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Mesenteric vascular occlusion in infants and children: Report of two cases and review of the literature

By

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Mesenteric vascular occlusion is a rare disease in the paediatric age group. The pertaining literature is reviewed and two cases are reported. In the first one the vascular obstruction developed after a Wilms tumour on the right side had been removed. Following bowel resection and end-to-end anastomosis the patient died of uraemia. Post mortem examination revealed a diffuse membranous glomerulonephritis and pseudoxanthomatosis in the remaining left kidney, the function of which had probably been affected by the shock associated with mesenteric thrombosis. In the second case a mixed mesenteric vascular occlusion was found without any previous disease; after ileal resection and ileo-coecal anastomosis the baby made a smooth recovery.

Mesenteric vascular occlusion due to thrombosis or embolism is a rare clinical and pathological entity in infants and children. The first reports date back to the second half of the last century [16, 31] and since then only 42 cases were described in the literature available [2, 3, 5, 7, 8, 9, 10, 11, 12, 13, 14, 15, 19, 20, 21, 22, 23, 25, 26, 27, 28, 29, 30, 32, 33]. (See Table I.) Most of these were single cases, only Ratner and Swenson [27] could collect 5 cases of the condition and even of these five, two were somewhat questionable.

The most striking characteristic of the disease is the very high (80%) mortality rate but this is well explained by the fact that 50% of the cases were diagnosed at necropsy and in the operated ones, surgery was mostly delayed. Among the patients (without the two to be described below) 15 were neonates, 8 infants and 19 children. All in all, 8 patients are known to have survived the condition; in 9 cases the outcome is unknown.

In half of the cases the occlusion was arterial, mostly in the area of the superior mesenteric artery. In the rest, it was venous or mixed. The occlusion was seldom due to embolism; it was mainly an aortic thrombosis that spread into the mesenteric artery [10, 22, 23]. The emboli originated mostly from the heart (right-left shunt) or from a patent ductus [4, 16]. The thrombosis was frequently preceded by some operation such as appendicectomy, laparotomy for intussusception, cardiac surgery. Two patients had undergone pyloromyotomy [3, 17]. In adult patients

TABLE I

Author and year	Age	Previous disease or operation	Length of infarcted bowel	Intervention	Nature of occlusion	Outcome
Klob [16] 1859	8 days	Patent ductus arteriosus	Not given	Diagnosis at autopsy	Superior mesenteric artery branches (clot from ductus arteriosus)	Died
Gruner [10] 1904	5 wks	None	Not given	None	Coeliac artery (clot from ductus arteriosus)	Died
Jackson et al. [13] 1904	1 mo 5 yrs 8 yrs	Details unknown				
Trotter [32] 1913	4 cases, age between 5–10 yrs	Unknown	Unknown	Unknown	1 arterial, 2 venous and 1 mixed thrombosis	Unknown
Frank [8] 1923	8 yrs	Peritonitis	Terminal ileum, coecum, ascending colon	None	Not given	Died
	10 yrs	Appendicectomy	70 cm small intestine	Resection and anastomosis	Superior mesenteric artery thrombosis	Survived
	12 yrs	Obstructing band, peritonitis	Not given	Explorative laparotomy	"Mesenteric vessels"	Died
McClanahan [19] 1926	4 mo	Intestinal obstruction	Distal 12–15 cm ileum	Explorative laparotomy	Not given	Died
Hogg [12] 1932	9 yrs	Blunt abdominal trauma	Distal ileum	Not given	Mixed thrombosis	Died
Feldstein & Goldstein [7] 1936	6 yrs	Ulcerative process of colon, Mastoiditis	Not given	None	Superior mesenteric artery and vein thrombosis	Died
Johnson [15] 1940	56 days	Intraperitoneal infection	Not given	None	Superior mesenteric vein	Died
Birnberg, Hansen [2] 1942	14 yrs	Migrating thrombophlebitis	Entire small bowel	Explorative laparotomy	Thrombosis of all mesenteric veins	Died
Gross [9] 1945	13 mo	None	Distal ileum	None	Inferior mesenteric artery thrombosis	Died

Morison [23] 1945	18 days	Umbilical infection after exchange transfusion	Descending and pelvic colon	None	Inferior mesenteric artery thrombosis, aortic thrombosis	Died
Miller, Bryant [21] 1952	30 mo	Appendicectomy	Terminal ileum and coecum	Resection and anastomosis	Not given	Survived
Carucci [3] 1953	1 mo	Pyloromyotomy	Lower ileum	Not given	Superior mesenteric artery thrombosis	Unknown
Rotschild et al. [29] 1953	6 days	Intestinal obstruction	Not given	Resection and anastomosis	Superior mesenteric artery thrombosis	Died
Mersheimer et al. [20] 1953	10 yrs	Traumatic splenic rupture, splenectomy	Proximal 35 cm jejunum	Resection and anastomosis	Not given	Survived
	6 days	Intestinal obstruction	Not given	None	Superior mesenteric artery thrombosis	Died
Ratner, Swenson [27] 1960	2 days	None	From upper jejunum to lower ileum (117 cm)	Resection and anastomosis	Superior mesenteric artery and vein thrombosis (Anomalous vessels)	Died
	3 days	None	20 cm long jejunoileal bowel	Resection and anastomosis	Arterial thrombosis	Survived
	6 mo	Septicaemia	Total ileum, coecum and ascending colon	Resection and anastomosis	Thrombosis of small mesenteric arteries. Panarteritis	Died
	5 days	Left sided diaphragmatic hernia. Operative reconstruction	Total small bowel	Laparotomy only	Superior mesenteric vein thrombosis	Died
	7 yrs	Appendicectomy	90 cm long jejunum	Resection and anastomosis	Superior mesenteric vein thrombosis	Survived
Schaffer [30] 1960	5 wks	Mesenteric cyst	Not given	Not given	"Mesenteric vessels"	Died
Miller et al. [22] 1961	33 hrs	Patent ductus arteriosus	Entire small bowel, coecum, first portion of ascending colon	Not given	Superior mesenteric artery thrombosis (clot from abdominal aorta)	Died
Lukács, Hittner [17] 1962	4 mo	Pyloromyotomy	120 cm long ileum	Resection and anastomosis	Superior mesenteric vein thrombosis	Survived

Author and year	Age	Previous disease or operation	Length of infarcted bowel	Intervention	Nature of occlusion	Outcome
DeMuth [5] 1962	7 yrs	Appendicectomy	70–80 cm long jejunum, sigmoid colon	None	Superior mesenteric artery thrombosis	Died
	9 days	None	Entire small bowel	Explorative laparotomy	Superior mesenteric artery thrombosis	Died
Otto [25] 1964	3 yrs	Adrenal tumour (exstirpation)	Total ileum	Not given	Not given	Unknown
Raffi [26] 1965	4 wks	Amniotic fluid aspiration. Enteritis	Total ileum, coecum	None	No histologic sign of thrombosis (?)	Died
	3 days	None	10 cm long lower ileum	Resection and anastomosis	Mixed thrombosis	Died
Rosenkrantz, Smiley [28] 1966	20 days	Transposition of great vessels, ventricular septal defect. Operative correction at 12 days of age	About 160 cm long small bowel	Embolectomy, resection and anastomosis	Superior mesenteric artery thrombosis	Died
Zerbes, Kluge [33] 1967	9 yrs	Appendicectomy and resection of diverticulum	120 cm long lower ileum, coecum, ascending colon and 2/3 part of transverse colon	Resection and ileotransversostomy	Mixed thrombosis	Survived
Jasonni et al. [14] 1972	7 days	None	Entire small bowel, coecum, ascending colon, half of the transverse colon	Explorative laparotomy	Superior mesenteric artery thrombosis	Died
Halmos, Teész [11] 1972	4 days	None	Entire small bowel	Explorative laparotomy	Superior mesenteric vein thrombosis	Died
Davies [4] 1973	11 mo	Imperforate anus, meningocele. Operation in neonatal period	35 cm long ileum	Resection and ileostomy	Superior mesenteric vein thrombosis	Survived
Present cases	2½ yrs	Wilms tumour (exstirpation)	40 cm long distal ileum	Resection and anastomosis	Superior mesenteric vein thrombosis	Died
	6 mo	None	60 cm long lower ileum to the ileocoecal valve	Resection and ileo-coccostomy	Mixed thrombosis	Survived

different other predisposing factors and diseases are found, such as endocarditis, hepatic cirrhosis, appendicitis, diverculitis, peritonitis, but these are uncommon aetiologic factors in the paediatric age group. Early periarteritis nodosa, disseminated panarteritis, chronic nephritis, serious diabetes mellitus, mongolism, juvenile arteriosclerosis may also cause vascular occlusion. In one case, mesenteric occlusion was induced by migrating thrombophlebitis that allowed an early preoperative diagnosis [2]. In the majority of cases, however, the aetiology remained uncertain or unknown. (Extensive bowel infarction caused by volvulus is not classified into this group.)

There are no characteristic clinical symptoms that would help to establish an early diagnosis. Usually, an acute abdomen is in the foreground: abdominal pain and distension, spastic attacks, nausea, vomiting and shock. In infants and young children shock symptoms are expressed. A characteristic bloody stool is an early sign; later, bowel movements stop altogether. There are no specific findings except an increased leucocyte count.

The prognosis is poor, the mortality rate is between 80-100%, especially in patients who are not operated upon or in cases where the intervention is performed too late. Early diagnosis and early surgery raise a certain hope to save the child, especially where the bowel infarction is localized to a small part of the gut [4, 27]. With early resection the mortality rate could be decreased to 50-84% [33]. Postoperative heparin therapy led to further improvement in adult patients [6]. Similarly, in some cases where the arterial occlusion was diagnosed early, embolectomy complemented with heparin therapy was successful. Such an intervention in a baby was published by Rosenkrantz [28], but the infant could not be saved.

In the following, two cases observed and treated by us will be reported.

Case reports

Case 1. A two and half year old female child was admitted because of an abdominal mass. A Wilms tumour weighing 340 g was removed without damaging the capsule (Fig. 1). Postoperative actinomycin D and therapeutic irradiation were tolerated well. Three weeks later, clinical signs of bowel occlusion developed suddenly, confirmed by X-rays. Laparotomy was performed and serosanguinous fluid was noted in the peritoneal cavity. A 40 cm long distal ileum was infarcted, with occlusion of the mesenteric vessels belonging to it. The infarcted bowel was resected, and

Fig. 1. Case 1. The removed Wilms tumour.

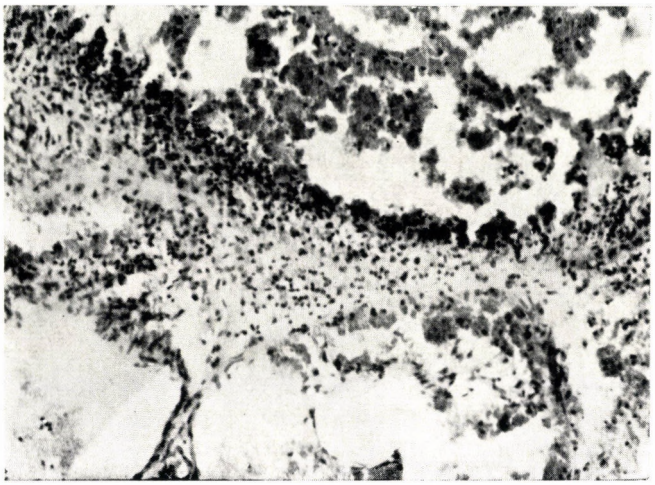

FIG. 2. Case 1. Resected mesenterium: diffuse blood clotting in the veins.

end-to-end anastomosis was performed. Histologic examination showed thrombosis in the resected mesenteric veins (Fig. 2).

Spontaneous bowel movements started on the 2nd postoperative day, but the state deteriorated gradually: the patient passed bloody stools, then became oliguric, finally, oedemas appeared in several areas, especially on the face. In spite of vigorous therapy the condition deteriorated rapidly and the child died of uraemia on the 23rd postoperative day. Post mortem neither tumour metastases nor a new thrombosis were found, but the remaining left kidney displayed diffuse membranous glomerulonephritis and pseudoxanthomatosis.

Case 2. A 6-months-old female infant was admitted with an acute abdomen, in grave shock. As a newborn she had had aspiration pneumonia then thrived well until 3 days before when she had run a fever. On admission the baby was unconscious and cyanotic, she had a distended, overtight and painful abdomen, she vomited several times and bowel sounds were not audible. In the faeces, *E. coli* was found. After the serious metabolic acidosis and shock had been relieved, a laparotomy was performed. Bloody fluid was evacuated from the peritoneal cavity. A 60 cm long portion of the lower ileum was infarcted up to the ileocoecal valve (Fig. 3), in the corresponding mesenterium the vessels were thrombotized. After resection of the infarcted ileum, the coecal opening was closed in two layers and an ileo-coecostomy was performed.

Histologic examination showed a mixed, arterial and venous occlusion of the mesenteric vessels. After broad spectrum antibiotic therapy the faeces became negative. On the 2nd postoperative day spon-

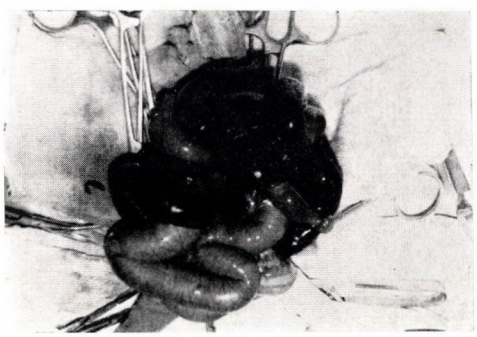

FIG. 3. Case 2. The infarcted ileum differentiates well from the normal bowel (Intraoperative photograph).

taneous bowel movements started and the baby passed normal stools. On the 6th day a faecal fistula developed, but it closed spontaneously 12 days later. In the next two weeks the condition improved quickly and on the 28th postoperative day the baby was discharged in a good condition. In the next few months the stool frequency changed from one to four daily, sometimes it was profuse and watery. An adequate diet and pancreatic extract were prescribed.

At the age of one year the baby weighed 7600 g, her somatic and mental development was normal. She has normal stools, once or twice daily.

Discussion

Infarction of the ileum was caused by venous thrombosis in the first case, and by a mixed obstruction in the second one. The aetiology remained obscure in both cases, but in the first patient, the mesenterial thrombosis had been preceded by nephrectomy and Wilms tumour resection. Otto [25] described a similar case in a 3 year old child where an adrenal tumour had been removed. The child died and necropsy revealed obstruction of the mesenteric vessels.

In our second case there had been no previous disease or operation except for the presence in the stools of *E. coli*. The aspiration pneumonia observed soon after birth was not likely to be in connection with the bowel obstruction. A coagulopathy did not develop and so postoperative heparin therapy was not applied in either of the cases.

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