Case Report

Advocating for Routine Post-natal Surveillance and Urgent Neonatal Inguinal Hernia Repair

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Abstract

Inguinal hernias are more common in preterm and neonates and incidence of incarceration are reported to be more in the first 6 months of life. Strangulation follows incarceration and various incarcerated and strangulated contents having been reported in the sac. The fistulation of the herniated content through the scrotal skin is quite rare with only a few reported cases. We present the case of entero-scrotal fistula in a neonate managed with staged repair along with a brief review of the literature. A 27-days-old, full-term male presented with faecal discharge from the right scrotum. He had no tell-tale signs of obstructed hernia. A faecal fistulous opening was located in the right hemi-scrotum. The terminal ileum was seen as the content with an antimesenteric perforation, divided ileostomy and scrotal debridement was done and later ileo-ascending anastomosis was performed electively. Neonatal hernias should be considered an urgency and we advocate early surgery. The resource-limited setting and poor post-natal surveillance may have added to the worries. In our case, swelling and fistulation occurred in a very brief period of 36 h. We managed the child with a diversion stoma followed by ileo-ascending anastomosis later.

Keywords: Early diagnosis, parental education, post-natal care, quality of health care, scrotal fistula

Introduction

Neonatal hernias are more often seen in pre-term babies. They are usually at a higher risk of incarceration.[1] The incident of hernia in children is 0.8%–4.4%, but in premature infants, this increases to 16%–25%.[2] However, it is not uncommon to have a complicated hernia in term infants. We present the case of a 27-day, full-term male with a perforated inguinal hernia and an entero-scrotal faecal fistula. We have also tried to review and summarise available literature on this topic.

Case Report

A 27-day-old, full-term, malnourished male presented with faecal discharge from the right hemi-scrotum. The parents had first noticed some redness over the scrotum 36 h before reaching the hospital from a remote location. They gave no prior history of inguinal or scrotal swelling. On examination, child was haemodynamically stable, afebrile with no tell-tale signs of obstructive or incarcerated hernia. His abdomen was soft and he had been passing stools per rectum also. Two, 1 cm × 1 cm fistulous opening with hyperaemic borders and faecal discharge were noted from the right scrotum [Figure 1].

Considering a complicated inguinal hernia, exploratory laparotomy, via right lower quadrant transverse incision was planned [Figure 2]. It showed terminal ileum up to ileocaecal junction was herniating through the right deep ring with severe adhesions of the ileal loop with inguinal canal and scrotal sac. On reduction of the above loop, a perforation was noted at the antimesenteric border of the herniated loop, 5 cm from ileocaecal junction. The right testis was viable. As the caecum and terminal ileum appeared very friable, a divide ileostomy was fashioned and thorough lavage of the region was done. The peritoneum was closed with a purse string suture through the abdomen. The scrotal wound was repaired primarily after a thorough wash with warm saline [Figure 3].

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Table 1: Previously Reported cases of Scrotal Fistulation in Children

Author	Location	Age	Presenting symptom	General status	Irreducibility	Side	Associated anomaly
Vost, 1915[3]	India	12 years	Irreducible hernia + fistulation	Healthy, stable	2 years	Right	
Stock, 1951[4]	Hong Kong	2 months	Scrotal swelling with faecal discharge	Healthy, STABLE	10 days	Left	Left UDT, gut malrotation
Mishalany, 1963[5]	Lebanon	5 months	Chronic discharging faecal sinus	Healthy, stable	3 months	Right	None
Mishalany, 1963[5]	Lebanon	5 months	Acute discharging faecal sinus	Healthy, stable	2 weeks	Right	None
Gupta and Rohatgi, 1993[10]	India	20 days	Inguinoscrotal faecal fistula	N/A	N/A	Right	None
Rattan and Garg, 1998[11]	India	20 days	Intestine protruding out of scrotum	Abdominal distension, tachycardia	14 days	Right	None
Kasat *et al*., 2000[12]	India	60 days	Faecal discharge from scrotum	Healthy, stable	9 days	Left	None
Ameh *et al*., 2022[13]	Nigeria	29 days	Irreducible swelling with scrotal faecal discharge	Refusal to feed, abdominal distension	7 days	Right	None
Ameh *et al*., 2022[13]	Nigeria	43 days	Irreducible swelling with scrotal faecal discharge	Bilious vomiting + irritability	8 days	Right	None
Chirdan *et al*., 2004[14]	Nigeria	21 days	Faecal discharge from scrotum	Dehydrated + febrile	3 days	Right	None
Sowande *et al*., 2006[15]	Nigeria	28 days	Inguinoscrotal hernia with faecal fistula	Healthy, stable	10 days	Right	Umbilical hernia
Ghritlaharey *et al*., 2007[6]	India	30 days	Faecal discharge from scrotum	Abdominal distension, hypothermia, cyanosis	7 days	Right	None
Roshan Khan *et al*., 2009[9]	India	28 days	Faecal discharge from left scrotum	Septic shock	2 days	Left	None
Sharif *et al*., 2010[16]	Pakistan	25 days	Faecal discharge from scrotum	Abdominal distension, vomiting, tachycardia	2 days	Right	None
Ezomike *et al*., 2012[17]	Nigeria	49 days	Faecal discharge from scrotum	Healthy, stable	14 days	Right	Testicular atresia
Panagidis *et al*., 2015[18]	Greece	25 days	Faecal discharge from scrotum	Septic shock	5 days	Right	None
Raj *et al*., 2017[19]	India	30 days	Faecal discharge from scrotum	Healthy, stable	2 days	Right	None
Smith *et al*., 2018[20]	USA	14 days	Faecal discharge from scrotum	Stable abdominal findings	2 days	Right	Patent ductus arteriosus
Ugwu *et al*., 2020[8]	Nigeria	31 days	Faecal discharge from two fistulas over scrotum	Healthy, stable	3 days	Right	None
Houegban *et al*., 2022[21]	Benin	21 days	Faecal discharge and swelling of scrotum	Healthy, stable	6 days	Right	None
Gupta *et al*., 2022[7]	India	45 days	Faecal discharge from scrotum	Healthy, stable	Few days	Right	None

Author	Content	Imaging	Approach	Surgery done	Testis	Complication	Outcome
Vost, 1915[3]	Caecum	None	Inguinal	Caecum repair + Bassinis's hernia repair	Normal	None	Improved
Stock, 1951[4]	Caecum appendix	None	Inguinal	Caecal repair + hernioplasty + scrotal debridement	Normal	Incarcerated contralateral inguinal hernia	Improved
Mishalany, 1963[5]	Appendix	IVU + fistulogram	Inguinal	Appendix stump inversion + herniotomy + scrotal drainage	Normal	None	Improved
Mishalany, 1963[5]	Caecum and appendix	None	Inguinal	Appendix stump inversion + herniotomy + scrotal drainage	Normal	None	Improved
Gupta and Rohatgi, 1993[10]	N/A	None	N/A	N/A	N/A	None	
Rattan and Garg, 1998[11]	Ileum	None	Inguinal	R/A of ileum + scrotal debridement + orchidectomy	Gangrenous	None	Improved
Kasat *et al*., 2000[12]	Ileum	X-ray	Inguinal	R/A of ileum	Normal	None	Improved
Ameh *et al*., 2001[13]	Ileum	None	Inguinal	R/A of ileum + herniotomy	Normal	Necrotising fasciitis of scrotum	Improved
Ameh *et al*., 2001[13]	Ileal wall	None	Inguinal	Wedge resection and ileal repair + herniotomy	Normal	Necrotising fasciitis of scrotum	Improved
Chirdan *et al*., 2004[14]	Ileum	Contrast X-ray	Inguinal	R/A of ileum + herniotomy	Normal	None	Improved
Sowande *et al*., 2006[15]	Ileum	None	Inguinal	R/A of ileum + herniotomy	Normal	Anastomotic leak + peritonitis	Improved
Ghritlaharey *et al*., 2007[6]	Ileum + Meckel's diverticulum	X-ray	Laparotomy	Resection of diverticulum and anastomosis + herniotomy	Normal	Pneumonitis and death	Died

Contd...

Table 1: Contd...

Author	Content	Imaging	Approach	Surgery done	Testis	Complication	Outcome
Roshan Khan *et al*., 2009[9]	N/A	X-ray	N/A	N/A	N/A	Severe septicaemia + death	Died
Sharif *et al*., 2010[16]	Ileum	None	Laparotomy	R/A ileum + transabdominal hernia repair	Normal	None	Improved
Ezomike *et al*., 2011[17]	Ileum	None	Inguinal	R/A of ileum + herniotomy	Atretic	None	Improved
Panagidis *et al*., 2014[18]	Appendix	X-ray	Inguinal	Appendectomy + inguinal canal repair	Normal	None	Improved
Raj *et al*., 2017[19]	Ileum	None	Inguinal	R/A of ileum + herniotomy	Oedematous, viable	None	Improved
Smith *et al*., 2018[20]	Ileum	X-ray	Laparotomy	Resection + divided ileostomy + transabdominal hernia repair	Not checked	DIC, respiratory failure, hypovolemic shock	Died
Ugwu *et al*., 2020[8]	Ileum	None	Inguinal	R/A of ileum + herniotomy + scrotal debridement	Normal	None	Improved
Hougeban *et al*., 2022[21]	Ileum	None	Laparotomy	R/A of ileum + transabdominal hernia repair	Atrophic	None	Improved
Gupta *et al*., 2022[7]	Long Meckel's diverticulum	None	Laparotomy	Diverticulectomy + R/A of ileal anastomosis + trans-abdominal hernia repair	Normal	None	Improved

N/A: Not available, UDT: Undescended Testis, IVU: Intravenous Urogram, R/A: Resection and Anastomosis, DIC: Disseminated Intravascular Coagulation

Figure 1: At the presentation in emergency

Figure 2: After cleaning under anaesthesia

Figure 3: Post-procedure

The post-operative course was uneventful and the patient was discharged on the 10th post-operative day. He was doing well at 3 months' follow-up. Later, elective restoration of bowel continuity was done with ileo-ascending anastomosis. The patient was doing well at 6 months post his second surgery.

Discussion

The earliest reported case of entero-scrotal fistulation goes back more than 100 years when an Englishman from Fyzabad reported it in a 12-year-old male.[3] Bassinis's repair and caecal closure were done under chloroform anaesthesia with an inguinal approach using silkworm gut. This patient had a prolonged history of ignoring the swelling for 1.5 months. This was followed by a similar report 40 years later in a premature infant from Portugal who had a Richter-type hernia on the left side which became irreducible and fistulated over 10 days. This was managed with an inguinal approach which again revealed caecum as the contents of the hernial sac with a perforated wall suggestive of associated midgut malrotation.[4]

In our case, the parents had noticed some redness over the scrotum which increased rapidly overnight leading them to seek medical attention. While travelling from a remote location to our centre, the scrotal fistula formed. The rapid fistulation may have occurred as the neonatal bowel has a thin wall and precarious blood supply. Moreover, the scrotal skin is also thin and devoid of fat in general. The poor nutritional status of the child may have also played an amplifying role in our case. There is an aspect of delay in the presentation as the child should have been brought to a clinician at the first sign of redness. Lack of parental education about common emergent medical conditions should also be considered in the index case as there is a possibility that a pre-existing hernia had gone unnoticed by the parents and only raised concern once erythema began to develop. The post-natal healthcare survey should have recognised the defect early but it appears otherwise.

Over the years, various contents have been reported to fistulate through the scrotum such as an appendix with the tip as faecal fistula[5] or ileal loop with Meckel's diverticulum.[6,7] All the cases reported by various authors in the past have been reviewed and summarised in Table 1. Few trends emerged across these cases and have been discussed here.

Right-sided fistulations are more common (19 vs. 3) and can be simply explained by the fact that right inguinal hernias are more common. Almost three-fourths of reported patients are < 1 month old with the acute onset of symptoms. This goes hand in hand with the increased incidence of incarceration in this age. Considering the history given by the parents, the index case has a very short time duration from (probable) irreducibility to fistula formation and further calls to attention the urgency of neonatal hernias. The contents of the sac have varied with a Richter-type hernia being the most common. The usual mode of presentation is with a discharging scrotal opening. Most of them have a history of delayed presentation with no symptoms of toxaemia while having a faecal discharging sinus. As discussed before by Ugwu *et al*.,[8] the scrotal opening acts as a pop-off mechanism which prevents peritoneal contamination and toxaemia.

There is only one report where the child's condition deteriorated before being explored.[9] The rapid deterioration of such neonates is also compounded by the delay in seeking medical attention. The post-natal surveillance of most childbirths in rural/remote locations is far from thorough in developing countries. Although the practice scenario has improved with time, we still have some distance to cover. We feel that these cases have been underreported as many similar cases would be have deteriorated even before seeking medical advice. With increased awareness of publication across the world, many case reports have come to light. Further proof of this assumption is that among 22 cases (including ours) with entero-scrotal fistula reported in the literature so far 15 have been reported in this millennium.

Conclusion

We report an extremely rare case of inguinal hernia in a neonate which progressed to a scrotal fistula in a short duration of 36 h and was managed surgically in two stages. We feel that the developing world has a lot of potential in improving the primary health care and referral system. With appropriate post-natal healthcare checks and better parental education, significant neonatal morbidities can be prevented. We advocate for considering neonatal hernia as an urgent surgery which will only be possible with improved post-natal survey and better grassroot healthcare services.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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