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CASE REPORT

An Unusual Case of Difficult Airway and a Serious Complication of Intubation Due to Forestier Syndrome

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Abstract

Forestier syndrome, also known as Diffuse Idiopathic Skeletal Hyperostosis (DISH), is characterized by typical findings of ossification and calcification primarily of the anterior longitudinal ligament, which result in osteophyte formation. This is a rare condition and is rarely associated with systemic diseases such as diabetes mellitus and obesity. It occurs mostly in the fifth and sixth decades of life and predominantly in males. This report describes a case of a 68-year-old man with DISH syndrome, who experienced a difficult airway and a serious complication during intubation.

Keywords: Difficult airway; Forestier; Hyperostosis.

Forestier syndrome was first described in 1950 by Forestier and Rotes-Querol, and was named diffuse idiopathic skeletal hyperostosis (DISH) by Resnick et al.[1] In 1975, with typical findings of ossification and calcification primarily of the anterior longitudinal ligament, which results in osteophyte formation[2]. Otolaryngological symptoms such as dysphagia, pharyngeal globus, dysphonia, difficulty in breathing, cough, and sore throat can be observed. Although the disease is not rare, it has a low diagnostic rate. Here, we present a case of Forestier syndrome, in which esophageal perforation developed after difficult intubation.

Case Report

A 68-year-old male presented for cataract surgery. His ASA physical status assessment was II (well-controlled diabetes mellitus/hypertension). The Mallampati score was recorded as grade four, and the patient's neck movements were slightly limited. There were no remarkable records in his preanesthetic examination notes or history.

Equipment for difficult intubation was also prepared. On laryngoscopic examination, the Cormack-Lehane scale score was grade IV. Laryngeal mask insertion was attempted twice, with different sizes, but the settlement

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of the LMA was not successful. A direct laryngoscopic attempt at endotracheal intubation resulted in esophageal intubation, followed by successful tracheal intubation via videolaryngoscopy. Ophthalmologic surgery and recovery periods were uneventful. The patient was discharged on the following day. Two days after the operation, the patient was admitted to the emergency department with a red, warm, swollen neck and difficulty in swallowing and breathing.

Radiography and computed tomography of the neck revealed widespread air images, starting from the left parapharyngeal region to the anterior mediastinum and supraclavicular region. On the sagittal plane, calcification of the whole anterior cervical ligament, osteophyte formation, and ossification between the C4 and C6 levels compressing the posterior esophagus were detected (Fig. 1).

Wide-spectrum antibiotics with anaerobic coverage were started in the ED, and the patient was transferred to the ICU and electively intubated using videolaryngoscopy, sedation and mechanical ventilation were started. When the patient's history was questioned in detail, a family member stated that he had been admitted to the emergancy departmant twice with the complaint of inability to swallow pills. The anesthesiologist stated that there was a light-colored, protruding lesion on the posterior wall of the hypopharynx, which she noticed during laryngoscopy and thought it might have been malignant lesions. The patient underwent multiple surgical consultations with the diagnosis of an esophageal perforation. The cervical CT scan was repeated on the 4th day. Abscess formation was detected in the anterior cervical region, at the level of the left thyroid gland and thyroid isthmus, tracing back to the glottis. An ultrasound-guided drainage catheter was placed, and a second catheter was placed on the superior side of the left thyroid gland.

Further manipulation or examination of the upper airway or gastrointestinal tract was avoided. The patient was kept intubated under sedation and mechanical support and fed parenterally. A tracheostomy was not possible because the abscess was located in the anterior region of the tracheal ring. After a week of deep sedation, light sedation, sedation holidays, asist modes of ventilation were applied to keep the patient cooperated and to facilitate weaning from the ventilator. On the 18th day, when the inflammatory status regressed, endoscopic assessment of the hypopharynx and esophagus was performed. At 25–30th mm distal to the epiglottic level, hyperemic mucosal tissue and a pock-shaped lesion covered by epithelium just above the compression site were observed, which was thought to be the healed esophageal perforation site (Fig. 2). The drainage catheters were withdrawn on the 25th day and weaned off the ventilator on the 30th day after intubation. He experienced moderate weakness in both limbs, and after electromyographic assessment, was diagnosed with critically ill neuromyopathy. The patient was discharged to the ward for further treatment.

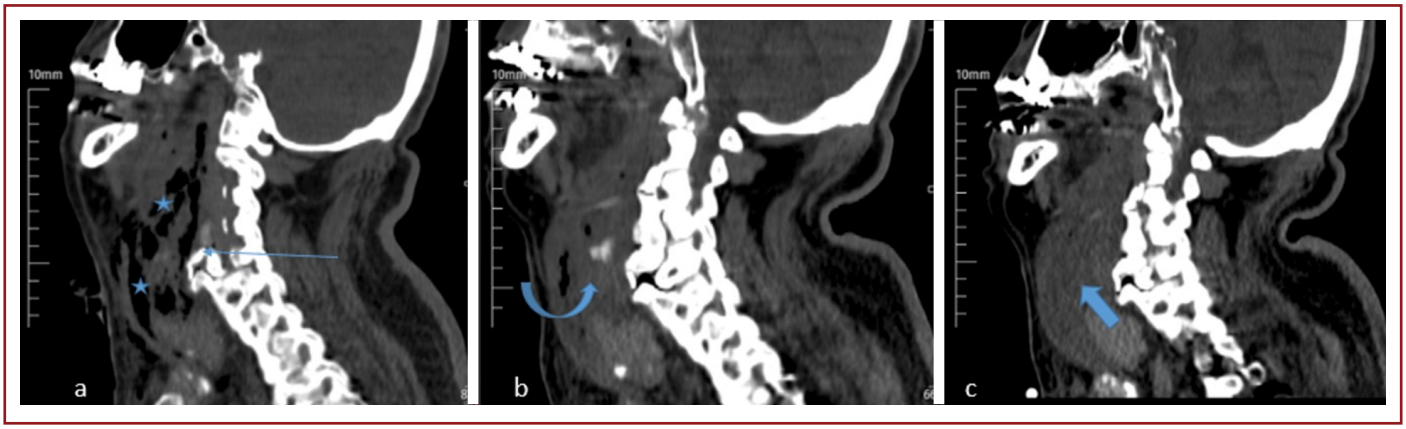

Figure 1. (a) On the 2nd day of the postoperative period, there was excessive skeletal hyperostosis on the C3–C7 vertebrae levels, which were mostly localized anteriorly to the end-plates and were almost bridging (arrow). They were protruding anteriorly and impinging upon the posterior wall of the esophagus at the C5–6 level, causing narrowing at the level of the osteophytes. Additionally, there were some air bubbles visualized in the adjacent soft tissue, which were suspected to indicate laryngeal or esophageal perforation (stars). **(b)** On the 6th day CT, air bubbles were almost completely resolved, but there was excessive soft tissue edema and hypodensity in the surrounding areas (curved arrow). **(c)** On the 10th day CT, reactive edema was persistent in adjacent areas, but no air was detected in the soft tissues (thick arrow).

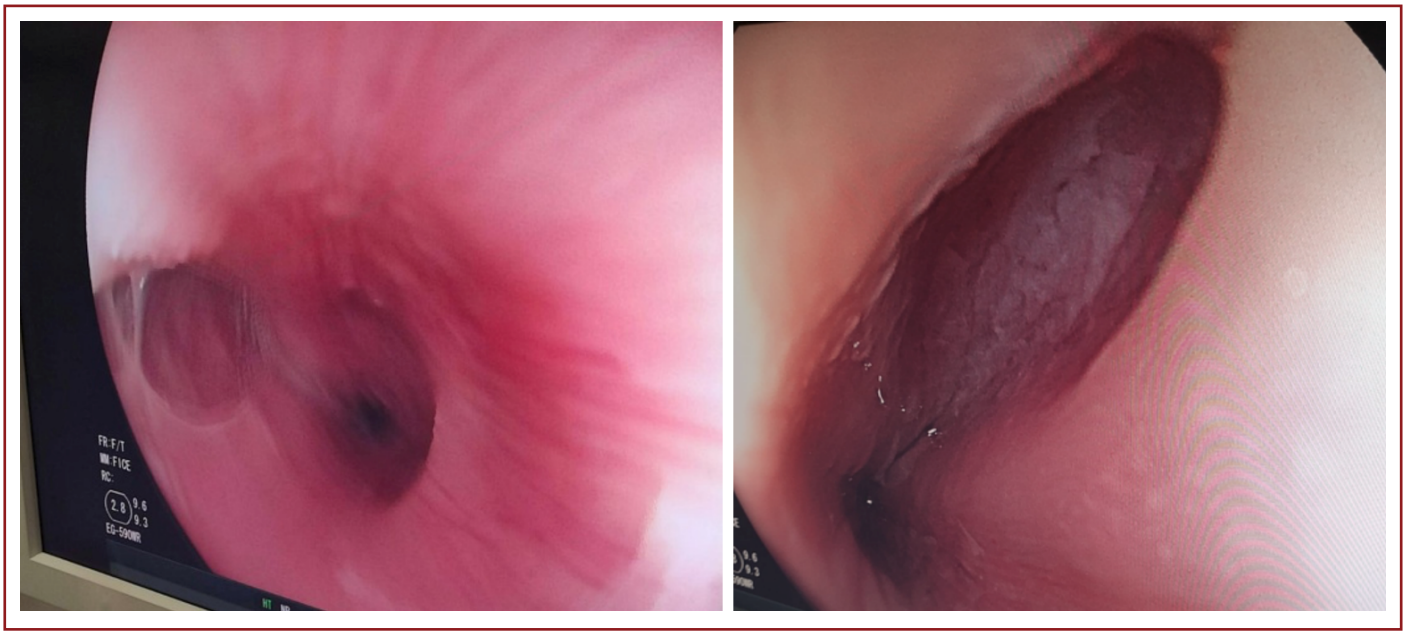

Figure 2. Endoscopic assessment of the oesophageus. At -25-30th mm distal to the epiglottic level-, hyperemic mucosal tissue and a pocket-shaped lesion covered by epithelium were seen, showing healed esophageal wall damage after perforation.

Discussion

The prevalence of DISH was reported to be 42.0%, depending on the classification criteria used and the presence of risk factors in the study population. DISH is associated with older age, male sex, obesity, hypertension, atherosclerosis, and diabetes mellitus[3].

The cervical spine is frequently affected, and large anterior cervical osteophytes cause compression of the esophagus; however, the upper respiratory tract is regularly reported in ENT and pulmonology journals, as well as by anesthesiologists, as a cause of intubation difficulties. When large osteophytes causing displacement of the pharynx and trachea are detected, awake intubation using fiberoptic bronchoscopy is preferred. Laryngoscopy, videolaryngoscopy, and laryngeal mask airway can be performed. Cases of subcutaneous emphysema and laryngeal perforation, sometimes leading to serious infections and death, have been reported following laryngoscopy and intubation in patients with DISH[4,5].

In our case, the risks of intubation could not be foreseen, as the patient was not evaluated in further detail during the preanesthetic examination. The anesthesia team prepared for difficult intubation but had no idea of the possibility of DISH. The posterior wall of the esophagus was probably damaged between the protruding osteophytes and endotracheal tube when the tube was mistakenly inserted into the esophagus.

Special attention was paid to avoid irritation and manipulation of the site of suspected perforation. Immediately after abscess formation was detected, drainage catheters were placed to prevent the development of mediastinitis. We performed frequent neck ultrasound assessments to follow up for inflammation and regression of the abscess site while keeping the patient under broad-spectrum antibiotics against aerobic and anaerobic microorganisms. We postponed extubation attempts after endoscopic assessment, which were also avoided in the first two weeks. Successful weaning was achieved, although not early.

Conclusion

DISH involving the cervical spine is a complicated cause of dysphagia and neck stiffness. Due to the anatomic variation of the pharynx secondary to DISH, patients undergoing any kind of endoscopic or bronchoscopic procedure are at risk of perforation. From an anesthesiologist's point of view, such a complication can be extremely disappointing, and for the patient, it is life-threatening. Any suspected patient presented for preanesthetic evaluation should be questioned for the signs and symptoms of DISH, and radiologic assistance should be sought.

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