Case Report
Central venous catheter malposition in the azygos vein and difficult endotracheal intubation in severe ankylosing spondylitis: a case report

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Abstract: Ankylosing spondylitis (AS) can be challenging for anesthesiologists because central venous access can be difficult, and the airway can be blocked due to the fixed flexion deformity of the spine. In this case, we attempted central access via the right subclavian vein, but the catheter was repeatedly inserted into the azygos vein, which was confirmed by radiology. After several attempts, the catheter position was corrected at the superior vena cava-atrial junction. Although several useful devices have been developed to address difficult intubation, in this case, fiberoptic bronchoscopy was the only applicable safe alternative because of the patient’s extremely severe chin on chest deformity and temporomandibular joint disease. We report a successful awake fiberoptic bronchoscopic intubation in a patient with extremely severe AS and recommend that the catheter placement should be confirmed with radiology to ensure proper positioning for severe AS patients.

Keywords: Ankylosing spondylitis, azygos vein, central venous catheters, difficult airway

Introduction
Ankylosing spondylitis (AS), an autoimmune seronegative spondyloarthopathy, is a painful chronic inflammatory disease that causes arthritis of the spine and sacroiliac joints. Clinical manifestations include limited mouth opening, atlanto-axial subluxation, fractures with little or no history of trauma, and limited cervical movement. AS patients present challenges for performing central venous access and secure airway procedures; however, to correct spinal deformity, it is important to obtain central venous access in order to manage massive bleeding, provide medication infusions, and secure the airway for general anesthesia. The possible complications of central venous catheterization are pneumothorax, vessel injury, and malposition. We tried to place the central venous catheter in the accurate position despite difficulties and malpositioning in the azygos vein. Ultimately, we carefully performed awake fiberoptic bronchoscopic intubation.

Case report
A 48-year-old male patient presented with extremely severe AS and had a severe chin on chest deformity that had been diagnosed 15 years previous with complaints of five-year symptoms of very severe back deformity, persistent backache, and inability to lie flat. Gradually, the deformity in his hip and back increased in the form of forward bending, and he was unable to look straight ahead. The patient’s height and weight were 120 cm and 65 kg, respectively.

The patient had a fixed flexion deformity of the cervical and thoracolumbar region, a chin-brow vertical angle fixed at 140 degrees, and the patient’s chin was fixed near the pubic symphysis (Figure 1). The patient’s Mallampati score was 4, and he was unable to open his mouth more than 2 cm due to temporomandibular joint disease. Additionally, there was little space between his chin and chest, which indicated a difficult airway. A whole spine computed tomography (CT) scan showed that the patient had advanced AS with bamboo spine formation, complete bony ankylosis, and severe kyphosis of the entire spine.

He was scheduled for sequential bilateral hip resectional arthroplasties and staged spinal
osteotomies, followed by bilateral total hip arthroplasties. A central venous catheter insertion was planned preoperatively in preparation for massive bleeding. We attempted a subclavian catheter insertion, in expectation of the difficulty of an internal jugular approach due to the fixed flexion of the cervical spine. The right subclavian vein was punctured using a micro-puncture needle under ultrasound guidance, and then a micro-wire and sheath insertion were performed, although several attempts were necessary due to severe resistance.
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However, when confirming the catheter location using a venogram, we determined that the contrast media was flowing into the azygos vein, although accurate imaging was not possible due to the spinal deformity. We next performed a CT angiography and confirmed that the catheter was located in the azygos vein (Figure 2). After the inserted catheter was removed, we again attempted to place the catheter at the superior vena cava (SVC) under fluoroscopy; however, it was not possible to intravascularly determine the course of the catheter. We determined, through a venogram, that the tip of catheter was located in the right ventricle, and we ended the procedure after withdrawing the catheter to the SVC-atrial junction (Figure 3).

We planned awake fiberoptic intubation to secure the airway and reduce the risk of neurological injury. Although the use of tracheostomy, retrograde intubation, or intubating laryngeal mask have been reported in other AS patients, those procedures could not be attempted in our patient due to the acute angulation of the chin on chest deformity. In the sitting position, the patient’s oropharynx was bathed with 10% xylocaine, and then an anesthesiologist performed fiberoptic bronchoscopy while standing in front of the patient (Figure 4). With light sedation using intravenous remifentanil and propofol, and local anesthesia using lidocaine around the vocal cord and trachea, the endotracheal tube was safely inserted into the trachea. After confirming adequate ventilation, the surgery was continued. All surgeries were performed six times over four months. After surgery, the patient could walk independently, had forward vision, and an improved psychological outcome.

Discussion

AS is a chronic autoimmune disorder that usually involves the spine and sacroiliac joints, and in some severe cases can cause organ compression or nerve injury. Typically, lesions emerge at sacroiliac joints and then gradually involve the total spine; the cervical spine is affected in half of AS patients. AS at the cervical spine causes anatomic abnormalities and limited cervical mobility, which lead to difficult head manipulation. Temporomandibular joint involvement causes limited mouth opening and can make it nearly impossible in advanced stages.

In severe AS corrective osteotomy, perioperative massive bleeding is anticipated, and central venous catheterization is commonly conducted for vascular access for transfusion and volume replacement, drug infusion, and central venous pressure monitoring. Although it is considered relatively safe, central venous catheterization should be performed carefully in order to avoid complications. Procedure-related complications including pneumothorax, hematoma,
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vascular injury, arrhythmia, and catheter malposition are all possibilities [1].

The optimal position for the central venous catheter tip is the junction of the SVC and right atrium, without contacting the pericardial reflection, in order to avoid cardiac perforation or tamponade and to reduce the risk of dysrhythmia [2]. Malpositioning of the central venous catheter is caused by placement into an incorrect vein or advancement too distally, which causes kinking in the catheter. Incorrect veins usually include major tributaries of the superior vena cava and, rarely, smaller braches of the central vein.

In cervical fusion cases such as AS, the internal jugular vein approach is generally very difficult. We expected that the internal jugular approach would not be possible in this patient and decided to try right side-subclavian catheterization under radiologic image guidance. In our case, CT angiography showed that the azygos vein was running alongside the inferior vena cava, which we suspected would allow the microwire tip to be easily inserted in the azygos vein. Inadvertent placement of the central venous catheter into the azygos vein is rare (1.2%) [3]. Risk factors for malpositioning into the azygos vein include left internal jugular venous access, use of catheters with long venous tips, and catheter insertion in a patient with fluid overload. Although a catheter is placed into the azygos vein intentionally as an alternative vein in case of severe venous occlusion [4], that involves several vulnerabilities caused by relatively small vascular caliber and direction of blood flow [5]. Furthermore, placement of a central venous catheter into the azygos vein carries a risk of venous perforation with mediastinal hemorrhage, venous thrombosis, and complete vein occlusion, which are related to significant morbidity and mortality, unless the complication can be recognized and corrected immediately. Hence, we tried to correct the catheter position, despite several failed attempts.

Drakonaki et al. reported a migratory azygos vein after kyphosis angled at the level of the fourth thoracic vertebra, and the authors demonstrated that verticalization of the azygos fissure could affect vein migration in spinal deformities [6]. Similarly, we assumed that the kyphosis of the thoracic vertebra of our patient could increase intrathoracic pressure, modify intrathoracic structure, and affect the course of the azygos arch, which could misdirect the catheter to the azygos vein.

In patients with difficult airways, airway management can be performed using fiberoptic bronchoscopic intubation, an intubating laryngeal mask, retrograde intubation, or tracheostomy [7, 8]. In addition, several different types of video laryngoscopes have been introduced, each with a different blade shape, user interface, and tube insertion strategy for promising intubation, for example, the Glidescope, Pentax Airway scope, or Airtraq laryngoscope; it has been reported that the Glidescope and Pentax Airway scope provide a significantly better...
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laryngoscopic view than the Macintosh laryngoscope in patients with limited cervical mobility [9, 10]. However, in severe AS cases, like our patient, such devices cannot be applied due to serious skeletal transformation. For patients with severe AS, the possibility of neurological injury caused by excessive neck extension due to chronic cervical kyphosis should be considered. Catastrophic neurological complications such as quadriplegia after emergent endotracheal intubation resulting in cervical fracture dislocation in a patient with AS have been reported [11]. In our patient, neither tracheostomy, retrograde intubation, nor other devices such as video laryngoscopes could be applied for airway management due to chin on chest deformity and mouth narrowing caused by temporomandibular joint involvement. Due to the angle of the oropharynx axis, laryngeal mask airway was also not appropriate in this case, especially because of concern for kinking and unsuitable placement over the trachea. Fiberoptic bronchoscope was the only available intubating device, and awake intubation was also useful for constant neurological assessment during endotracheal tube insertion. Awake fiberoptic intubation is considered the safest and most effective method that allows continuous neurological monitoring in suspected cases of difficult airway or unstable cervical spine [12].

In conclusion, our experience indicates that awareness of the possibility of central venous catheter malposition and difficulty with intubating a patient with AS are important for successful procedures and to attenuate complications. It is important to identify an accurate catheter position to reduce the risk of complications in AS patients. In particular in patients with risk factors, it is difficult to detect the malposition of the catheter tip during a blind procedure; therefore, using image guidance such as ultrasonography, venogram, or CT scan might be crucial for identifying malposition. Finally, death from loss of airway can occur in difficult airway, especially severe AS patients, so it is important to select the safest airway access judiciously when treating patients with severe AS.

Disclosure of conflict of interest

None.

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