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Recommended Citation

Amjad, A., Qazi, S., Nazir, Z. (2008). Enterocutaneous fistulae in children - a management challenge. *Journal of the College of Physicians and Surgeons Pakistan*, 18(8), 526-7.

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Enterocutaneous Fistulae in Children – A Management Challenge

Ali Imran Amjad¹, Saqib Hamid Qazi² and Zafar Nazir³

Enterocutaneous Fistula (EF) is an unexpected complication of intestinal surgery and is associated with high mortality and morbidity.¹ It can occur spontaneously in inflammatory bowel disease or subsequent to radiation therapy for abdominal or pelvic malignancies. Although many adult series are reported in literature, only one study describes experience in paediatric population.¹⁻⁵ We report a case series of seven children with EF to highlight the difficulties encountered in management in developing world.

Seven patients (age ranging from 14 days to 13 years; mean 7.1 years) with EF were admitted to paediatric surgical service at the Aga Khan University Hospital (AKUH), Karachi, from January 2003 to December 2006. Patients were self-referred after an initial treatment at other healthcare facilities. Important clinical features of patients are summarized in Table I.

At presentation, 6 out of the 7 patients had fluid-electrolyte imbalance and sepsis (positive blood cultures). Initial management included correction of fluid-electrolyte imbalance and control of sepsis (drainage of intra-abdominal abscesses under CT or ultrasound guidance in four children and broad-spectrum antibiotics). All patients were malnourished (weight loss >10%, low serum albumin; ranging from 1.2 to 2.3 g/dl) and received Total Parenteral Nutrition (TPN) for a mean duration of 32.5 days (ranging from 16 to 55 days). Somatostatin analogue (10 mcg/kg subcutaneously every 12 hours) was used in 2 patients, which reduced the fistula output substantially. None of the fistulae healed spontaneously. Six patients underwent surgical procedures including adhesiolysis

Table I: Enterocutaneous fistulae in children (n=7).

Parameters	n
Male : Female	6:1
Etiology (underlying surgical condition)	
Hirschsprung's disease	2
Typhoid perforation	2
Surgery for adhesive bowel obstruction	1
Congenital small bowel atresia	1
Bowel trauma	1
Site of Fistula	
Abdominal esophagus	1
Duodeno jejunal junction	2
Ileum	4
Fistula output	75-718 ml/day (mean 372 ml/day)
Outcome	
Survived	6
Death	1

(n=6), closure of perforation (n=3), and resection and anastomosis of small bowel (n=3). The operative time ranged from 7 to 10 hours (mean 8.8 hours) due to extensive intra-abdominal and inter-loop fibrotic adhesions. One patient died of uncontrolled sepsis (multi-drug resistant *Pseudomonas aeruginosa*) and liver dysfunction (total bilirubin 8.8 mg/dl) and albumin 1.2 g/dl) before surgery during the initial stabilization phase. Six patients were discharged after 23 to 93 days (mean 56 days) of hospitalization. The treatment costs ranged from PKR 325,435 to PKR 744,430 (US\$ 5,388 to US\$ 12,325). These figures, however, do not include the personal expenses by the family on illness before presenting to the hospital.

Enterocutaneous Fistula (EF) in the paediatric population is a rare entity.² In these patients, EF occurred following unintended shortfalls in operative techniques for common paediatric surgical procedures (e.g. Hirschsprung's disease, bowel perforation etc). This is in contrast to the adult population where majority of EF develop following bowel resections for intra-abdominal or pelvic malignancy.^{1,3} Guidelines for safe abdominal surgery are well-established and include control of sepsis (peri-operative antibiotics, mechanical bowel preparation), gentle tissue handling and attention to nutrition. Violation of these principles can result in EF. All of the reported patients were referred from resource-limited areas of the country without medical institutions with specialized paediatric surgery units. Presence of well-trained paediatric surgeons has shown to improve the outcome of various paediatric surgical procedures.⁶

Even for adults, the management of EF is a challenge.^{1,3} In children, the management is even more challenging due to the peculiar nature of their physiology. However, principles laid down by Edmunds *et al.* for the

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Received January 2, 2008; accepted July 30, 2008.

management of EF in adults (correction of fluid and electrolyte imbalance, control of sepsis and malnutrition) can safely be applied to children.³ Total Parenteral Nutrition (TPN) has revolutionized the management of EF allowing reduction in fistula output, healing of abdominal wound and attaining positive nitrogen balance thus reducing mortality.⁴ However, it is expensive and there is an increased risk of line sepsis introduced through intravenous routes and liver dysfunction. Line sepsis occurred in 4 patients requiring change of central line. The role of somatostatin and its analogues (e.g. octreotide) is well-established in the management of EF in adults.⁵ Our limited experience endorsed by one case-report demonstrates its safety in infants and children.⁷ The surgical procedure in patients with EF is demanding due to extensive intra-abdominal adhesions and friable intestines. Extensive resection of small bowel can reduce absorptive surface area and can lead to short bowel syndrome.^{1,3,5}

The development of EF can have deleterious effects on the growing child. The prolonged course of illness, hospitalization and multiple operative procedures affects children both physically and psychologically. They are often socially deprived and lose opportunities to learn and play. The present report shows that these children take months before being functionally normal. Cost of treatment, the family had to bear for this non-anticipated complication is phenomenal. Considering the per capita income of Pakistan to be PKR 43,200 (US\$ 720),⁸ the expenditures on hospitalization and treatment of unexpected complications following common paediatric surgical procedures is disproportionately high. This is burden on an already stressed health system of the nation and underscore the importance of establishment of specialized paediatric surgical services at district

headquarter hospitals readily available to children with comprise 45% population of Pakistan.

Enterocutaneous fistula is a serious complication of paediatric abdominal surgery. It is a result of inadvertent operative techniques and postoperative management. Attention to basic principles of bowel surgery such as gentle tissue handling, vascularity in intestine and consideration of bowel stoma in case of extensive intra-abdominal sepsis can avoid this complication.

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