Anesthetic management of an adolescent with congenital glossopharyngeal web

Sir,

Glossopharyngeal web is a rare congenital anomaly, which extends from the posterior part of the tongue to the posterior pharyngeal wall. It represents the embryological remnant of the buccopharyngeal membrane (BPM) and causes an anesthetic challenge due to the difficult airway. Here, we report the airway management of an adolescent with the partial glossopharyngeal web.

A 15-year-old, 50 kg adolescent male presented with a history of difficulty in talking and swallowing for the past 10 years. He was evaluated and found to have a glossopharyngeal web, which extended from the base of the tongue on the right side to the posterior pharyngeal wall [Figure 1]. He was operated 9 years before elsewhere for coarctation of the aorta. The previous anesthesia and surgery were uneventful according to the patient’s mother. Preoperative echocardiography revealed a systolic pressure gradient of 34 mmHg across the coarctation segment, normal left ventricular systolic function, and ejection fraction of 58.7%. There was no other significant medical history.

He was scheduled for the release of the glossopharyngeal band. He was adequately fasting and premedicated. In the operating room, a 20 gauge cannula was inserted into a peripheral upper limb vein. Monitors including electrocardiography, noninvasive blood pressure, and pulse oximeter were connected. Baseline blood pressure readings were taken from all four limbs with appropriate sized cuffs.

Figure 1: Preoperative photograph of the patient showing the presence of glossopharyngeal band on the right side (permission obtained from the patient)

There was no significant difference. Since difficult airway was anticipated, difficult intubation cart with fibreoptic bronchoscope was kept at hand.

Following preoxygenation, patient was induced with fentanyl 2 mcg/kg and propofol 2 mg/kg. After ensuring ability to mask ventilate, he was paralyzed with vecuronium. After paralysis patient was able to be ventilated easily without any airway adjuvants. Laryngoscopy using Macintosh 3 left sided blade revealed a distorted anatomy with the epiglottis being pulled toward the band (Cormack and Lehane laryngoscopy grade 4). The view did not improve with BURP and the working space on the right side was too narrow, a decision to intubate using a fibreoptic bronchoscope was made. Patient was

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Intraoperatively, anesthesia was maintained with intermittent positive pressure ventilation using air/oxygen/isoflurane. Adequate analgesia was provided with intravenous morphine and paracetamol as well as infiltration of local anesthetic by surgeons. Intravenous dexamethasone was administered soon after intubation for its anti-edema effect. He was hemodynamically stable during the procedure.

At the end of the procedure, a direct laryngoscopy using Macintosh 3 blade revealed a grade 2 view. There was minimal edema around the base of the tongue on the right side. Neuromuscular blockade was adequately reversed using neostigmine and glycopyrrolate and the patient was extubated awake in the lateral position. Immediate postoperative period in the postanesthetic care unit was eventful, and he was transferred to the ward in a stable condition.

Consent for his case to be reported was obtained from the patient before writing the article.

Congenital persistent BPM is an exceedingly rare anomaly.[1-3] The BPM (oropharyngeal) is a barrier between the stomodeum (primitive mouth) and the early pharynx during embryonic development.[3] This membrane usually breaks down by the 4th week, opening the oral cavity to the pharynx. Failure of its resorption will result in persistent BPM.[4-6]

There are three presentations of the persistent BPM namely:

1. Complete membrane: No communication between the mouth and the pharynx, the patient may suffer from early respiratory compromise.[9] Direct laryngoscopy may be unsuccessful and emergency tracheostomy needed for securing the airway.[10]
2. Partial or total BPM: Patients were able to breathe easily and present with dysphagia between 1 and 3 months of age. These patients may need tracheostomy and surgery on the BPM.
3. BPM as an incidental finding: Partial BPM presents with a partial connection between the oral cavity and the pharynx permitting normal respiratory and swallowing functions.

Table 1: Reported cases and congenital anomalies associated with a persistent buccopharyngeal membrane[5-8]

| Author  | Year | Sex | Age | Membrane description | Associated congenital anomalies |
|---------|------|-----|-----|----------------------|--------------------------------|
| Verma   | 2009 | F   | 0   | Total membrane       | Transposition of great arteries, patent ductus arteriosus, atrial septal defect |
| Lee     | 2007 | F   | 0   | Silt perforation     | Ventricular septal defect, patent ductus arteriosus, vertebral anomalies |
| Kara    | 2007 | F   | 18  | 2 cm central perf    | None                           |
| Takahashi | 2007 | M   | 0   | Total membrane       | Atrial septal defect           |
| Tan     | 2006 | M   | 0   | Total membrane       | None                           |
| Ooi     | 2005 | M   | 27  | 2 cm central perf    | Atrial septal defect, unilaterial choanal atresia |
| Lim     | 2005 | M   | 1 day | Total membrane     | Atrial septal defect, unilateral choanal atresia |
| Thauvin | 2002 | F   | 0   | Total membrane       | Hypomandibular faciocranial dysostosis |
| Kangaputra | 2003 | F   | 0   | Total membrane       | Aglossia, micrognathia, microcephaly, absence of mandibular teeth |
| Bent    | 1997 | F   | 0   | 5 mm perforation     | Transposition of great arteries |
| Agarwal | 1996 | M   | 8 days | Total membrane     | None                           |
| Agarwal | 1996 | M   | 13 days | Total membrane     | None                           |
| Ludman  | 1993 | F   | 0   | Total membrane       | Hypomandibular faciocranial dysostosis, bicornuate uterus |
| Gartlan | 1992 | M   | 0   | Pinpoint hole        | Micrognathia, microglossia     |
| Schimke | 1991 | F   | 0   | Total membrane       | Hypomandibular faciocranial dysostosis, patent ductus arteriosus |
| Flannery| 1989 | F   | 0   | Total membrane       | Mandibulofacial dysostosis, downsloping palpebral fissures, epicantthal folds, broad nasal bridge, costovertebral anomalies |
| Neidich | 1988 | F   | 0   | Total membrane       | Severe first branchial arch anomalies, atrial septal defect |
| Arcand  | 1988 | M   | 3 yrs | Partial membrane    | None                           |
| Hoffman | 1979 | M   | 30 days | Partial membrane    | None                           |
| Chandra | 1974 | F   | 28 days | Total membrane    | None                           |
| Seghers | 1966 | M   | 30 days | Total membrane    | Auricular and vertebral anomalies |
| Longacre| 1951 | F   | 90 days | Partial membrane   | None                           |
| Fridenberg | 1908 | M   | 21 yrs | Partial membrane   | None                           |
In our case, the patient had a partial persistent BPM where the glossopharyngeal band on the right side extended from the posterior part of the tongue to the right posterior pharyngeal wall.

Patients with persistent BPM can present at any age, and the symptom also varies depending on the degree of the embryological remnant. These patients can pose anesthetic problems like airway difficulty\[10,11\]. Airway management includes emergency tracheostomy for respiratory distress\[10\] or tracheostomy for securing the airway during surgery\[11\]. Pirat et al. have described retrograde intubation for a child even after the excision of the glossopharyngeal web.\[11\] Some patients, although asymptomatic may need fiberoptic intubation.

Moreover, these patients with persistent BPM can have other associated congenital anomalies [Table 1] including congenital cardiac defects. In our case, the patient had coarctation of the aorta, which had been operated before the presentation of the glossopharyngeal web.

To conclude, patients with such rare anomalies could be managed better by a detailed preoperative workup, the anticipation of intraoperative difficulties, and a thorough search of sources available.

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Conflicts of interest
There are no conflicts of interest.

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