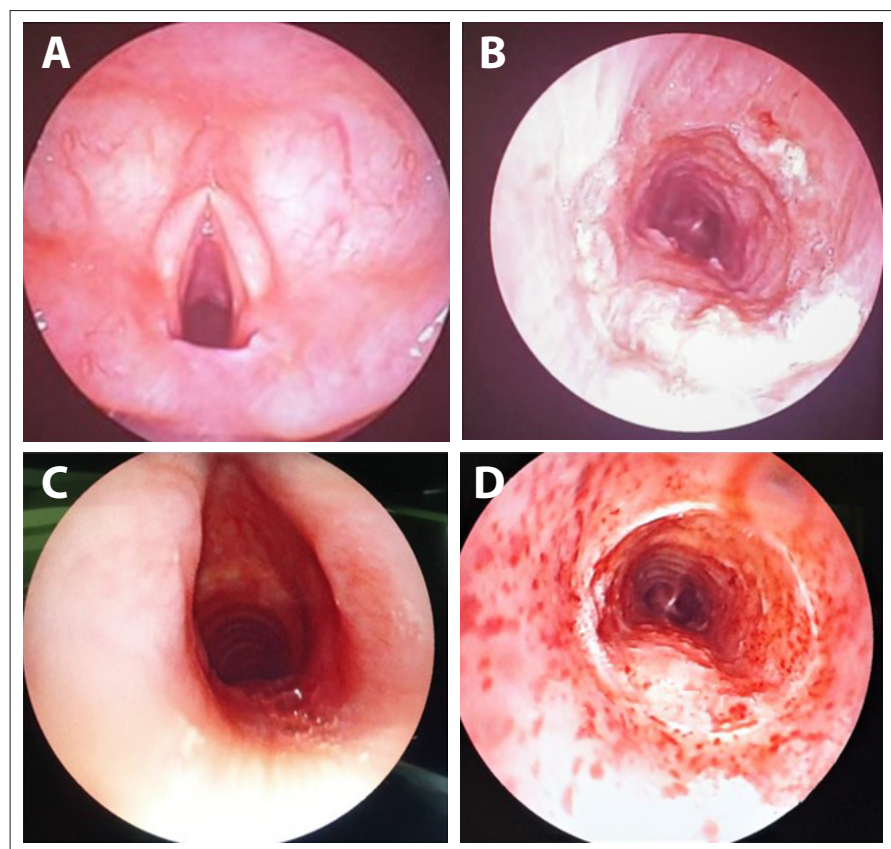


Fig. 3. Postoperative bronchoscopy. (A and B) Early post-operation bronchoscopy. (C and D) Six months post operation and radiotherapy.

were attributed to asthma and normal physiological changes of pregnancy until marked symptoms developed in the 3rd trimester.<sup>[4]</sup> Although a diagnosis of upper airway obstruction by a tumour is not difficult in a patient with obvious symptoms of dyspnoea and stridor, tumours such as ACC that have a slow-growing nature and precisely do not cause haemoptysis, may be frequently missed at the early stages.<sup>[4,5]</sup> The physiological changes of pregnancy and limitation of investigations for prevention of hazards to the mother and fetus are important explanations for misdiagnosis or delayed diagnosis.<sup>[4]</sup> Physicians and obstetricians should be aware of such a rare tumour of the upper airway in a pregnant woman with respiratory complaints and symptoms.<sup>[4]</sup>

Shafiee *et al.*<sup>[4]</sup> reported a case of ACC during pregnancy. They reviewed the literature and found 13 cases of tracheal tumours in pregnant women. Five cases were primary malignant tracheal tumours and only two were ACCs.<sup>[4]</sup>

Considering the literature research conducted by Shafiee *et al.*,<sup>[4]</sup> our patient is the 4th case of primary ACC in pregnancy. Diagnosis of tracheal tumour was established with stroboscopy and CT scan before referral to our institution. At the outset, we maintained a secure upper airway by fibre-optic guided intubation and subsequent tracheostomy. Adequate biopsy was taken by partial cryoablation. Total endobronchial cryoablation was not indicated because of extraluminal extension. Partial cryoablation was repeated twice more to have partial patency of the airway and preserve phonation until definitive surgical resection. At this time, the surface of the tumour was covered by smooth scar tissue rather than a cauliflower appearance (Figs 1A and 2A), which helped to maintain partial patency of the airway and preserve phonation.

The tumour originated from the posterior tracheal wall, very close to the cricoid cartilage. The selected proximal margin was grossly normal, and we intended to preserve the larynx in this young patient, and we know she had an extraluminal extension. In this scenario, the frozen section does not change the plan of resection; therefore, to save time during operation, it was nullified. Microscopic involvement is

accepted and it is not unusual in ACC.<sup>[4,6]</sup> She received postoperative radiotherapy for microscopic proximal involvement and extraluminal extension.<sup>[4,6]</sup> Despite the heterogeneity and different follow-up times for such a rare tumour, she is planned to receive lifelong follow-up.<sup>[6]</sup>

## Conclusion

Primary tracheal tumours are very rare in pregnant women. It is often missed, although it can be diagnosed by unusual symptoms in pregnancy with high suspicion. Staged or definite treatment can be done safely by individualised case and tumour histology.

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**Conflicts of interest.** None.

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