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5AP1-4

General anesthesia in a 9-year-old child with Kartagener syndrome: a case report

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Background: Kartagener syndrome is a subset of primary ciliary dyskinesia, a rare autosomal recessive condition in which ciliary structure and/or function is abnormal; it is characterized by the clinical triad of situs inversus, chronic sinusitis and bronchiectasis. Clinical manifestations are more severe during the first decade of life and include chronic upper and lower respiratory tract disease due to impaired mucociliary clearance. Recurrent otitis media is a common manifestation. Even though surgical treatment is often needed, this syndrome is rarely encountered in paediatric anaesthesia practice.
Case report: A 9-year-old male child with history of situs inversus with dextrocardia and hypoplasia of paranasal sinuses was scheduled for adenoidectomy and bilateral myringotomy tube placement due to chronic adenoiditis, sleep-disordered breathing and chronic otitis media. He presented frequent episodes of breathlessness and mucopurulent productive cough often needing postural drainage associated with antibiotic and bronchodilator therapy. Pre-operatively, postural drainage and chest physiotherapy were performed. Antibiotics and steroids were administred before induction. Anesthesia was induced with midazolam, propofol and alfentanyl and maintained with sevoflurane. The patient was ventilated with a humidified mixture of oxygen and air in the pressure-controlled mode. Intraoperatively, despite remaining hemodynamically stable, gentle endotracheal tube suction due do accumulated secretions with associated decrease of achieved tidal volumes was needed on three occasions. Bronchodilators were administered prior to deep extubation but mild bronchospasm still followed, requiring additional bronchodilators and assisted mask ventilation. Administration of supplemental oxygen and epinephrine nebulization were maintained during the immediate post-operative period, which was otherwise uneventful.
Discussion: Patients with Kartagener syndrome are prone to airway obstruction, air trapping and hypoxemia. If airway management is required, there may be an increased risk of complications such as bronchospasm or acute respiratory failure. Careful clinical evaluation and pre-operative measures such as chest physiotherapy and administration of steroids and bronchodilators are crucial.

5AP1-5

Anesthetic management of a very rare disease: HSAN type IV

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Background: Hereditary Sensorial Autonomic Neuropathy Type IV (HSAN-IV) a.k.a. Congenital Insensitivity to Pain with Anhidrosis (CIPA) is a rare congenital disease characterized by recurrent unexplained fever, total insensitivity to pain, anhidrosis, and mental retardation in some cases. These patients occasionally develop corneal scarring, teeth eruptions, multiple fractures, osteomyelitis, buccal and lower limb wounds, and injuries of finger tips caused by self-mutilating behaviors; so they often undergo surgery.[1] We report our experience with HSAN-IV patient developed tibia fracture.
Case report: A 7 year old-male patient admitted to hospital with left tibia fracture. He had clinical diagnosis of HSAN-IV; history of ulna fracture, minor burns on the finger tips, and circumcision without anesthesia. The patient was scheduled for elective operation under monitored anesthesia care (MAC). Routine monitorization was made, including electrocardiogram, pulse-oximeter, non-invasive blood pressure and body temperature measurement in the operating room. Closed reduction and external fixation of tibia was performed without any complications. The patient was discharged after one night uneventful follow-up.
Discussion: Patients with CIPA have insensitivity to pain, but MAC can sometimes yield problems. Tactile stimulus can cause hyperesthesia and unpleasant feeling during surgery. Patients with CIPA are prone to regurgitation and cardiac dysrhythmias. Appropriate airway control must be provided. Body temperature monitorization is vital, because the regulation is impaired due to anhidrosis. We suggest routine application of general anesthesia to these diazepines and inhalation agents have been safely used in CIPA.[2] In conclusion, although patients with CIPA have insensitivity to pain, general anesthesia is suggested in them for operations, and body temperature monitoring must be a part of routine monitorization.
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1. E. Achouri, M. Gribaa, J. Bouguila et al. HSAN type IV: A report on two cases Archives de Pe'diatrie 2011;18:390-3.
2. Sheng-Chin Kao, Chien-Kun Ting, et al. Desflurane Used in a Patient with CIPA during Septic Shock. J Chin Med Assoc 2004;67:305-7.
Learning Points: Important features of anesthesia for CIPA.

5AP1-6

Apparent mineralocorticoid excess (AME) syndrome - anaesthetic management of Omani siblings with hypertension and hypokalemia

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Background: Congenital deficiency of 11β-hydroxysteroid dehydrogenase type-2 enzyme, which normally inactivates cortisol and confers aldosterone specificity on the mineralocorticoid receptor, leads to Cortisol behaving as a potent mineralocorticoid, Hypertension and hypokalemia are thus grave risks for concomitant general anaesthesia.
Case report: Two Omani sisters aged 6 yrs (weight-13 kgs; BMI-6.7) and 10 yrs (weight- 20kgs; BMI-12.1) with Hypertensive Retinopathy were scheduled for Ophthalmological examination under General Anaesthesia. Initially presenting with severe failure to thrive, hypertension, hypokalemia (K-2.1) and polyuria-polydipsia, they also had progressive learning disability, easy fatigability, enuresis and bladder dysfunction. Cortisol level was normal but 24-hr urine cortisol/ cortisone ratio was raised 50-fold. Treated with potassium supplements, Ameloride and Atenolol. Anaesthesia assessment revealed normal airway and clear chest. Propofol and Fentanyl induction led to atropine-sensitive bradycardia. Maintenance was with Sevoflurane through LMA proseal. No intraoperative hypertension and smooth recovery.
Discussion: Hypokalemia and Hypertension are potential anaesthetic risks. Intraoperative cardiac arrhythmias, hypertensive cardiac failure/cerebrovascular accidents and delayed recovery due to poor muscle strength are realistic possibilities. It is imperative to optimize and also be vigilant intraoperatively, while managing patients with such rare syndrome.
This case report is to highlight the potential clinical catastrophe that are incipient in anaesthesia management of these children. Regional anaesthesia is also potentially harmful in view of contracted intravascular volume and altered autoregulation; with significant perfusion loss after sympatholysis. Active multi-disciplinary optimization in the peri-operative phase is essential to ensure smooth anaesthesia conduct.
References:
1. Al-Harbi T, Al-Shaikh AJ. Apparent mineralocorticoid excess syndrome: report of one family with three affected children. Pediatr Endocrinol Metab. 2012;25(11-12):1083-8.
2. Parvez Y, Sayed OE. Apparent mineralocorticoid excess (AME) syndrome. Indian Pediatr. 2013 Apr;50(4):416-8.
Learning Points: Hypertension and hypokalemia in children with rare congenital syndromes are important potential harbingers of Stroke and heart failure during anaesthetic management. Diagnosis and pro-active multi-disciplinary management are imperative for continued care.

5AP1-7

Anesthetic management of child with anterior mediastinal mass: a case report

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Background: General anesthesia in children with anterior mediastinal mass may be accompanied by serious life-treating complications, as a result of