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Case Report

High-flow nasal cannula and Kartagener's syndrome in a patient with in vitro fertilization conception

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Abstract

Kartagener's syndrome is a rare autosomal recessive disorder characterized by a triad of situs inversus, chronic sinusitis, and bronchiectasis. It is typically diagnosed during childhood and is caused by a defect in the ultrastructure of the microtubules in epithelial ciliated cells, which leads to impaired ciliary function. This impairment results in recurrent bronchitis, sinusitis, and bronchiectasis. A significant concern associated with this syndrome is the increased susceptibility to infections, particularly of the pulmonary system during anesthesia. We present the anesthetic management of a 43-year-old primigravida who conceived via in vitro fertilization and was currently 29 weeks pregnant with twins. She was under regular follow-up for known Kartagener's syndrome and presented for an emergency lower segment cesarean section.

Keywords: HFNC, Kartagener syndrome, Subarachnoid block.

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1. Introduction

Kartagener's syndrome (KS) is a subset of primary ciliary dyskinesia. Kartagener's syndrome is a rare, inherited disorder characterized by a triad of medical conditions of bronchiectasis, chronic sinusitis, and situs inversus. **Figure 1**). In 1904, Siewart first described this condition, but it was Kartagener who, in 1933, reported four cases and established the etiological correlation within the triad.[1,2] The estimated prevalence is 1 in 30,000 live births.[3] In KS, there is ultrastructural impairment of ciliary motility, leading to recurrent infections of the ear, nose, throat, and pulmonary system.[4] Early diagnosis and prompt treatment are required to preserve pulmonary function and improve the quality of life and life expectancy for patients with this disease.

Figure 1: Manifestations of Kartagener's syndrome

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2. Case Report

A 43-year-old primigravida with a twin gestation of 29 weeks, conceived via in vitro fertilization (IVF), presented to the outpatient department with bilateral limb edema lasting two days. She was a known case of KS under regular pulmonology follow-up since childhood. Her medication regimen included a metered-dose inhaler of Forglyn (glycopyrrolate and formoterol fumarate), tablet Acebrophylline 100 mg twice daily (BD), and syrup N-acetylcysteine (NAC) during acute episodes. She had experienced recurrent lower respiratory tract infections since childhood, leading to multiple hospitalizations. Additionally, her brother had a similar medical history and was also infertile. The patient faced challenges with infertility, including multiple failed intrauterine inseminations (IUI) and IVF attempts. Recently, she was diagnosed with gestational hypertension and was prescribed tablet Labetalol 100 mg BD; she was also known to have hypothyroidism and was on tablet Eltroxin 25µg. Furthermore, she was on tablet Aspirin 75 mg and injection Enoxaparin 40 mg subcutaneously once daily. Throughout her pregnancy, she received both prophylactic and therapeutic antibiotics. The twin pregnancy was complicated by reversed diastolic flow and gestational hypertension (160/90 mmHg) at 29 weeks, necessitating an emergency cesarean section (C-sec).

On examination, she was 165 cm tall and weighed 75 kg. She was conscious and oriented but exhibited dyspnea at rest, with a respiratory rate of 32-36 per minute. Bilateral air entry was present, accompanied by coarse basal crackles and wheezing throughout the lung fields. Her blood pressure was 160/90 mmHg. The patient's oxygen saturation on room air was 90%, which improved to 95% with supplemental oxygen at 5 L/minute. Laboratory tests were within the normal range. Echocardiography showed situs inversus with dextrocardia and good biventricular function.

The patient was counseled for a subarachnoid block (SAB), taking into account the risk-benefit ratio and the primary concern of avoiding pulmonary system involvement. Antacid prophylaxis with intravenous (IV) pantoprazole 40 mg and metoclopramide 10 mg was given. Blood grouping and crossmatching were done. She was positioned on the operating table with 30º head-end elevation and connected to a high-flow nasal cannula (HFNC) delivering 50% FiO2 at 37°C. To reduce the risk of respiratory complications, IV Deriphylline 110 mg was given prophylactically, and her routine medications were kept ready.

Standard ASA monitors were applied, with ECG chest leads placed on the right side and invasive blood pressure monitoring initiated. The patient was preloaded with 250 ml of Ringer's lactate solution and positioned in the left lateral position for SAB. At the L3-L4 interspace, a 26-gauge Quincke needle was introduced, and a total of 1.8 ml of 0.5% heavy bupivacaine was injected intrathecally. Surgery commenced after confirming the dermatomal level at T6. A lower segment cesarean section was performed, resulting in the delivery of a female baby weighing 890 grams with APGAR scores of 7/10 and 8/10 at one and five minutes respectively, while the other baby was delivered in utero demise. An IV infusion of 10 IU of oxytocin and an additional 10 IU intramuscularly were administered immediately after delivery. IV tranexamic acid 1g and cefotaxime 1g were also given. The patient's blood pressure dropped from 160/90 mmHg to 120/80 mmHg. A maintenance fluid of 750 ml was administered cautiously due to pre-existing lung congestion, with an estimated blood loss of around 400 ml and a urine output of 600 ml, ensuring a negative fluid balance. Any drop in blood pressure was managed with inotropes. The patient experienced dyspnea and desaturation during delivery, which improved with administration of her metered-dose inhaler (Forglyn) and nebulized salbutamol, while slight head elevation was maintained throughout the procedure.

After surgery, the patient was transferred to the Surgical Intensive Care Unit (SICU). HFNC was continued, then tapered and weaned off gradually. Nebulization with bronchodilators and NAC was advised every two hours. Chest physiotherapy was given to enhance respiratory function. Adequate postoperative pain relief was provided. The postoperative period was uneventful.

3. Discussion

The characteristic triad of KS consists of bronchiectasis, sinusitis, and dextrocardia, typically identified during childhood. Additional features may include infertility in both males and females, recurrent respiratory infections, and either situs inversus totalis or solitus. [5] The absence of ciliary motility or presence of dyskinesia leads to recurrent respiratory infections, which are the primary concern in this condition.[5] There are limited case reports addressing anesthesia management for KS. A critical consideration is minimizing pulmonary complications, as these patients face an increased risk of postoperative respiratory infections after general anesthesia.[6,7] Preoperative optimization is essential for any planned elective procedures or in emergency situations, as demonstrated in our case. Precautions to prevent perioperative infections should include the administration of peri-operative antibiotics, provision of adequate analgesia, implementation of postoperative chest physiotherapy, postural drainage, and early mobilization.

In this case, the patient experienced multiple unsuccessful IUIs and IVFs over a span of 15 years of infertility. Ultimately, she achieved a successful twin pregnancy through IVF. However, she presented with a reversal of diastolic flow, necessitating an emergency C-sec.[8,9] After evaluating the risk-benefit ratio, we opted to perform the C-sec under SAB. [6,7] To mitigate any potential decline in respiratory function, prophylactic administration of theophylline was given, and her regular medications were

kept ready. The patient was positioned on the operating table with a slight incline, and an HFNC was utilized.[10] HFNC delivers heated, humidified air, improving mucociliary clearance, alveolar recruitment, and work of breathing, making it ideal for KS patients.[11,12]

Patients with KS are at a heightened risk of respiratory infection; therefore, strict aseptic protocols were adhered to during all invasive procedures.[5] IV antibiotics were administered prior to the incision. We ensured stable hemodynamics through volume replacement and inotropes, maintaining a negative fluid balance throughout the procedure. Manipulating the airway in patients with KS increases the risk of infections, which can elevate morbidity and mortality.

4. Conclusion

Patients with Kartagener's syndrome can be successfully managed using a subarachnoid block and a high-flow nasal cannula, provided adequate preoperative optimization is performed. Adequate postoperative analgesia, early mobilization, and chest physiotherapy help in reducing the risk of peri-operative infection.

5. Declaration of Patient Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

6. Source of Funding

None.

7. Conflict of Interest

None.

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