Idiopathic Spontaneous Intestinal Perforation in Neonates; Explorative Laparatomy or Primary Peritoneal Drainage?

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Abstract

Objective: To highlight our management of idiopathic spontaneous intestinal perforation (ISIP) in two medical teaching hospitals.

Methods: Retrospective study was done between Sept. 2003-Sept 2011.

Results: The total number of patients was 9. The gestational age ranged between 27-29 weeks. The age at presentation ranged between 8-21 days.

All the patients were males. Clinically there was severe tympanic abdominal distention. Radiologically there was severe pneumoperitonium. Operatively there was single perforation with normal surrounding bowel. The treatment was primary explorative laparotomy in 4/9 patients and primary peritoneal (PPD) drainage in 5/9 patients. The total survival rate was 77.7%.
The total morbidity rate was 25%. The morbidity and mortality rate after PPD were zero.

Conclusions: ISIP is a serious condition associated with a high mortality rate if not promptly recognized and treated. To overcome the high mortality, PPD is a good option to rescue and stabilize the patient before the definitive treatment.

Keywords: spontaneous, intestinal, perforation, neonates.

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Introduction

Gastrointestinal perforations in neonates remain a major threat despite advancement in antibiotic therapy and modern intensive care. The pathogenesis of intestinal perforations in general can be secondary to necrotizing entercolitis, ischemic necrosis, occlusive anomaly of the bowel, and it can be of no obvious cause (1). Necrotizing entercolitis still the leading etiology of neonatal intestinal perforations, as well as it is well described in the literatures for many years (2) (3). A catastrophic condition of an idiopathic spontaneous intestinal perforation (ISIP) is increasingly reported in the recent years all over the world.

Although the understanding of its

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pathophysiology continue to evolve, yet is not clearly understood. Many factors have been implicated in its pathogenesis, but still no single cause can be regarded as a basic etiological factor. Additionally it seems to be quite distinct entity from necrotizing entercolitis (4). ISIP is a surgical emergency associated with significant morbidity and mortality if it is not addressed early(5). The outcome largely depends on the early recognition and vigorous medical therapy and expeditious surgical approach to salvage the affected baby.

The current increase in the reported cases of ISIP is correlated with the increased number of survival of prematures and low birth weight babies especially in the developing countries (6).

The aim of the current issue is highlight our observation on neonates had idiopathic spontaneous intestinal perforation with concern on their management and outcome in two medical teaching hospitals.

METHODS

Retrospective study of all cases with final diagnosis of Idiopathic spontaneous intestinal perforation (ISIP) were reviewed.

The study was undertaken during the period Sept.2003- Sept.2011, at Al-Butnan Medical Teaching Center, Tobruk and Al-Beida Teaching Hospital, Al-Beida, Libya.

The data collected included: age, gender, birth weight, associated diseases with special focus on patent ductus arteriosus and pulmonary dysplasia, history of postnatal drug intake like corticosteroids and indomethacin, clinical features, time onset, radiological findings, operative findings and histo-pathological examination. The treatment options included: primary explorative laprotomy in one group and primary peritoneal drainage and lavage followed by delayed laparotomy in the second group. The morbidity and mortality rate were assessed. All the data were analyzed and evaluated.

RESULTS

The total number of patients was 9. All the cases were males. The birth weight ranged between 1.4 – 1.8kgm. The gestational age ranged between 27 weeks - 29 week.The age at presentation ranged between 8 days- 21 days. The onset was acute with rapid development of severe abdominal distention, reluctant to feed and deterioration in the vital signs. There was no family history of the same condition in all the patients. Additionally there were no any concomitant diseases other than prematurity, and no history of postnatal intake of corticosteroids or indomethacin. None of the patients had neither patent ductus arteriosus nor pulmonary dysplasia. All the patients showed pneumperitonium on plain X-ray of the abdomen with absence of both portal vein gas and pneumatosis intestinalis. Operatively all the patients showed single perforation of the small intestine at distance ranged between 10-18cm from the ileocaecal valve. All the perforations were seen on the antimesenteric boarder of the small intestine. The diameter of the perforations ranged 0.35- 0.50 cm. The surrounding bowel were apparently normal. There were neither macroscopic features suggestive of necrotizing entercolitis nor evidence of occlusive bowel disease. Histo-pathologically there were perforation with necrosis involving the muscular layer only without evidence of ischemia.

The treatment strategy: 1- In the first group primary explorative laparotomy was done in four patients (44.5 %) with V-shaped resection and primary anastamosis. One of the earlier cases was transferred early from our nursery, he survived the operation. The remained three were referred to our hospital late with a delay range from 36hrs- 48 hrs, two died after four days due to septicemia and respiratory failure, the remaining case was required re-exploration because of anastamotic leakage from the site of the previous anastamosis. The leaking segment was exteriorized as ileostomy. 2- In the second group of five patients (55.5 %) were treated primarily by peritoneal drainage and lavage because they were referred late to our hospital with significant abdominal distention and respiratory embarrassment. 24- 48 hours later explorative laparotomy and exteriorizion of the perforated segment as ileostomy. Closure of the stomas were done at later a period ranged from 3- 4 weeks, when the general condition of the patient became stable. The mortality rate for the whole group was 22.2% and the total survival rate was 77.7%. The mortality rate in the second group was zero, compared with a mortality rate of 50% after primary explorative laparotomy in the first group. Morbidity rate after explorative laparotomy group was 25%, while after primary peritoneal drainage group was zero.

DISCUSSION

ISIP still a rare cause of intestinal perforation. The development of ISIP seems to be multi-factorial. Congenital muscle abnormality, air embolism and intake of indomethacin were all have a recognized association with ISIP (6). Affected neonates perhaps were subjected to repeated venous arterial embolism from intravenous infusions. The foramen ovale may remain opened for several weeks in those babies, allowing a tiny air bubbles pass to the left side of the heart (4). Additionally neonates with persistent patent ductus arteriosus who received indomethacin are at a risk to develop isolated intestinal perforation, the cause may be due to the fetal circulation itself rather than the medical treatment of patent ductus arteriosus with indomethacin (4) (6). Moreover, the early administration of high dose of corticosteroid for treatment of pulmonary dysplasia is suggested as a significant risk factor for the development of ISIP (7) (8). Our patients neither had patent ductus arteriosus nor history of receiving indomethacin or corticosteriods. Animal model of spontaneous ileal perforation suggest that nitric oxide, insulin-like growth factor and epidermal growth factor skew trophism of the ileum (thinning of the submucosa concomitant with hyperplasia of the mucosa. Global depletion of the nitric oxide is associated with disturbed intestinal motility and transforming growth factor-alpha in the muscularis externa. This constellation of insults may make the distal ileum vulnerable to perforation during recovery of motility (8). On the other hand neonate with ISIP is likely to have severe placental chorioamnionitis with foetal vascular response. 93% of the mothers of neonates with ISIP have received antibiotics before or at delivery and 31% of the newborns had coagulase- negative staphylococcal sepsis versus 6% in the control subjects (9)(10). None of our patients had history of perinatal, labour and post partum problems.

ISIP has been shown to affect extreme prematures and low birth weight babies (8)(11) compare with our patients. A neonatal stress consequent to preterm birth is regarded as

determining factor for the etio-pathogenesis of ISIP. Neonates under 28 weeks of gestational age with a low birth weight show a particular predisposition to ISIP with a risk accounting to 96% (2). ISIP has no unique preoperative clinical features and usually the definitive diagnosis mostly depends on the preoperative findings.

The recognition of a pneumoperitoneum with absence of pneumatosis intestinalis and portal vein gas are highly suggestive features (4), compare with our patients. Neonatal bowel takes several hours to fill with gas down to the rectum, so timing abdominal X- ray in suspected patients may avoid delay in diagnosis (12). However, occult intestinal perforation without peritoneal gas was reported (11). Per-operatively the jejunum and the ileum are mostly affected (4)(6)(13) compare with our patients. Histo-pathologically there is focal perforation with necrosis of the muscularis externa and no sign ischaemic damage (8)(10) compare with our patients.

The fundamental principle of management of ISIP are general supportive measures, rationale antibiotic therapy and timely surgical intervention.

The management needs multidisciplinary team to deal with prematures and low birth weight babies, whom they have a high prevalence of bacterial sepsis than other infant and children (14). In the recent years the treatment of ISIP has undergone radical re-thinking, depending on the clinical condition of the patient and the severity of the disease. Apart from the standard explorative laparotomy, a primary peritoneal drainage (PPD) has been used. PPD was effective as a definitive treatment in only 11% surviving neonates, the rest either had delayed laparotomy or died (15). Indeed PPD alone as a definitive procedure was not used in our patients because the documented results still not encouraging. The efficacy of the PPD as a definitive procedure instead of laparotomy needs to be further evaluated (16). However, PPD as a definitive procedure is encouraged by others (4)(17). Additionally spontaneous sealing of the site of perforation was recorded, which enhance the idea of why surgery needs to be avoided in some cases and managed by PPD only (18).

In comparable with our study when the neonate is too small or too sick for laparotomy, PPD is used as a rescue and stabilizing measures rather than a sole surgical management, but as adjunct therapy to laparotomy (15)(16). To decrease the potential high morbidity and mortality rates associated with PPD (6)(19), other recent promising and effective techniques are used in the management of PPD. Window enterostomy and T-tube ileostomy are more applicable procedures than the conventional surgery with lower operative time, lower the time to full oral feeding and high postoperative weight gain (1)(20). A six months survival rate with the use of PPD and explorative laparotomy are 48.6% and 36.4% respectively (15) compare with our patients.

CONCLUSION

ISIP remains a surgical emergency with potential risks if there is any delay in recognition and optimum treatment. The conventional explorative laparotomy remains the standard procedure, however, the use of PPD followed by delayed laparotomy, are

good applicable options in delayed patients or too small neonates and even when the facility of tertiary pediatric surgery is not available, where a first line management can be applied and then transfer the patient to a tertiary centre. Advances in the intensive neonatal care and the application of the above less invasive techniques in the management of ISIP will result in a better prognosis and a high survival rate.

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انثقاب الأمعاء التلقائي مجهول السبب عند حديثي الولادة

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الملخص

الهدف: التركيز على طرق علاج انثقاب الأمعاء التلقائي الذاتي عند حديثي الولادة، أجريت في اثنين من المستشفيات التعليمية.

الطريقة: دراسة راجعة أجريت بين شهر أيلول 2003 إلى شهر أيلول 2011.

النتائج: العدد الكلي للمرضى كان 9. مده الحمل تراوحت بين 27-29 أسبوع. عمر المرضى تراوح بين 8-21 يوم. جميع المرضى كانوا من الذكور. سريريا كان هناك انتفاخ شديد في البطن. شعاعيا كان هناك استرواح بريتوني شديد. المشاهدات أثناء العملية كانت وجود ثقب واحد في الأمعاء. الأنسجة المحيطة بالثقب تبدو طبيعية. العلاج كان استكشاف التجويف البريتوني في 9/4 من المرضى، وبزل التجويف البريتوني الأولي في 9/5 من المرضى. النسبة الكلية للبقاء على الحياة كانت 7707%.

النسبة المرضية الكلية كانت 25%.نسبة الموات والنسبة المرضية لعملية بزل التجويف البريتوني الأولي كانت صفراً %.

الاستنتاجات: انثقاب الأمعاء التلقائي الذاتي عند حديثي الولادة مصحوب بنسبة عالية من نسبة الموات، إذا لم يعالج بصورة حازمة وبدون تأخير. للتغلب على هذه المشكلة فإن بزل التجويوف البريتوني الأولي يعد خياراً جيداً، لإنقاذ واستقرار حالة المريض، قبل إجراء العملية الجراحية النهائية.

الكلمات الدالة: عفوية، الأمعاء، انثقاب، حديثي الولادة.