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Case Report

The Anesthetic Implications of Goldenhar Syndrome: A Case Report

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ABSTRACT

Goldenhar syndrome or oculo-auriculo-vertebral spectrum is associated with multiple craniofacial, ocular, auricular, and vertebral anomalies in different combinations. These defects often lead to significant anesthetic challenges, frequently involving airway management, cervical spine instability, and cardiac defects. We present the case of a 14-year-old adolescent female diagnosed with Goldenhar syndrome, exhibiting left-sided auricular appendage, limbal dermoid in the left eye, and hemifacial atrophy with facial asymmetry. She had an excision of the auricular appendage from the preauricular area under general anesthesia. The perioperative anesthetic considerations, challenges, and successful management strategies in such patients are discussed.

Key words: Adolescent, anesthesia, Goldenhar syndrome

INTRODUCTION

Maurice Goldenhar first described Goldenhar syndrome (GS) in 1952. [1] GS falls within the oculo-auriculo-vertebral spectrum, with an estimated incidence of 1 in 3500 to 1 in 7000 live births. [1] Unilateral craniofacial anomalies, including hemifacial microsomia, microtia, preauricular tags, ocular dermoid, and vertebral defects, are commonly described components of GS. [2] While on the one hand, mandibular hypoplasia and facial asymmetry are often associated with complicated airway management, associated anomalies like congenital heart disease and vertebral anomalies potentiate the perioperative risks on the other hand. [3, 4] The case reported here highlights the anesthetic challenges in managing a patient with GS undergoing auricular surgery.

CASE REPORT

A 14-year-old female with a weight of 42 kg and a height of 156 cm was diagnosed with GS. Her clinical features included an auricular appendage on the left side, left-sided hemifacial atrophy with mandibular hypoplasia, and a previously diagnosed left limbal dermoid (**Figure 1**). There was no history of prior anesthetic exposure. She had been worked up and investigated in the outdoor clinic. With a plan for excision of the auricular appendage and reconstruction thereafter, she was referred to the pre-anesthetic clinic.

During pre-anesthetic evaluation, facial asymmetry and left hemifacial hypoplasia were evident (**Figure 1**). Her Mallampati classification was III; mouth opening was 3 cm, with

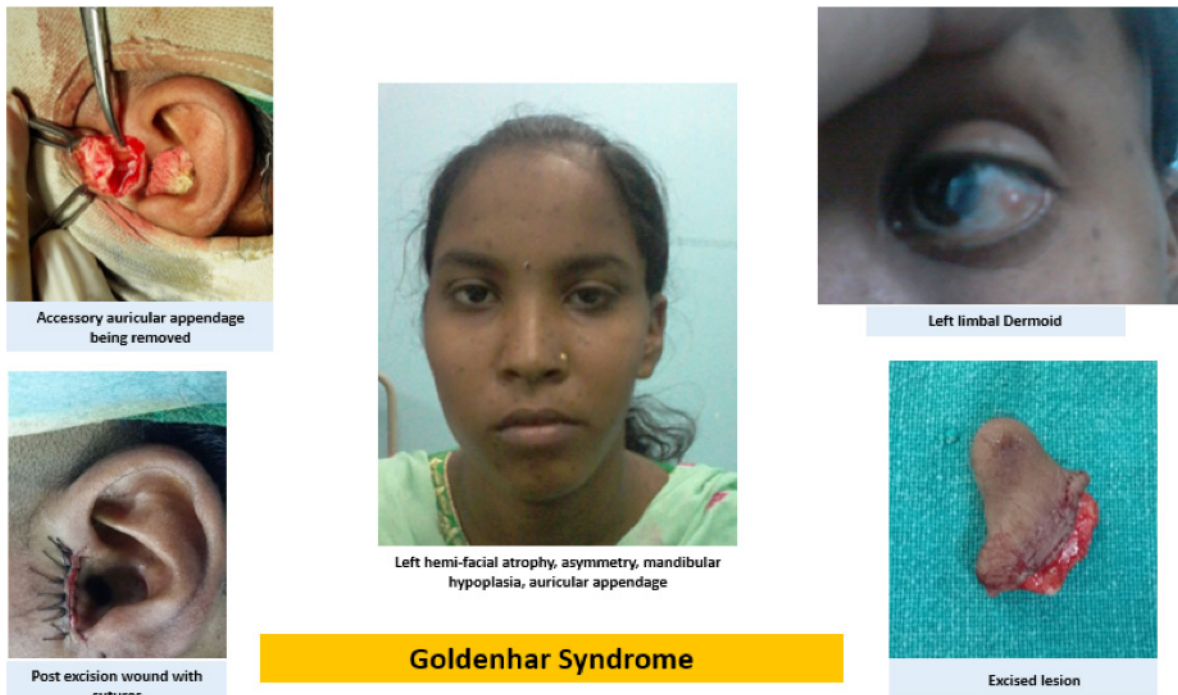


Figure 1: Clinical and intraoperative pictures of the patient.

some restriction in the protrusion of the mandible. Anticipating a difficult airway, the thyromental and sternomental distances were also measured with values of 6 cm and 13 cm, respectively. These distances were measured with a rigid scale with the neck extended and the mouth closed. These values fell in the borderline region as per the normal reported values. She also had misaligned teeth. Although there was no restriction in the neck movements, an X-ray of the cervical spine was performed due to the known association with vertebral anomalies; results revealed no fusion or instability. A baseline echocardiogram showed normal cardiac anatomy and function, ruling out any associated cardiac anomaly. There were no respiratory or renal anomalies.

Routine investigations, including complete blood counts, renal and liver function tests, were within normal limits. Difficult airway equipment, including video laryngoscopes, fibreoptic bronchoscopes, supraglottic airway devices, and an emergency tracheostomy kit, was all kept ready. Informed consent was taken; the possibilities of a difficult airway and tracheostomy were explained to the attendants.

Before induction, standard American Society of Anesthesiologists monitoring (electrocardiogram, pulse oximetry, non-invasive blood pressure, capnography, skin temperature probes) was applied. Intravenous access was secured with an 18-G cannula, and glycopyrrolate (0.2 mg) was administered intravenously. Preoxygenation was done for 3 minutes. Fentanyl (2 µg/kg) and propofol (2 mg/kg) were used for the induction of anesthesia. Facial asymmetry and lack of a prominent chin made it difficult to achieve a tight seal during mask ventilation; posterior displacement of the tongue and restricted mobility of the jaw added to these problems, and all these led to difficulty in mask ventilation. The introduction of an oropharyngeal airway and the use of the two-handed mask ventilation technique helped to optimize

mask ventilation. Also, the remnant gap between the mask and the face had to be occluded with Vaseline-soaked gauze by another assistant to achieve a seal.

For neuromuscular blockade, rocuronium (1 mg/kg) was instituted. This was higher than the normal recommended dose but was used due to a difficult airway and the need to rapidly intubate the patient, considering difficulties in mask ventilation. Video-laryngoscopy using a Macintosh blade type provided a Cormack-Lehane grade II view (with only partial view of the glottis), enabling successful intubation with a 6.5 mm cuffed endotracheal tube. For maintenance of anesthesia, sevoflurane (1.5%–2%) in an oxygen-air mixture was used; intermittent boluses of rocuronium ensured muscle relaxation. Intravenous paracetamol (15 mg/kg) and fentanyl supplementation were used for analgesia.

The intraoperative period was uneventful. The surgical procedure lasted for 45 minutes with minimal blood loss. Reversal of neuromuscular blockade was done using neostigmine (2.5 mg) and glycopyrrolate (0.4 mg). When fully awake with adequate spontaneous respiration and positive reflexes, the patient was extubated. The postoperative period was uneventful, with no respiratory or hemodynamic complications.

DISCUSSION

Patients with GS have multiple anesthetic concerns, which include difficulties in airway management, the presence of cervical vertebral anomalies, cardiac anomalies, mask ventilation difficulties, and potential airway risks in the immediate postoperative period (**Figure 2**). All these make the anesthetic procedure challenging in such cases.

Airway management is often difficult in patients with GS due to mandibular hypoplasia, facial asymmetry, limited mouth



Figure 2: Perioperative challenges in a case of Goldenhar syndrome.

opening, and higher Mallampati grades. [5] In this patient, left hemifacial atrophy and mandibular underdevelopment predicted a possibility of difficult laryngoscopy, necessitating preoperative preparation with advanced airway assisting devices. In our patient, mask ventilation difficulties arose due to facial asymmetry, mandibular hypoplasia, and recessed chin; these can sometimes be compounded by limited cervical spine mobility, and often tracheal intubation can also be difficult in these cases. A pre-defined, step-wise airway management is therefore necessary. Adjuncts such as oropharyngeal airways and two-handed techniques were needed in our case for effective mask ventilation. Preoxygenation and readiness for failed intubation scenarios were crucial. Anticipating a difficult airway, the successful use of video laryngoscopy is consistent with evidence in the literature suggesting its advantage over direct laryngoscopy in such cases. This can be attributed to improved visualization of glottic structures. [6] Although fiberoptic bronchoscopy is the gold standard for anticipated difficult airways, in our case, video-laryngoscopy with Macintosh blades sufficed. Had it been difficult with video-laryngoscopy, we would have used the fiberoptic bronchoscope in the next step.

Before taking up this patient for Anesthesia, we also had our backup plans (in case our routine airway management failed). This included the use of Supraglottic airway devices, which can serve as an effective rescue tool for ventilation and may also act as conduits for fiberoptic-guided intubation. A video laryngoscope was also arranged for better glottic visualization. In case of failure of these techniques, emergency

plans for cricothyrotomy or tracheostomy were also discussed beforehand with the otorhinolaryngologist. A plan for smooth, delayed, and monitored extubation was also made, given the risks of airway edema in the post-operative period and difficulty with re-intubation. In this way, meticulous planning, arrangement of multiple airway devices, and action through this pre-planned difficult airway algorithm helped us in the airway management of this patient with GS.

Cervical spine anomalies, including vertebral fusion, scoliosis, or instability, are common in GS. [7] Although no abnormalities were detected in this patient, cervical spine imaging is recommended in all such cases before surgery.

Cardiac anomalies, such as ventricular septal defects, tetralogy of Fallot, and patent ductus arteriosus, occur in approximately 5% to 58% of patients with GS. [8] Hence, preoperative echocardiography is mandatory to detect any hemodynamic compromise that may influence anesthetic drug selection or intraoperative fluid management.

Neuromuscular blocking agents, such as rocuronium, are preferred for their rapid onset and reversibility (as in this case). Sevoflurane was chosen for maintenance due to its favorable hemodynamic profile and rapid emergence.

Postoperative care involved close monitoring for airway obstruction due to residual hypoventilation or airway edema. Patients with craniofacial anomalies are at risk of obstructive sleep apnea, necessitating prolonged observation.

Multidisciplinary involvement, including Pediatric Anesthesiologists, otorhinolaryngologists, and intensivists, is essential for optimal outcomes.

CONCLUSIONS

GS presents significant anesthetic challenges due to craniofacial abnormalities, airway difficulties, potential cervical spine anomalies, and associated systemic defects. Proper preoperative evaluation, anticipation of airway challenges, appropriate preparation with advanced airway devices, and meticulous intraoperative monitoring are pivotal for safe anesthetic management. This case emphasizes the importance of individualized planning in the successful conduct of general anesthesia in an adolescent with GS undergoing auricular reconstruction.

CONSENT

Written informed consent was obtained from parents for the publication of this case report and all associated images.

AUTHORS' CONTRIBUTION

All authors have significantly contributed to the work, whether by following the case at the bedside, conducting literature searches, drafting, revising, or critically reviewing the article. They have given their final approval of the version to be published, have agreed with the journal to which the article has been submitted, and agree to be accountable for all aspects of the work.

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CONFLICT OF INTEREST

None.

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