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Airway Management Using the I-gel Supraglottic Airway Device in Patients with Grisel's Syndrome -Case Report-

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그리셀증후군 환자에서 I-gel 성문상기도유지기를 사용한 기도관리 -증례보고-

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Abstract Grisel's syndrome is a non-traumatic subluxation of the atlantoaxial joint with an inflammatory condition in the adjacent soft tissues. Due to the instability of the cervical spine, careful airway management is crucial to prevent potential cervical spinal cord injury following airway manipulation. We successfully secured the patient airway using a supraglottic airway device (I-gel) in a patient who had previously diagnosed with Grisel's syndrome. The operation was successfully completed, and the patient recovered without any neurological complications. I-gel can be a good option for airway management during general anesthesia in a patient diagnosed with Grisel's syndrome.

Key words : Atlantoaxial instability, Cervical spinal cord injury, Difficult airway, Grisel's syndrome, Saglottic airway

요 약 그리셀 증후군은 환축관절의 비외상성 탈구로 인하여 전신마취 시 경추손상이 발생할 가능성이 있어 신중한 마취관리를 요한다. 이에 우리는 과거에 그리셀 증후군을 받은 진단받은 환자의 전신마취 하 전립선 레이저절제술 시 I-gel을 이용한 기도관리를 성공적으로 진행하였고, 환자는 신경학적 합병증 없이 잘 회복된 사례를 보고하고자 한다. 그리셀 증후군 환자의 마취 관리 시 기도관리를 위하여 I-gel을 사용하는 것이 좋은 대안이 될 수 있을 것이다.

주제어 : 환축관절불안정, 경부척수손상, 어려운 기도관리, 그리셀증후군, 성문상기도유지기

1. Introduction

Grisel's syndrome is a non-traumatic subluxation of the atlantoaxial joint with an inflammatory condition in the adjacent soft tissues [1]. In 1930, Pierre Grisel first described three patients with pharyngitis and torticollis who had dislocation of the C1-C2, the

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atlantoaxial joint [2]. Since then, the condition has been called Grisel's syndrome (GS), with numerous papers indicating a strong association between its occurrence and several infectious disease or surgery in head and neck region [3-5]. Anesthetic challenges for GS includes difficult airway management due to cervical spine instability. Laxity of the atlantoaxial ligaments and/or bony erosion of the insertion site of the ligaments causes various degree of atlantoaxial joint subluxation and instability [6]. Consequently, any sudden movement of patient's head and neck during airway manipulation such as laryngoscopy and tracheal intubation might cause neurological damage. In this case report, we report a case of successful airway management using supraglottic airway device (I-gel) in a patient who had previously been diagnosed with GS.

2. Case Report

The authors received written informed consent from the patient for his case details and the accompanying images published. A 72-year-old man (height: 170 cm; body weight: 68 kg) was requested to undergo anesthesia for holmium laser enucleation of the prostate (HoLEP) for benign prostate hyperplasia (BPH). The patient had a previous history of receiving surgery and subsequent chemotherapy for tonsillar cancer about 20 years ago. The patient stated that normal airway management procedures were performed at the time of tonsillar surgery. And 8 years ago, the patient was diagnosed with GS. At that time, the patient complaint of posterior neck pain, and magnetic resonance imaging (MRI) revealed the broad inflammation around odontoid process of C2, atlantoaxial joint, and adjacent soft tissue and concomitant atlantoaxial subluxation and cord compression. Conservative treatment such as cervical collar apply, antibiotic treatment, and percutaneous drainage of abscess was performed because the patient refused to receive surgery, and the patient was lost to follow-up.

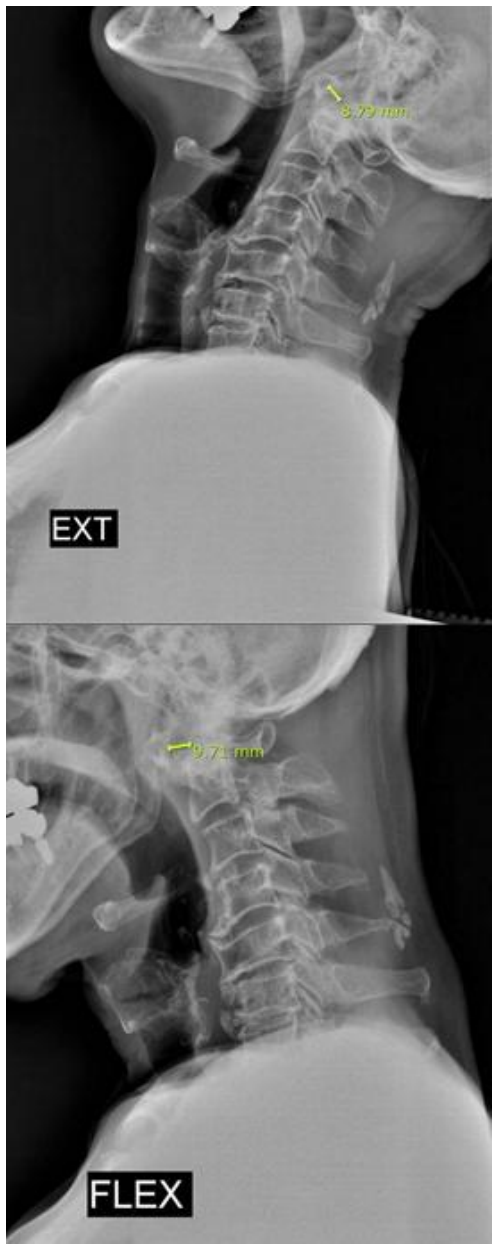

Fig. 1. Measurement of ADI in extension and flexion of lateral cervical spine x-ray indicating atlantoaxial joint subluxation

Cervical spine plain x-ray and MRI were performed at 6 weeks prior to the surgery, and the findings were assessed by neuroradiology and neurosurgery specialists. As shown in Fig. 1, compared to the previous imaging study, the inflammation had improved, but atlantoaxial joint instability was still observed with an atlantodental interval (ADI) of 8.79 mm at extension and 9.71 mm at flexion. The MRI scan of Figure 2 also showed that the ADI was increased to 8.73 mm. Normal measurement of ADI ranges 1-3 mm in adults [7], although age and sex may affect their normal value [8,9]. The patients had complaint no severe and critical symptoms such as posterior neck pain or weakness of extremities, and he again refused to undergo surgical treatment for GS.

The patient's lung was appropriately ventilated in a pressure-controlled ventilation mode with peak inspiratory pressure of 14 cmH_2O and I:E ratio of 1:2. End-tidal CO_2 was maintained at 35 to 40 mmHg during the operation. The operation time was approximately 1 h, and no remarkable events occurred. There was no additional dose of neuromuscular blocking agent until the end of surgery. At the end of surgery, inhalation anesthetics were discontinued, and residual neuromuscular blockade was reversed with neostigmine 2.0 mg and glycopyrrolate 0.4 mg at the reappearance of fourth twitch response during train-of-four count. The I-gel was removed just after the recovery of consciousness and appropriate spontaneous breathing. The patient was discharged to the postanesthesia care unit for close monitoring, and he didn't admit any neurological deterioration. On the 5^{th} postoperative day, he was discharged without any perioperative complications.

Fig. 2. Measurement of ADI in cervical spine magnetic resonance imaging

3. Discussion

GS is a rare syndrome characterized by nontraumatic subluxation of the atlantoaxial joint and inflammatory condition on the adjacent soft tissues [1]. Pathologic laxity of the ligaments and/or bony erosion of the insertion site of the ligaments surrounding the upper cervical spine causes the atlantoaxial joint subluxation and instability [6,11]. A recently proposed hypothesis on the pathogenesis of GS suggests that inflammation of the paravertebral tissue may extend to the atlantoaxial joint through the pharyngo-vertebral veins via numerous lymphovenous anastomoses [1,12]. Several infectious disease or inflammatory condition following otolaryngologic surgery is associated to the development of GS [3-5]. Although the majority of the affected patients present pediatric onset, it can also occur in adult age group [5]. In the current case, local inflammation and abscess following the surgery and subsequent chemotherapy for tonsillar

cancer might be the cause of adult-onset GS of the patient.

The early stage GS might be successfully treated with conservative treatment including cervical brace and antibiotic treatment when needed in patients with no definite displacement [13]. However, higher degree of atlantoaxial instability with ventral displacement, greater than 5 mm of ADI, may require more advanced care such as surgical treatment or halovest apply. In our patient, the ADI value was 8.73 mm, and this corresponds to type III by Fielding and Hawkins classification, which surgical intervention is usually indicated [14]. However, the neurological symptoms of the patient had been minimal for about 8 years, and the patient refused the surgical correction. Therefore, specialized anesthetic plan, based on that of traumatic cervical spine instability patient, was applied for his HoLEP surgery.

Several researches emphasized that airway manipulations including chin lift/jaw thrust as well as conventional direct laryngoscopy might cause narrowing of spinal canal and consequently cord injury in cadaveric model with atlantoaxial instability [15,16]. However, laryngeal tube, one of the old-generation supraglottic airway device, barely caused dural sac compression compared to conventional direct larygoscopic endotracheal intubation [16]. Other supraglottic airway devices also manifested successful performance on airway management with less cervical motion compared to conventional laryngoscopy in a cadaveric study [17]. I-gel, a second-generation supraglottic airway, is non-inflatable, preformed, and gel-like cuff with an esophageal vent [18]. It can be easily inserted without neck extension in neutral position, and it also has advantage of unnecessity for position change after cuff inflation. Therefore, it might also be safely applied to patients with suspected cervical spine injury as suggested by Haske et al. [19].

Although the effect of manual in-line stabilization technique on preventing spinal cord injury is controversial [20-23], it has been still recognized as one of the point of care during anesthesia induction for traumatic cervical spine injury patient. This technique, however, often disturbs glottic view, and even result in failed intubation. Moreover, I-gel sometimes fails to ventilate patient's lung appropriately. In these situations, fiberoptic-guided intubation can be optimal for placing endotracheal tube by visualizing vocal cord indirectly. Thus, fiberscope should also be prepared in advance for the potential I-gel failure.

In summary, we performed airway management using the I-gel, a supraglottic airway device, in a patient who had been previously diagnosed with GS, for HoLEP surgery under general anesthesia. Securing airway with I-gel with manual in-line stabilization might be carefully tried in patients with atlantoaxial instability. Fiberoptic-assisted endotracheal intubation should be prepared for potentially failed I-gel insertion or ventilation.

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