

Spontaneous intestinal perforation



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Neonatal necrotizing enterocolitis, the most common gastrointestinal emergency in neonates, typically affects the preterm infants and carries high mortality. Classic clinical trad consists of abdominal distension, bloody stools, and pneumatosis intestinalis. Occasionally, signs and symptoms may not be classic and a number of papers have highlighted the difficulty of relying on radiological features to make a definitive diagnosis of necrotising enterocolitis.

We report herein a case of neonate referred to us as a case of obstructed inguinal hernia and was diagnosed as a necrotizing enterocolitis perforation revealed by a rare manifestation of pneumoscrotum.

KEY WORDS: NEC, Pneumoscrotum, Spontaneous intestinal perforation.

Introduction

Evidence of pneumoperitoneum on x-ray usually indicates bowel perforation. There are myriad of causes for bowel perforation in a neonate. Neonatal necrotizing enterocolitis, the most common gastrointestinal emergency in neonates, typically affects the preterm infants and carries high mortality. A localized noninfectious enterocolitis with spontaneous perforation is an unusual cause but can affect small or large bowel. Occasionally, signs and symptoms may not be classic and neonate with radiological evidence of free air but without much clinical evidence of peritonitis can create management dilemma¹. We report a neonate who was referred to us as obstructed inguinal hernia, but turned out to be having pneumoperitoneum with pneumoscrotum apparently caused by spontaneous idiopathic bowel perforation.

Patient report

A 5-day-old 33 weeks preterm male baby, weighing 1.7 kg born to a primigravida mother through normal vaginal delivery was brought to pediatric emergency with complaints of right side inguinal swelling for the past 12 hours. The mother gave history of slight abdominal distension and constipation as well. There was a history of intake of non-steroidal anti-inflammatory drug (NSAID) in antenatal period but there was no significant perinatal event. Patient had passed meconium and urine within 16 hours of birth and was being breast fed. The birth APGAR was 10/10. The patient was referred to us as obstructed inguinal hernia by the attending pediatrician. On general examination his vital signs were within normal limits with baby being pink, well perfused and having a good cry. Physical examination disclosed that the abdomen was mildly distended but there was no evidence of abdominal wall erythema, tenderness or muscle guarding. There was a right sided inguinoscrotal swelling which was tense, nontender, translucent with a tympanic note, and could not be reduced (Fig. 1). Examination of chest cardiovascular system and central nervous system revealed no abnormality. Laboratory

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examination showed hemoglobin to be 18.3 g%, TLC was 7700/cc and platelet count was 72000/cc. The serum biochemistry was normal. Stool for occult blood was negative, blood culture was sterile. Upright abdominal X-ray film showed free air in the both sub-diaphragm and the presence of pneumoscrotum on right side (Fig. 2). Radiological evidence of pneumoperitoneum and pneumoscrotum suggested the possibility of spontaneous bowel perforation or NNEC (neonatal necrotizing enterocolitis). Since the patient had a stable general condition with lusty cry, good perfusion and no signs of peritonitis,

he was managed conservatively with needle aspiration of the scrotal swelling, nil per oral, nasogastric decompression, peripheral TPN and empirical intravenous antibiotics. The baby was closely monitored in intensive care with regular general physical and abdominal examination. Over the period of next three days the baby showed significant improvement in the form of return of bowel peristalsis and resolution of pneumoperitoneum on x-ray. Oral intake was started on the sixth day at hospital and slowly weaned off parenteral nutrition and graduated to full feeds. The baby was referred back to neonatology unit for further nutritional rehabilitation. The patient is doing well in follow up.



Fig. 1: Clinical photograph showing the right sided inguinoscrotal swelling. Note the tense and shiny appearance of the scrotum.



Fig. 2: Abdominal radiograph of the patient showing free air under diaphragm and right sided pneumoscrotum.

Discussion

Necrotizing enterocolitis (NEC), typically seen in premature babies, can also be witnessed in a term neonate from day one of life to several months age. Although the exact pathogenesis of NEC remains elusive, innumerable causes have been suggested. The most common implicated causes are prematurity, low birth weight and elemental formula, pathogenic microorganism, and bowel compromise: coalesce in an at risk neonate to produce the bowel injury². The clinical presentation of NEC is variable ranging from mild form to severe systemic disease with lethargy, recurrent apnea, temperature instability, bradycardia and abdominal signs². Spontaneous intestinal perforation is a localized disease which can affect gut anywhere from stomach to rectum. There are numerous causes implicated for spontaneous bowel perforation³. The causes implicated are bowel wall ischemia secondary to shock, septicemia, direct trauma during resuscitation measures⁴. SIP (Spontaneous Intestinal Perforation) is supposed to be separate disease from NEC most commonly affecting the very low birth weight (VLBW) and low birth weight (LBW) babies without much clinical manifestations.⁵ Antenatal NSAID intake has an association with this entity⁵. Our patient was low birth weight, was relatively stable with no signs and symptoms of NEC and mother had antenatal history of NSAID intake.

Pneumoscrotum, denoting the presence of gas within the scrotum, is an uncommon finding in neonates⁶. The cause of pneumoscrotum can be production of air locally or movement of air from the peritoneal space. Local air production suggests scrotal trauma or gas gangrene due to infectious causes and is usually fatal unless treated appropriately but it is rare in neonates. On the other hand, pneumoperitoneum from any cause can lead to movement of free air from the peritoneal cavity to scrotum due to persistent processus vaginalis⁷. Besides perforated NEC, there are other rare reported causes leading to pneumoscrotum and these are perforated Meckel's diverticulum⁸ and ileal atresia with perforation^{8,9}. Presentation of spontaneous intestinal perforation with

pneumoscrotum and clinical features mimicking as obstructed hernia has not been reported so far. In this case we based our decision for conservative treatment because patient's vital signs were stable, the abdominal signs were minimal and the pneumoscrotum was not due to local air production but originated from the pneumoperitoneum. Since the pneumoscrotum did not show any intestinal contents and the patient had an excellent recovery, the need for any peritoneal drain or exploration was obviated.

Therefore, it is essential for an attending pediatrician to consider spontaneous intestinal perforation, besides other causes of pneumoperitoneum, in the differential diagnosis of a patient presenting with a scrotal swelling (especially if the scrotum is tympanic on percussion) but with relatively stable general condition and few or absent abdominal signs so that an undue morbidity and mortality can be prevented.

Riassunto

L'enterocolite necrotizzante neonatale, l'emergenza gastrointestinale più comune in neonati, interessa tipicamente neonati prematuri e comporta un'alta mortalità. Triadi cliniche classiche consistono nella distensione addominale, in sgabelli sanguinanti e pneumatosi intestinale. Occasionalmente i segni ed in sintomi non possono essere classici ed un certo numero di articoli ha evidenziato la difficoltà di contare sulle caratteristiche radiologiche per fare una diagnosi definitiva di enterocolite necrotizzante.

Gli Autori segnalano il caso di un neonato per il quale era stata diagnosticata un'ernia inguinale ostruita che è

stata invece diagnostica come la perforazione di un'enterocolite necrotizzante rivelatrice di una manifestazione rara del pneumoscrotum.

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Commento e Commentary

PROF. NICOLA PICARDI
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Il caso presentato è molto interessante ed istruttivo nella sua rarità, specialmente per il tipo di presentazione clinica e soprattutto per la sua evoluzione positiva.

Per quanto riguarda la sua etiologia è opportuno avanzare sommessamente qualche ipotesi, differente da quella proposta dagli Autori, specialmente considerando la sostanzialmente spontanea risoluzione favorevole. Non è facile accettare semplicemente la diagnosi di enterite necrotizzante per completa assenza di altri segni generali ed addominali di coinvolgimento maggiore del piccolo paziente.

Il bimbo era un neonato di pochissimi giorni, ed anche se prematuro il parto aveva certamente rappresentato un recentissimo trauma. È possibile ipotizzare che durante il parto un qualche tipo di danno meccanico abbia interessato l'intestino, provocando una piccola perforazione dell'intestino - distensione diverticolare di Meckel o altro, con diffusione nel cavo peritoneale di aria praticamente sterile. Quindi si sarebbe verificata la risoluzione della situazione con riassorbimento dell'aria ed in assenza di altri segni di qualche importanza.

The case report is very interesting and instructive in its rarity, especially for its clinical presentation and moreover for its positive evolution.

However as for its aetiology it is proper to advance in a low voice some other hypotheses different from that of the Author especially considering the rather spontaneous favourable outcome. It is not easy to plainly accept the proposed diagnosis of necrotizing enteritis, because of the complete absence of other abdominal and general signs and of a major involvement of the little patient.

The baby was just newborn, and even if premature, the birth represented anyways certainly a very recent trauma. It is possible to think that during the labour some kind of mechanical injury affected the gut provoking a very little perforation - distended diverticle of Meckel or so - with diffusion of practically sterile gas in peritoneum. Then followed the recovery of the situation, with absorption of the air and without other signs of some importance.

Point to point Clarification to queries by Respected Prof. Nicola Picardi

ANSWER TO QUERY N. 1: We have clearly mentioned that it was Spontaneous Intestinal Perforation (SIP) a variant of necrotizing enterocolitis where in the abdominal signs and general signs are not marked; plus it was managed conservatively with a very good outcome which again goes in favor of SIP. That is why we did not consider any other diagnosis.

ANSWER TO QUERY N. 2: During delivery such type of trauma to cause the perforation of existing Meckel's Diverticulum would be a very significant trauma. This type of trauma causing Meckel's perforation is not known even in adults who are exposed to very high degrees of trauma. Therefore we did not consider this in diagnosis.