

Airway Management of a Patient with Undiagnosed Diffuse Idiopathic Skeletal Hyperostosis – DISH

Peixoto AR*, Silva R, Antunes MJ and Moreira A

Department of Anesthesiology, Centro Hospitalar Universitário de São João, Hernâni Monteiro, Portugal

Abstract

Diffuse idiopathic skeletal hyperostosis (DISH), also known as Forestier's syndrome was first described in 1950 by Forestier and Rotes-Querol. It is a rheumatologic disease, with unclear etiology. DISH is characterized by spinal osteophyte formations and thickening and calcification of the ligaments, resulting in abnormal ossification of the paravertebral ligaments and muscles. DISH incidence is high but is often undiagnosed, leading to significant morbidity and mortality. Herein, we report the case of a 72-year-old female scheduled for surgical correction of carpal tunnel syndrome. She was classified as American Society of Anesthesiologists classification II. As the anesthesia was induced, we realized that it was a "cannot ventilate, cannot oxygenate" situation, and a supposedly simple procedure became a life threatening emergency.

Keywords: Diffuse idiopathic skeletal hyperostosis; Difficult airway; Perioperative complications

Introduction

Diffuse idiopathic skeletal hyperostosis (DISH), also known as Forestier Disease, was first described in 1950 by Forestier and Rotes-Querol [1]. Due to abnormal bone cell activity under the influence of metabolic factors as characterized by an ossifying diathesis, it affects the spinal and peripheral locations. Affection of the cervical spine by DISH can make airway management difficult in various ways [2-4]. Here, we report a case of undiagnosed DISH, which revealed an extremely difficult airway management situation that led to the subsequent diagnosis of DISH.

Case Report

A 72-year-old female patient with a body mass index of 30.1 kg/m², American Society of Anesthesiologists classification II (arterial hypertension and dyslipidemia), no medical allergies, and diagnosed with carpal tunnel syndrome, was scheduled for surgical decompression of the median nerve under general anesthesia. Four years earlier, the patient was scheduled for surgical correction of lumbar stenosis under general anesthesia without any anesthetic complications. Preoperative results of biochemical studies and electrocardiography were normal. Airway evaluation showed a Mallampati grade II, upper lip bite test class I, tiromentonian distance of >6 cm, interincisive distance of >3 cm, without mouth opening limitation, but a limited cervical mobility. Standard monitoring and bispectral Index were used. A good preoxygenation was performed. For anesthesia induction, an intravenous fentanyl (0.002 mg/kg), lidocaine (1 mg/kg), and propofol (2 mg/kg) were administered. Patient lost consciousness and respiratory drive; hence, we tried to ensure oxygenation by manual ventilation using oropharyngeal airway (Guedel airway tube) but no improvement in the ventilation or oxygenation was achieved.

A very stiff neck, without mobility was noted. After position optimization, ventilation with 4 hands was done but without any improvement. As initially planned, we placed a supraglottic device (iGel[®] laryngeal mask number 4), but later on realized that manual or controlled ventilation was impossible. Vital signs remained stable. After replacing and adjusting the supraglottic device without success, we asked for assistance, and then decided to place an orotracheal tube. Direct laryngoscopy was performed by an experienced anesthesiologist, but it revealed a Cormarck Lehane grade IV. Therefore, a blind intubation was tried but unfortunately tracheal intubation was impossible. Any advanced equipment like videolaryngoscope or fiberoptic was immediately available, and as the patient started to desaturate, we decided to perform a surgical cricothyroidotomy. Fortunately, as we were ready to perform the procedure, the patient started to breathe spontaneously, peripheral oxygen saturation started to rise, and the entire anesthetic and surgical team decided to wake the patient and cancel the procedure to investigate the reason for the difficult airway since she had a

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*Correspondence:

Ana Ritinha Peixoto, Department of Anesthesiology, Centro Hospitalar Universitário de São João, Porto, Portugal.

Tel: 00351933859520

E-mail: ritinhapeixoto@hotmail.com

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Figure 1 and 2: Showing severe ossification and spondylosis of anterior longitudinal ligament between C3 and C6 with deformation of the posterior wall of hypopharynx.



Figure 3 and 4: Showing a beak-like projection of osteophytes on the 4th and 5th cervical vertebrae compressing the esophagus and consequentially the posterior wall of the trachea.

previous general anesthesia without any problem.

To investigate this situation, many examinations were carried out. A laryngofiberscopy was performed by an otolaryngologist, which showed protrusion of posterior pharyngeal wall.

Magnetic resonance imaging was done, and surprisingly it revealed a severe ossification and spondylosis of anterior longitudinal ligament between C3 and C6 with deformation of the posterior wall of hypopharynx. A beak-like projection of osteophytes on the 4th and 5th cervical vertebrae was compressing the esophagus and consequentially the posterior wall of the trachea (Figures 1 to 4). These findings were consistent with DISH, which was considered to be the cause of difficult intubation. The patient denied dysphagia, cough, sore throat, hoarseness or foreign body sensation, but mentioned cervical pain and stiffness that had worsened from past few months.

Discussion and Conclusion

DISH is a rheumatological condition whose primary manifestation is the ossification of the anterior longitudinal ligaments, which mostly affects the axial skeleton. It is a common disease of old age, but as it remains asymptomatic in many cases, diagnosis needs high suspicion. If undiagnosed, it can lead to significant morbidity and mortality. It is characterized by multiple axial and extra axial involvement and symptoms such as dysphagia due to distortion or compression of the

esophagus, cough, sore throat, hoarseness or foreign body sensation in the pharynx, sleep apnea, cervical pain and stiffness [5], but usually is asymptomatic and therefore latent. These changes are most often seen in the spine; however, DISH can affect almost any part of the skeleton, including the hips, knees, ankles, feet, shoulders, hands and ribs [5]. It affects mostly the thoracic levels, but may affect cervical spine too [6]. Mechanisms of difficult intubation are poorly understood, but some reports mention difficulty intubation and insertion of supraglottic devices due to mass-like protrusion of posterior pharyngeal wall as in our case [4].

Several studies have revealed a significant association between DISH and metabolic disorders, such as obesity, diabetes mellitus, hyperuricemia, dyslipidemia, cardiovascular disease and use of certain medications like isotretinoin [4,7].

Some studies show difficult intubation and spinal cord injury when managing the airway in patients with DISH. The exact etiology of DISH is unknown but, histopathological features include the following: focal and diffuse calcification and ossification of the anterior longitudinal ligament, paraspinal connective tissue and annulus fibrosus, degeneration of the peripheral annulus fibrosus fibers, anterolateral extension of fibrous tissue, hypervascularity, chronic inflammatory cellular infiltration, and new bone formation at the anterior surface of the vertebral bodies [8-10].

The presence of cervical stiff, recent dysphagia, dyspnea or sleep apnea, should alert all the anesthesiologists about the upper-airway obstruction due to large anterior cervical osteophytes. This mechanical distortion may complicate laryngoscopy and tracheal intubation.

We decided to report this case since it was a non-predictable difficult airway management. Fortunately, it is a rare situation, but is a real emergency if it happens. This report could help anesthesiologists to be alert of some symptoms such as recent dysphagia and cervical stiffness, which are sometimes devalued.

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