Case report

Spontaneous enteroscrotal fistula following an incarcerated inguinal hernia in a neonate: Case report and literature review

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ABSTRACT

Introduction: In Africa, frequent delays in seeking medical attention due to cultural considerations and lack of financial support could explain some particular clinical presentations. An enteroscopy fistula following an incarcerated inguinal hernia is a rare condition in neonates.

Presentation of case: A 21-day-old infant was brought to our consultation for persistent stool discharge from the right scrotum for three days. The parents reported an inguinoscrotal swelling that had been evolving for ten days before admission and became painful two days later. He was diagnosed with a case of enteroscopy fistula. After correction of electrolytes disorders, a laparotomy was performed through a right lower transverse incision. Operative findings were strangulated inguinal hernia with ileal perforation. Resection and end-to-end ileo-ileal anastomosis was performed. He was discharged from the hospital on the 6th post-operative day. No recurrence within the first year of follow-up.

Discussion: Thirteen cases have been reported in the literature until now. Most of them were from developing countries, including ours.

Conclusion: Early seeking medical attention of incarcerated inguinal hernias is necessary to reduce their morbidity and mortality. Paediatricians should emphasize providing adequate information, education and communication during routine examinations of neonates. We advocate the popularization of universal health insurance to facilitate health care.

1. Introduction

Inguinal hernia is a common surgical disease in children and incarceration is a well-known complication. The care of children's surgical diseases in Africa presents multiple challenges and is complicated by the large number of sick children, late presentation, and, often, advanced pathology at presentation. The family's attitude towards the management of surgical diseases is modified by tradition and culture that is peculiar to the community from which the child is coming [1]. This situation could explain certain dramatic and exceptional conditions like enteroscopy fistula following an incarcerated inguinal hernia. We report a case of a 21-day-full-term boy successfully managed in our unit and review the literature. This case report was presented using the SCARE 2020 guidelines [2].

2. Patient information

A 21 day-old male was brought from a village to our consultation, with a chief complaint of persistent discharging faeces from the scrotum and swelling at the right groin for three days. His parents noticed an initially reducible right inguinoscrotal swelling on day of life 11. Then, it became irreducible on day of life 13, with inflammation and pain. It has been mistakenly considered as an abscess, and they applied on an ointment twice a day for five days (till day of life 18). A short period of vomiting and mild fever was reported. No abdominal distension. The child continued to feed and pass stool. On day of life 18, the swelling had

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spontaneously fistulated through the scrotum, discharging faeces.

Medical history revealed an uneventful delivery, a full-term boy with a birth weight of 2.680 kg.

2.1. Clinical findings

At presentation, the cry, tone, and activity were normal. Vital signs were normal. The abdomen was flat. No evidence of intestinal obstruction.

On local examination, there was a large wound in the right hemiscrotum discharging faeces (Fig. 1).

2.2. Diagnosis assessment

He was diagnosed with a case of enteroscrotal fistula. The left inguino-scrotal area was normal. Blood investigations were within normal limits except Hb: 10.2 g/dL and Na: 131 mmol/L. Any imaging investigations were carried out.

2.3. Therapeutic intervention

After correction of electrolytes disorders, a laparotomy was performed through a right lower transverse incision.

Operative findings were strangulated inguinal hernia with ileal perforation leading to enteroscrotal fistula (Fig. 2). Resection and end-to-end ileo-ileal anastomosis was performed and the hernia was repaired from internally the abdomen with purse-string sutures. No drain was left in place. The homolateral testis was atrophic. Antibiotics (Ceftriaxone, Metronidazole) were administrated for five days.

2.4. Follow-up and outcome

Postoperative course was uneventful and the neonate was discharged from the hospital on the 6th post-operative day. The hernia did not recur till 12th months but the neonate was then lost to follow-up.

3. Discussion

The main complication of inguinal hernia is incarceration, which may result in bowel obstruction. It concerns around 12% of infants and young children. The incidence increases to 30% in children less than one year of age and 60% in the first six months [3]. Reduction can take place in up to two-thirds of cases after hospitalisation, leaving only one-third with a need for emergency surgery [4]. Within these one-third, spontaneous enteroscrotal fistula is an uncommon complication [5] but possible in developing countries due to delay in seeking medical attention, cultural considerations burden, financial constraints. In our case, parental ignorance and lack of resources were found. The parents were not very alarmed hence took him to a non-emergency consultation.

To the author’s knowledge, only thirteen previous cases have been reported in English medical literature [5–16]. Most of these cases published were from developing countries such as Nigeria [6–8], India [5,9,10–13] and Pakistan [14]. In developed countries, reported cases had particular features: extremely low birth weight premature neonate [15] and in Greece [16], a mild one. All the previous cases are summarized in Table 1.

In our reported case, the neonate's first symptom was swelling of the groin and faeces discharging from the right scrotum.
| Series No | Country | Year of publication | Case particularity | Duration (days) | Side of involvement | Surgical approach | Imaging investigation | Ipsilateral testis | Operation done | Complication if any | Outcome |
|-----------|---------|---------------------|-------------------|----------------|-------------------|------------------|---------------------|-------------------|----------------|----------------------|---------|
| Case 1 [5] | India  | 1993               |                  | 20             | Right             | Inguinal          | –                   | Gangrenous orchidectomy | R/A* of ileum  | –                  | Survived             |
| Case 2 [9] | India  | 1997               | Intussusception  | 20             | Right             | Inguinal          | –                   | Gangrenous orchidectomy | R/A* of ileum  | –                  | Survived             |
| Case 3 [10] | India  | 1999               |                  | 08             | Left              | Inguinoscrotal    | Plain abdominal radiograph | Normal          | R/A* of ileum  | –                  | Survived             |
| Case 4 [6] | Nigeria| 2002               |                  | 07             | Right             | Inguinal          | –                   | Normal            | R/A* of ileum  | Scrotal wound infection | Survived |
| Case 5 [6] | Nigeria| 2002               | Richter          | 43             | Right             | Inguinal          | –                   | Normal            | R/A* of ileum  | –                  | Survived             |
| Case 6 [7] | Nigeria| 2004               |                  | 07             | Right             | Inguinal          | Fistulogram        | Normal            | R/A* of ileum  | –                  | Survived             |
| Case 7 [8] | Nigeria| 2006               |                  | 10             | Right             | Inguinal          | –                   | Normal            | R/A* of ileum  | Anastomotic leak     | Survived             |
| Case 8 [11]| India  | 2007               | Meckel Diverticulum | 07        | Right             | Inguinal          | Plain abdominal radiograph | Normal          | R/A* of ileum  | Pneumonia, septicaemia | Died after surgery |
| Case 9 [12]| India  | 2009               |                  | 28             | Left              | –                 | –                   | –                | R/A* of ileum  | Severe sepsis        | Died prior to surgery |
| Case 10 [14]| Pakistan | 2010             |                  | 25             | Right             | Abdominal         | –                   | Non indicated   | R/A* of ileum  | –                  | Survived             |
| Case 11 [16]| Greece | 2014               | Amyand           | 05             | Right             | Inguinoscrotal    | Plain abdominal radiograph | Normal          | R/A* of ileum  | Severe sepsis        | Survived             |
| Case 12 [13]| India  | 2017               |                  | 11             | Right             | Inguinoscrotal    | –                   | Normal            | R/A* of ileum  | –                  | Survived             |
| Case 13 [15]| USA    | 2017               | Richter          | 02             | Right             | Abdominal         | Plain abdominal radiograph | Unknown        | R/A* of ileum  | Left chylothorax, severe respiratory disorders | Died after surgery (day 46 post-op) |
| Case 14 Present case | Benin  | 2021               | Richter          | 07             | Right             | Abdominal         | –                   | Normal            | R/A* of ileum  | –                  | Survived             |

Foot notes: R/A* – Resection and anastomosis, herniotomy and scrotal debridement.
gript followed by discharging faeces. Any features of intestinal obstruction were noticed, suggesting a Richter’s type hernia in which the lumen is not obstructed.

The right side was often involved [5–9,11,13–16] and only two authors reported left one [10,12]. Inguinal hernias are thought to be slightly common on the right side. Enteroscrotal fistula diagnosis is mainly clinical hence requires no investigations; even though around 50% of authors completed abdominal x-ray [7,10,11,12,15,16]. It showed gaseous shadow in the scrotum but didn’t modify the indication. Babygram could be optional as a meticulous examination was performed at admission.

However, resuscitation using intravenous crystalloid fluid, broad-spectrum antibiotics, and sometimes blood transfusion is obvious. It allows better general health prior to surgery and may decrease morbidity. In our case, no blood transfusion was required.

Various types of surgical approaches have been reported: inguinal, inguinoscrotal and lower abdominal [6,15,16]. All of them offer good exposure and facilitates necrotic tissues excision. Compression of the spermatic vessels may compromise the viability of the ipsilateral testis. Further progression may be testis gangrene so that orchidectomy could be inevitable [5,9]. The other complications ranged from wound infection to severe sepsis leading and death in some cases [11,12,15]. Only one author noted a death before surgery [12] showing the emergency degree of this condition.

4. Conclusion

Prompt and early seeking medical attention of incarcerated inguinal hernias is necessary to reduce their morbidity and mortality. Moreover, paediatricians should put the emphasis on information, education and communication during routine examinations of neonates. Finally, in countries with limited resources, we advocate popularization of universal health insurance in order to facilitate health care.

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Ethical approval

Ethical approval has been exempted by my institution for reporting this case.

Consent

Clinical figures were taken in this manuscript after parents informed consent obtained. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Guarantor

Dr. Amoussou Sedjro Clotaire Romeo Houegban is the guarantor for the work.

CRediT authorship contribution statement

Dr. Amoussou Sedjro Clotaire Romeo Houegban contributed to the concept and design of study, data collection, data interpretation and analysis, approval of final manuscript.

Data collection, revision, approval of final manuscript were done by Dr. Beaudelaire Romulus Assan and Dr. Medard Ayawo Guedennon.

Dr. Sourou Bruno Noukpouzounkou, Dr. Mahnakpon Vihtogbe Léon Samuel Boris Gogan and Dr. Monsoia Gildas Yassegoungbe were involved in approval of final manuscript.

Dr. Antoine Seraphin Gbenou supervised the work and were involved in approval of final manuscript.

Declaration of competing interest

Authors have no conflicts of interest to declare.

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