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CASE REPORT

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Anesthetic management of diphallia with anorectal malformation posted for colostomy: a rare association

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Abstract

Background Duplication of the penis or Diphallia is a rare abnormality of the urogenital tract present once in every 5–6 million live births with varied presentations and associated systemic anomalies.

Case presentation We present the anesthetic management of a neonate presenting with duplication of penis, urethra, and anorectal malformation.

Conclusions With this rare case, we aim to shed light on the various perioperative anesthetic concerns of such neonates presenting with surgical emergencies and their successful management.

Keywords Anesthesia, Anomalies, Diphallia, Neonates

Background

Diphallia or duplication of the penis is a rare entity occurring once in every 5–6 million/live births. It may be associated with genitourinary, gastrointestinal, neuromuscular, vertebral, and sometimes cardiac abnormalities (Gyftopoulos et al. 2002). There are two schools of thought regarding its origins—polygenic and/or environmental causing an embryological defect in urethral folds and genital tubercles during 23rd to 25th days of gestation (Habib et al. 2023).

Case presentation

Here, we present the anesthetic management of a 2.5 kg, 1-day-old neonate with high anorectal malformation (ARM) posted for colostomy in October, 2020. The neonate had an oxygen saturation of 98% on room air, respiratory rate of 40/min and bilateral equal air entry on auscultation with a heart rate of 160/min, loud S1, and no murmurs. On examination, a distended abdomen, penile duplication, pigmented anal dimples, and a sacrococcygeal mass were found as shown in Figs. 1 and 2, respectively. Invertogram revealed distended bowel loops and pubic symphysis diasthesis. Ultrasound examination confirmed the presence of true diphallia along with two distinct urethra and a single bladder. Family and antenatal history were insignificant. Baseline investigations (complete blood count, renal function tests, liver function tests) were within normal limits. The American Society of Anaesthesiologists (ASA) fasting guidelines were followed.

The neonate was shifted to the prewarmed operating room after obtaining informed consent. Standard monitoring devices—oxygen saturation and temperature probes, electrocardiogram (ECG), noninvasive blood pressure (NIBP), and end-tidal carbon dioxide (e.g., CO2), were attached, and baseline vitals were recorded and monitored continuously. Premedication with glycopyrrolate 0.04 mcg/kg intravenous (IV) and fentanyl 1 mcg/kg (IV) was given via an already secured 26-gauge IV cannula. Following preoxygenation, intravenous (IV) induction was done using propofol 2.5 mg/kg and

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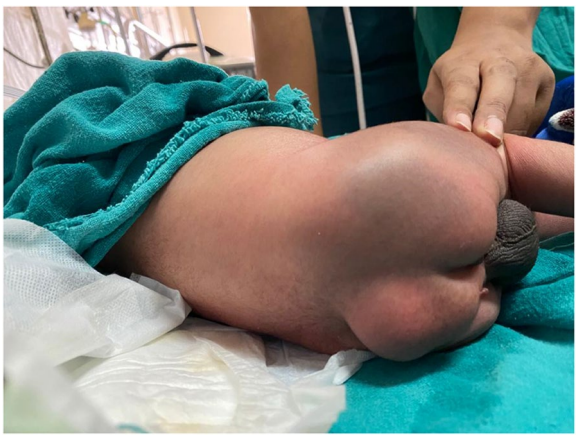

Fig. 1 Sacrococcygeal mass

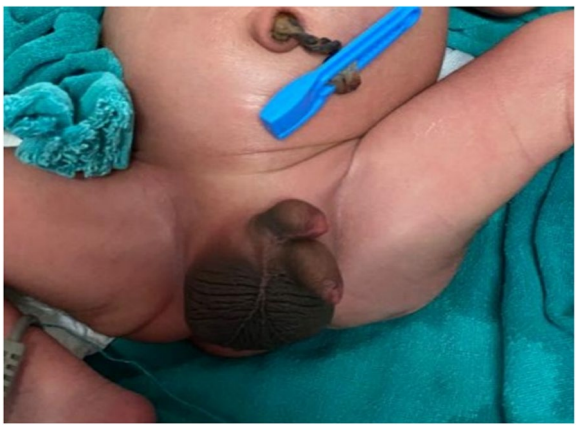

Fig. 2 Duplication of penis

atracurium 0.5 mg/kg, and the child was intubated after 3 min of gentle bag mask ventilation with a 3.0-mm uncuffed endotracheal tube under direct laryngoscopy using Miller's Blade 0. Anesthesia was maintained using a mixture of oxygen + air + sevoflurane at 0.75 minimum alveolar concentration (MAC). Paracetamol 10 mg/kg IV was given at end of surgery for postoperative analgesia. Lactated Ringer's solution (10 ml/kg) with 1% dextrose was infused via burette set. The neonate remained hemodynamically stable. Following surgery (colostomy performed 24 h after birth), he was reversed with neostigmine 0.05 mg/kg IV and glycopyrrolate 10 mcg/kg IV and extubated and shifted to the nursery on 99% saturation.

Discussion

Diphallia has an epigenetic mechanism involving seven genes and fetal exposure to toxins, stress, or infections (Frolov et al. 2018). Due to the paucity of literature and its unique presentations, anesthetic management is tailored according to the surgery and presenting features. Associated VACTERL (vertebral defects, anal atresia, cardiac defects, tracheoesophageal fistula, renal defects, limb hypoplasia) anomalies should be assessed for. Unanticipated difficult intubation can occur due to anteriorly placed larynx with/or subglottic stenosis in neonates. Increased chances of aspiration and impaired ventilation due to distended abdomen are a possibility in ARM (Harless et al. 2014). Appropriate fluid administration is essential. Avoidance of hypoglycemia, hypovolemia, hypothermia, and hypoxia is important. Hypothermia can be prevented by using warm IV and irrigating fluids, wrapping head and limbs, and maintaining optimum ambient temperature. Careful positioning and padding below joints are done in case of vertebral defects. Thorough cardiovascular system evaluation is necessary to prevent hemodynamic instability. Appropriate pain management and timely extubation aid in early discharge (Pani and Panda 2012).

Conclusions

Rare surgical cases present new learning opportunities for the anesthesiologist as well. Detailed preoperative assessment followed by adequate perioperative anesthesia and analgesia tailored according to the patient needs aid in successful recovery.

Abbreviations

ARM	Anorectal malformation
IV	Intravenous

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Authors' contributions

(1) AS, writing manuscript, analysis and interpretation of patient data, research, and compilation of data and facts; (2) ND, proofreading and correction of manuscript; (3) RC, data rechecking and analysis; (4) PR, proofreading and correction of manuscript; and (5) HM, data collection and interpretation. All authors have read and approved the final manuscript. All authors give consent to publish.

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Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

Taken.

Competing interests

The authors declare that they have no competing interests.

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