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Case Report Open Access

Anorectal Malformation without Fistula in Female Neonate and Colonic Atresia: A Rare Association

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Abstract

The combination of colonic atresia and anorectal malformations is uncommon. The etiopathogeny of this association was obscure with an unknown frequency. Few cases were reported worldwide since the first description. We report a sporadic occurrence of this association in a 3-day Ivorian female newborn which to our knowledge has not been previously reported in sub-Saharan Africa.

Keywords: Anorectal malformations; Colonic atresia; Female newborn

Introduction

Anorectal Malformations (ARM) are relatively rare condition with an estimated incidence of 1/4000-5000 live births. This anomaly may be isolated or part of a poly malformation syndrome [1]. Colonic atresia (CA) is one of the rarest causes of neonatal intestinal obstructions [2]. It represents 1.8% to 15% of all intestinal atresia with an incidence of 1/40000 live births [3]. The combination of CA and ARM in female neonate is uncommon. Worldwide, coexistence of these anomalies in a single patient was first reported in 1959 by Trusler, et al. [4]. We have no knowledge so far of such report from Ivory Coast. We are hereby reporting a rare case of this association in a female neonate.

Case Report

A 3-day full-term baby female suffering from emesis and failure to pass stool since birth, was transferred to the pediatric surgery department at Yopougon Teaching Hospital. There is no history of drug administration or exposure of any known teratogenic agents during pregnancy. The newborn was born by vaginal delivery at term. At birth the female weighed 2.850 kg and measured 49 cm. Her head circumference was 32 cm, and APGAR score was 7 and 8 at 1 and 5 min respectively. There was no issuance of meconium. At admission, the baby weighed 2.9 kg. Her skin was pink. She was febrile (T=38°C) and tachypnea with a respiratory rate of 70 c/min. The heart rate was 170 b/min. Clinical examination revealed a grossly distended abdomen with peritonitis signs. An imperforate anus with an obvious anal dimple, with poorly developed gluteal muscle and presence of two perineal ports: urethral and vaginal without visible fistula. She had normal female external genitalia. No other physical abnormalities were noted. Abdominal X-rays shows extensive bowel dilatation (Figure 1). The transverse laparotomy exploration revealed intact gastrointestinal system from proximal stomach to the ascending colon. However, distal to the transverse colon existed a blind-ending colon with a massive dilatation of proximal colon followed by multiple CAs. There was no rectum and no visible fistula. There was no evidence of other abnormality (Figure 2). Transverse colostomy uni-tubular was performed. Renal and cardiac ultrasound found no associated anomalies. Rachis X-rays was normal. Post-operative recovery was uneventful. She was discharged home in good conditions. Definitive surgery will be performed in the next step.

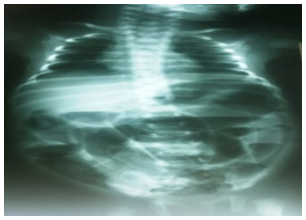

Figure 1: Abdominal X-rays shows extensive bowel dilatation.

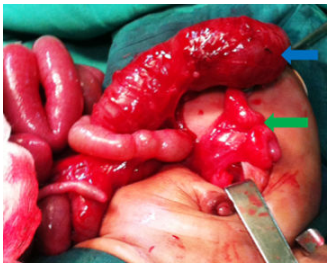

Figure 2: Blind-ending distending transverse colon (blue arrow), multiple atresia (green arrow).

Discussion

Since the first description, few cases have been reported in the literature [5-8]. The aetiology of both conditions coexisting remains obscure with unknown frequency. No risk factors have been identified to be responsible for the simultaneous occurrence of ARM and CA.

There are postulates implicating genetic, familial and environmental factors [6].

Diagnosis of CA sometimes poses a special diagnostic problem. In some cases, it must be suspected when mucus and no meconium are encountered during initial anorectoplasty after opening the rectum, and failed to pass meconium at post-operative time [7].

Alimentary tracts anomalies associated to ARM have been known to be relatively less common than associated anomalies of another organ system. [9].

The occurrence of ARM without fistula is a rare entity. This anatomic presentation appears to be uncommon in female [10]. In our case, it was a female neonate with ARM without fistula (imperforate anus) in which it was found to intraoperative colonic Grosfeld's atresia type IV [11]. The gap was significant and concerned the descending colon below the splenic flexure. This case underscores the importance of a high index suspicion for CA in case of ARM without fistula in female.

In emergency, we performed at first time a colostomy to avoid neonatal death. Delaying delicate surgical and anaesthetic procedure in a newborn baby is particularly advantageous in our environment where neonatal anaesthesia, parental nutrition, and neonatal intensive care unit are unavailable.

Delayed repair will consist of blind-ending colon pull-through. Combination of these anomalies requires multi-stage operations in particular and extensive medical management to establish bowel continuity, optimal intestinal absorption and unimpaired defecation.

Conclusion

The coexistence of anorectal malformation and colonic atresia is rare. We therefore recommend that patients with anorectal malformation without perineal fistula, especially in female, should be evaluated for colonic atresia at first operation time.

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