Bifid Epiglottis: What Perioperative Physician Should Know about It?

The Editor,

A 7-year-old male child visited preoperative clinic for preanesthetic evaluation. He was posted for urological procedure. On history, he was first child of normal parents. On examination, he was 35 kg in weight and having course facial features. At birth, he was diagnosed to have high anorectal malformations (ARMs), penoscrotal hypospadias, polydactyly, bilateral congenital talipes equinovarus, and left renal agenesis. Child's mental progress was also not aged appropriate. He has undergone surgery for high ARM at birth uneventfully. His clinical examination and laboratory investigations were within normal limits. His chest radiograph, 12 lead electrocardiography and two-dimensional echocardiography were also normal. Hormonal assay showed a testosterone level of 1.6 nmol/L (normal 9–35 nmol/L for males). No chromosomal abnormality was diagnosed on karyotyping. Airway examination revealed a mouth opening of 2 cm with large tongue. Previous anesthesia records were not available. Routine anesthesia induction was done with sevoflurane in oxygen, 70 μ fentanyl, and 20 mg atracurium. On direct laryngoscopy for tracheal tube advancement, a bifid epiglottis with complete slit up to base of epiglottis [Figure 1] was seen. Backward, upward, and rightward pressure was applied by assistant *in lieu* of high placed glottis and a size 5 ID tracheal tube was advanced into trachea with slight difficulty. Anesthesia was maintained on sevoflurane (inhaled concentration 1.5%–2%) in oxygen and air (40:60). After 4 h at the end of surgery, moderate inspiratory stridor was present after tracheal extubation. He was kept in lateral position in postanesthesia care unit and supplemental oxygen was given by venturi mask (FiO_2 0.5). His recovery was uneventful and shifted to ward on evening.

Congenital anomalies of the epiglottis are rare.[1] True bifid epiglottis is defined as cleft in the epiglottis that extends at least two third of its length.[2] It is present mainly as part of one of the syndrome but in extreme rare condition, present as an isolated anomaly. Till date, around 27 cases of bifid epiglottis and 1 case of trifid epiglottis[3] have been reported. These may be asymptomatic or can present with life-threatening severe inspiratory stridor and airway obstruction, sometimes may require tracheostomy. Our case is a true bifid epiglottis (defined as a cleft extending at least 2/3 of the length of epiglottis toward its base). The bifid epiglottis is associated with complex malformations and almost all reported cases were presented with respiratory stridor. As a part of the syndrome, it may be present in Pallister-Hall syndrome (an autosomal dominant syndrome, characterized by bifid epiglottis, hypothalamic hamartoblastoma, hypopituitarism, imperforate anus, polydactyly, and micrognathia), Bardet–Biedl syndrome (bifid epiglottis, retinitis pigmentosa, mental growth problem, hypogonadism, renal and anal abnormalities, polydactyly, micropenis, and hypospadiasis), and Joubert syndrome (autosomal recessive disorder characterized by bifid epiglottis, polydactyly, hypotonia, ataxia, abnormal eye movements and a respiratory pattern of alternating tachypnea-apnea, hypotonia, renal abnormalities, and facial dysmorphism).[3] Bifid epiglottis may also present as familial disorder involving structural congenital cardiac anomalies such as atrial septal defect, ventricular septal defect, patent ductus arteriosus, hypoplastic left heart, and hypoplastic aorta and these patients may be posted for cardiac surgical procedure any time after birth.[4] Bifid epiglottis may present with or without structural abnormalities having some airway symptoms[3,5] and even requiring microsurgical excision to relieve the stridor.[5] In some cases, bifid epiglottis was associated with laryngeal cyst and cricoid stenosis.[6] A careful laryngoscopy and tracheal intubation and extubation is required during general anesthesia. The child developed stridor in immediate postexubation period that was managed conservatively. Each leaflet (halve) of bifid epiglottis becomes lax and may prolapse into laryngeal inlet and can obstruct the airflow. It can also mimic laryngomalacia and present as airway emergency. The stridor present in our case and other reported cases can be explained by this pathophysiology.

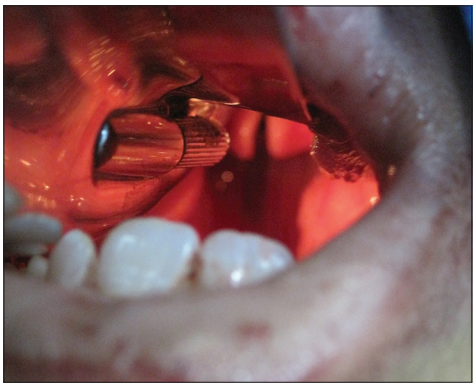

Figure 1: A bifid epiglottis on direct laryngoscopy with a complete slit up to the base of epiglottis

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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Quick Response Code:

Website: www.annals.in

DOI: 10.4103/aca.ACA_84_17

How to cite this article: Yadav R, Solanki SL, Doctor JR. Bifid epiglottis: What perioperative physician should know about it? Ann Card Anaesth 2017;20:479-80.