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Case Report

Anaesthesia Section

Anaesthetic Considerations for a Patient with Kartagener's Syndrome Undergoing Emergency Surgery: A Case Report

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ABSTRACT

Kartagener Syndrome (KS) is a rare genetic disorder resulting from autosomal recessive inheritance and is characterised by ciliary dyskinesia. It typically presents with the distinctive triad of chronic sinusitis, situs inversus, and bronchiectasis. KS poses distinct challenges during the perioperative period due to its potential impact on pulmonary function and cardiovascular anatomy. The present case details the successful perioperative management of a 55-year-old male patient with KS who underwent emergency knee-spanning external-fixator application under regional anaesthesia. The patient's medical history included a chronic diagnosis of bronchiectasis, for which he was on irregular medications. His clinical examination revealed Grade 2 digital clubbing. Auscultation findings included heart sounds auscultated on the right hemithorax, with the apical impulse palpable in the right fourth intercostal space, indicative of dextrocardia associated with situs inversus. Diagnosis of Kartagener syndrome was confirmed with Computed Tomography (CT), which showed the cardiac apex and aortic notch on the right side and bronchiectasis changes predominantly involving the bilateral lower lobes. Given the immediate need for surgery, neuraxial anaesthesia under ultrasound guidance was planned to address the patient's complex respiratory profile. In this case, employing a combined spinal-epidural anaesthesia technique facilitated effective pain management and haemodynamic stability while minimising the respiratory complications typically associated with general anaesthesia.

Keywords: Intraoperative management, Respiratory pathology, Ultrasound-assisted neuraxial technique

CASE REPORT

A 55-year-old male presented to our emergency department after a fall from a significant height, resulting in a severe injury to the right lower extremity. Orthopaedic assessment identified a complex post-traumatic injury, namely a Gustilo-Anderson grade III B open fracture of the proximal tibia with concomitant patellar dislocation. The surgical plan was the application of a knee-spanning external fixator to stabilise the injury and facilitate optimal healing.

The patient's medical history included a chronic diagnosis of bronchiectasis for 20 years, managed with Deriphyllin (a combination of theophylline and etophylline) 150 mg. He was also being treated for Type 2 Diabetes Mellitus (T2DM) with metformin 500 mg twice daily. Additionally, he had a history of chronic sinusitis for the past 20 years, though adherence to pharmacotherapy for this condition had been irregular. The patient was assessed as American Society of Anesthesiologists (ASA) III due to poorly controlled T2DM and chronic bronchiectasis.

Upon clinical evaluation, the patient presented with productive cough and sputum production, consistent with the existing pulmonary pathology. Examination revealed Grade 2 digital clubbing. Auscultation showed heart sounds on the right hemithorax, with the apical impulse palpable in the right fourth intercostal space, indicative of dextrocardia associated with situs inversus. A CT scan of the thorax confirmed situs inversus totalis, with the cardiac apex and aortic notch on the right side. Imaging also revealed findings consistent with chronic bronchitis and bronchiectasis, predominantly affecting the bilateral lower lobes. Based on these findings, the diagnosis of KS was established. Young syndrome (triad: chronic rhinosinusitis, bronchiectasis, obstructive azoospermia) was considered as a differential but was ruled out due to the presence of situs inversus, which favour KS.

Laboratory investigations were unremarkable, showing no acute biochemical abnormalities. A 12-lead Echocardiogram (ECG) was performed with reversed precordial leads and limb leads, demonstrating sinus rhythm. A screening ECG was also normal. The patient is classified as obesity class I with a Body Mass Index (BMI) of 32 kg/m^2. Vitals: Heart Rate (HR) 78 Beats Per Minute (BPM) (regular), Blood Pressure (BP) 130/80 mmHg, SpO_2 93-94% on room air.

Given the urgency of the situation and the need for surgical intervention, combined spinal-epidural anaesthesia with ultrasound assistance was planned to address the patient's complex respiratory profile. Nebulisation was administered before transfer to the operating room.

In the operating theatre, wide-bore Intravenous (IV) access was established and standard monitoring (ECG, noninvasive blood pressure, and pulse oximetry) was applied. The patient was positioned sitting after receiving an ultrasound-guided femoral nerve block with 15 mL of 0.25% bupivacaine. Under ultrasound guidance, the L2-L3 and L3-L4 interspaces were precisely identified, and epidural and spinal anaesthesia were administered, respectively. A subarachnoid block was achieved with 3 mL of 0.5% bupivacaine. The four-hour surgery proceeded with the patient remaining haemodynamically stable throughout. He was discharged on postoperative day 7. In the postoperative period, epidural top-ups were provided per our institution's Acute Pain Service (APS) protocol to ensure effective pain management.

DISCUSSION

Primary Ciliary Dyskinesia (PCD) comprises a group of autosomal recessive disorders. Among these, KS is the most prevalent subtype [1]. KS is characterised by bronchiectasis, chronic recurrent rhinosinusitis, and situs inversus. The pathophysiological basis is linked to dysfunction in the dynein arms within the ciliary axoneme, which compromises the coordinated movement of epithelial cilia and disrupts mucociliary activity. This dysfunction leads to impaired mucociliary clearance, resulting in mucus retention, ineffective clearance of pathogenic organisms, and

subsequent chronic respiratory infections, which may ultimately culminate in bronchiectasis [2].

Anesthetic considerations for individuals with KS are predominantly centred on the respiratory system. Factors such as preoperative respiratory tract infections and patterns of obstructive pulmonary function contribute to increased morbidity [3] in these patients. The complications of general anaesthesia include atelectasis, suppression of the cough reflex, increased airway secretions, and aspiration, resulting in perioperative respiratory tract infections [3]. Considering the risk-benefit balance, we preferred regional anaesthesia for our patient. In this case, employing a combined spinal–epidural anaesthesia technique facilitated effective pain management and haemodynamic stability while minimising the respiratory complications typically associated with general anaesthesia [4]. Ultrasound guidance proved advantageous in overcoming anatomical challenges due to the patient's obesity, enabling precise identification of the midline, vertebral level, intervertebral space, and anticipated depth to both epidural and intrathecal spaces. Ultrasound imaging also informed optimal angle and approach decisions for successful block placement [5]. Compared to traditional landmark techniques, ultrasound guidance reduced the number of needle passes and skin punctures required. The spinal level was maintained below T10 to preserve pulmonary function. Integrating epidural analgesia in the postoperative care plan can mitigate pulmonary complications by providing superior analgesia and facilitating early mobilisation. It is crucial to emphasise aseptic techniques during regional anaesthesia to address the abnormal neutrophil chemotaxis observed in these patients [5]. Reidy et al., have documented the incidence of postoperative respiratory tract infections following general anaesthesia for diagnostic laparoscopy in a patient with KS [6]. It is imperative to consider complications such as accidental endobronchial intubation and pulmonary aspiration when planning for general anaesthesia in this patient population.

In the context of dextrocardia, typically associated with situs inversus, no additional cardiac anomalies are usually present. Anesthetic considerations for this condition include adjustments for lead placement during ECG monitoring, paddle positioning for external defibrillation, and facilitation of left-sided central venous cannulation [1]. Patients with KS undergoing elective surgical procedures should be thoroughly optimised through intensive physiotherapy, supportive pulmonary care, and prophylactic antibiotic administration to prevent the progression of respiratory tract infections. Given that our case involved an emergency procedure, the patient was managed with nebulisation as a precautionary measure prior to surgery.

CONCLUSION(S)

In patients with KS undergoing emergency orthopaedic surgery, meticulous anaesthetic planning and tailored interventions are essential for achieving successful outcomes. The primary advantage of employing regional anaesthesia in these cases lies in its ability to preserve respiratory muscle function both intraoperatively and postoperatively, thereby enabling the patient to effectively clear airway secretions. Additionally, regional anaesthesia provides superior analgesia, facilitating early mobilisation during the postoperative period. Preprocedural ultrasound imaging of the spine considerably reduces the technical challenges associated with central neuraxial blockade, requiring fewer needle passes and skin punctures. Cardiopulmonary complications can be largely mitigated through appropriate perioperative suctioning of pharyngeal and pulmonary tract secretions.

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