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Remnants of vitelline duct: analysis of 66 cases

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In twenty years, 66 infants and children with remnants of vitelline duct requiring surgery have been admitted. The patients were classified into three groups: patent vitelline duct (20 cases); Meckel's diverticulum as the primary surgical disease (19 patients); and Meckel's diverticulum found incidentally at surgery (27 patients).

The male preponderance in the groups of patent vitelline duct and symptomatic Meckel's diverticulum was 9 : 1. In contrast with other data, Meckel's diverticulum requiring surgery occurred with nearly equal frequency up to fourteen years. The gravest complication in the cases of patent vitelline duct were a T-shaped protrusion of ileum and a small bowel volvulus around the fibrous cord or the patent duct; and in the cases of Meckel's diverticulum causing symptoms, intestinal obstruction, bleeding peptic ulceration or inflammation.

Three deaths occurred in newborn age in connection with patent vitelline duct, and one patient died who belonged to the group of asymptomatic Meckel's diverticulum.

The vitelline duct (vitello-intestinal duct or omphalomesenteric duct) connects the primitive midgut and the embryonic yolk sac in early intrauterine life. At about the fifth week, once placental nutrition has become established, a progressive narrowing of the duct occurs; it is completely obliterated by the seventh week of fetal life. Arrest of this obliterative process at different stages results in a variety of congenital abnormalities capable of producing a wide variety of clinical disturbances.

If the vitelline duct remains widely patent (Fig. 1a) a prolapse or intussusception of the ileum through the abdominal wall (Fig. 1b) may occur. This is the most terrifying complication of this congenital malformation. The prolapse causes a T-shaped protrusion of the small bowel (Fig. 1c). If the duct is narrowly patent, it may discharge mucus, gas bubbles or faeces.

If the distal end fails to obliterate, an incomplete fistula (Fig. 1f), or a so-called sinus develops (Fig. 1e) which opens at the umbilicus and usually secretes mucus. On inspection a bright red, slightly haemorrhagic polypoid formation with a central opening is seen. The anomaly often leads to inflammations which are usually diagnosed as omphalitis, umbilical granuloma, or umbilical sepsis.

The vitelline duct cyst is a rare anomaly; it appears as an abdominal

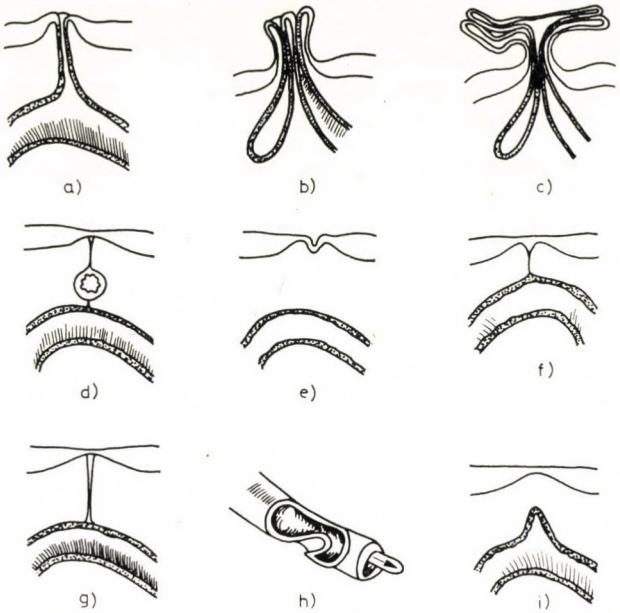

FIG. 1. Remnants of vitelline duct; a) persistent vitelline duct; b) intussusception through vitelline duct; c) T-shaped protrusion of the duct; d) incomplete duct; e) umbilical sinus; f) vitelline duct cyst; g) Meckel's diverticulum, h) Meckel's diverticulum causing ileal intussusception; i) fibrous band between ileum and umbilicus

mass, mainly in newborn infants (Fig. 1d).

The totipotential cells of the vitelline duct sometimes persist and thus the duct fails to obliterate and gives rise to ectopic tissue. The most common form of this is Meckel's diverti- or culum (Fig. 1i). Gastric mucosa, pancreatic tissue, duodenal glands colonic mucosa may be found in it. This ectopic tissue accounts for at least two of the complications occurring in the diverticulum with resulting haemorrhage or perforation. The diverticulum may invaginate into the lumen of the small bowel and serve as the leading point of ileal intussusception (Fig. 1h). Acute inflammation of the duct may be due to peptic ulceration or non-specific inflammation, the pathogenesis being probably similar to diverticulitis elsewhere.

Around the obliterated or patent duct (Fig. 1g) a small bowel volvulus or internal herniation may develop resulting in life-threatening intestinal obstruction, strangulation or ischaemic necrosis.

The aim of the present study is to analyse the main features of the remnants of the vitelline duct with special reference to its clinical signs, radiological and histological appearance, and variations. The operative findings and procedures and the follow-up studies are also detailed.

MATERIAL

During the twenty year period 1956 to 1976, 66 infants and children with various

forms of remnants of vitelline duct requiring surgery were admitted. Because of the wide variations in the clinical appearance and the problems presented by these malformations, the material was classified into three groups: patent vitelline duct, Meckel's diverticulum requiring emergency surgery, and Meckel's diverticulum found incidentally at surgery. Each group will be considered separately.

Patent vitelline duct

Twenty patients belonged to this group.

Sex distribution: there was a marked male preponderance (male : female = 17 : 3).

In age, the patients ranged from a few hours to seven years; 15 were under three months of age (Table I).

In most cases the diagnosis on admission already indicated or suggested the kind of congenital anomaly (Table II).

Diagnosis

In five patients there was a widely patent duct discharging gas bubbles or faeces, or a duct with partial ileal prolapse, or a T-shaped small bowel intussusception. These cases did not require diagnostic manoeuvres. In further ten patients where the diagnosis was obscure, injection into the duct of radio-opaque material demonstrated its connection with the intestine (Fig. 2) or showed a sinus or indicated a patent urachus duct. In two patients diagnosis was made in the course of surgery.

Associated anomalies

Major congenital abnormalities were encountered in one patient (oesophageal atresia, rectal agenesis, and severe heart failure). Other anomalies were exomphalos (3 patients), persistent urachus duct, cleidocranial disostosis, mesenterial defect (1 patient each) and malrotation of intestines (2 patients) (Table III).

All the findings were proved and classified at surgery. One persistent vitelline duct and one fibrous cord resulted in volvulus and severe subsequent intestinal obstruction. In a newborn infant the proximal part of the persistent vitelline duct was

Table I
Age at admission

	No. of patients		No. of patients
< 24 hours	3	1— 3 months	2
1—7 days	6	3—12 months	1
1—4 weeks	4	> 1 year	4

Table II
Diagnosis at admission

	No. of patients		No. of patients
Vitelline duct	6	Umbilical hernia	1
Omphalitis	2	Abdominal emergency (ileus)	2
Exomphalos	2	Appendicitis	1
Umbilical granuloma	4	No data	1
Umbilical sepsis	1		

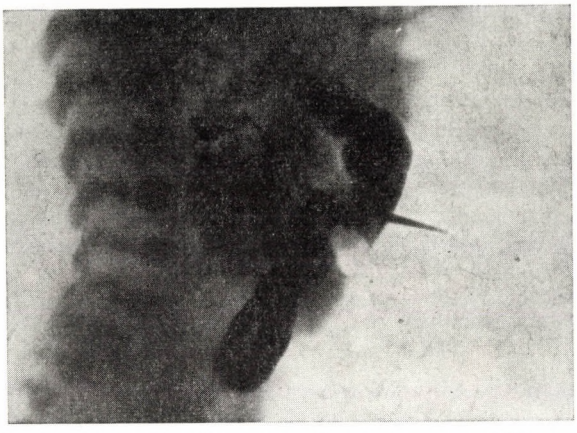

FIG. 2. Fistulography demonstrating the connection between umbilicus and small intestine

TABLE III

Types of patent vitelline duct

	No. of patients
Persistent vitelline duct without prolapse	7
Partial prolapse of ileum	2
Intussusception with T-shaped protrusion	2
Persistent vitelline duct + exomphalos	3
Fibrous cord + volvulus	1
Persistent vitelline duct + Meckel's diverticulum	1
Incomplete vitelline duct (sinus)	3

as wide as a Meckel's diverticulum and continued in a narrow but still patent duct to the umbilicus, this being patent on the abdominal wall (Table III).

In most cases, the vitelline duct was removed by careful wedge resection of the ileal wall. Manual reduction of the ileal prolapse followed by a resection of the duct was performed in two patients. Infants with T-shaped intussusception of ileum and one patient with volvulus required partial resection of the small bowel. The continuity of the ileum was re-established by single layer end-to-end anastomosis. The smallest patient of this group was a 2000 g premature infant. The newborn infant with multiple congenital anomalies was operated on only for treatment of its oesophageal atresia and rectal agenesis. (Table IV).

Type of mucosa

In five patients out of the eight with ileal mucosa in the vitelline duct, inflammatory changes were seen. In a newborn infant gastric mucosa was found. In the remainder no histological examination was carried out.

The patient with mechanical obstruction had to be subjected to repeated laparotomy in the early postoperative period

TABLE IV

Surgical procedures

	No. of patients
Removal of the duct	11
Removal of the duct + a segment of ileum	3
Removal of the duct + exomphalos	3
Removal of the duct + appendix	1
Incision of an abscess and later removal of the duct	1
Thoracotomy, gastrostomy, colostomy	1*

* No surgery for vitelline duct was carried out.

TABLE V

Postoperative period

Healing without complication		14
Healing with complication		3
Long-term suture suppuration	1	
Intestinal obstruction	1	
Sepsis	1	
Death		3
Persistent vomiting	1	
Generalized sepsis	1	
Multiple anomalies	1	

because of strangulation. A 30 cm length of small bowel was ischaemic and required resection; following ileal reanastomosis the patient recovered.

In this group there were three deaths. The cause of death in one case remained unclear even at necropsy. This infant, following resection of the vitelline duct, had a free intestinal passage but continued to vomit and failed to thrive, eventually dying of marasmus. The second death occurred after segmental small bowel resection, due to leakage of the suture line with subsequent peritonitis and generalized sepsis. (Table V).

The surviving patients were followed for an average of 11 years with a maximum of 20 years. All the surviving patients showed a good or sufficient physical development. Two of them have constipation and one is moderately retarded mentally.

MECKEL'S DIVERTICULUM

There were 46 patients with Meckel's diverticulum. Nineteen presented with various complications and required emergency surgery. In the remaining twenty-seven the diverticulum was found incidentally at surgery for some other condition.

MECKEL'S DIVERTICULUM CAUSING SYMPTOMS

Sex incidence: there was a very suggestive male to female ratio (18 : 1).

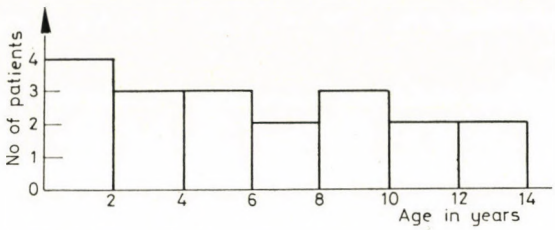

FIG. 3. Age distribution in manifest diverticulum

TABLE VI

Complaints before requiring hospitalization and surgery

Chronic paraumbilical colic	2
Dyspepsia	1
Chronic anaemia	2
Melaena	1
Vomiting	3
No previous complaints	8
No data	2

TABLE VII

Presenting complaints and findings on admission

Melaena	7
Vomiting	13
Abdominal colic pain	4
Abdominal distension, meteorism	9
Localized or diffuse peritonitis	7
Palpable abdominal mass	8
Bulging pouch of Douglas	1

Patients with symptomatic diverticula ranged in age from under two years to 14 years, mean 5.2 years. There was a slightly greater incidence in the first two years of life, which then remained steady with increasing age (Fig. 3).

Table VI demonstrates that half of the patients had had some complaints before hospitalization suggesting the possibility of Meckel's diverticulum.

The most characteristic presenting symptom was rectal bleeding which varied from mild to massive and was usually painless. Pain appeared in the older age group. One-third of the patients were seriously ill on admission, with fever, marked dehydration and some evidence of shock (Table VII).

Clinical picture

The patients displayed one of three clinical pictures verified at surgery (Table VIII).

TABLE VIII

Clinical picture of manifest diverticula

I Obstruction		8
Intussusception	6	
Band + volvulus	2	
II Peptic ulceration		8
Massive bleeding	6	
Perforation	2	
III Inflammation		3
Non-perforated	2	
Perforated	1	

In six patients with intestinal obstruction, an ileo-ileal or ileo-ileo-colonic intussusception with Meckel's diverticulum as the leading point was seen. The history of one of these patients was remarkable: a 6-year-old boy presented with severe abdominal pain. A fortnight before he had sustained a mild blunt abdominal injury producing no clinical symptoms. On admission, abdominal rigidity could not be estimated due to the very extensive pain in the abdomen. On rectal examination on the right side of the pouch of Douglas a bulging mass was palpated. Since the X-ray showed no opacity in the upper left abdomen, the mass was thought to be the spleen with a long pedicle squeezed into the small pelvis. At surgery it proved to be an ileo-ileo-colic invagination; its leading point was a Meckel's diverticulum with ectopic gastric tissue. Because the intussusception was irreducible, a segmental resection of ileum together with the diverticulum was performed. Fig. 4 shows the resected intestine with a phalliform Meckel's diverticulum.

In two patients the obstruction was due to volvulus of the small bowel around a Meckel's diverticulum which was attached by a fibrous band to the underside of the umbilicus.

Peptic ulceration caused usually painless intestinal bleeding in six patients. In the remaining two patients belonging to this group, the ulcer had perforated resulting in peritonitis and shock but no melaena.

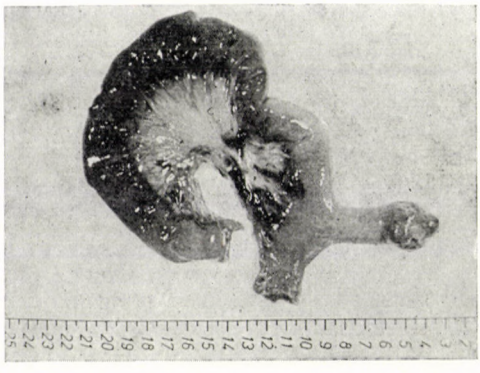

FIG. 4. Resected segment of small bowel with Meckel's diverticulum

Inflammation of Meckel's diverticulum was seen in three patients. Here, pain was the major presenting symptom and the clinical picture was indistinguishable from acute appendicitis. In one of the three patients inflammation led to a perforation of Meckel's diverticulum and subsequent severe peritonitis.

Mucosal histology. From the 19 patients, 13 had heterotopy (12 gastric and one colonic mucosa). In line with the literature a heteropic predominance was found in children with peptic ulceration.

Therapy

Simple diverticulotomy was performed if the diverticulum could be removed without compromising the lumen of the small bowel. In the four patients in whom the adjacent bowel was necrotic as a result of intussusception or volvulus, a segmental resection and an end-to-end anastomosis of the ileum was made. There was no death in this group of patients but complications developed in three children: septic infection, intestinal paralysis lasting for ten days, and mechanical obstruction requiring a temporary ileostomy for a couple of weeks. Follow-up showed a good mental and physical development in all patients.

Asymptomatic Meckel's diverticula

In 27 patients, Meckel's diverticulum was an incidental finding at surgery for other conditions like appendicitis, intestinal obstruction, mesenteric adenitis, Hirschsprung's disease, strangulated hernia, intra-abdominal testis, intussusception.

The age incidence was different from that of the diverticula causing symptoms. More than half of the patients were over ten years of age. In all but two Meckel's diverticulum was removed when it was detected. We only leave a diverticulum in place if there is a ruptured appendicitis, severe peritonitis, intestinal paralysis or mechanical obstruction, or in poor risk patients, etc. In a 5-month-old infant who was operated on for intussusception and a Meckel's diverticulum was found, a few months later a second operation was performed to remove the diverticulum.

As to mucosal histology, in only two children of the 27 patients was gastric mucosa found in the diverticulum; in the rest, normal ileal mucosa was detected in it.

There was one death in this group. An infant of three months was referred to us with strangulated irreducible inguinal hernia. At herniotomy a loop of small bowel and the adherent sac were not reducible and so a hernio-laparotomy was performed. During hernia repair a Meckel's diverticulum without associated pathology was found. Simple diverticulotomy was carried out and the closure was with 5—0 atraumatic silk thread. Unfortunately, in the postoperative period leakage of the suture-line with subsequent peritonitis led to the death of the infant.

Discussion

A considerable variation in the types of remnant vitelline duct has been found in our patients, and the incidence of the anomaly in our material amounted approximately to 1 : 2000 in the last twenty years.

The persistent vitelline duct including Meckel's diverticulum shows a marked male preponderance. Soderlund [16] reported a male to female ratio of 7 to 1 in a series of 54 cases causing symptoms, while in the asymptomatic cases the incidence in the two sexes was equal. Gross [5] observed a male to female ratio of 3 to 1 in a group of 149 manifest cases, while in our 66 children the ratio was about 4 : 1. If we consider the cases which required emergency surgery the ratio was 9 : 1, but in the asymptomatic diverticula only 3 : 1.

As with some other types of congenital malformation, prematurity was found to be a relevant factor in the patients with patent vitelline duct; only four of Singer's [15] ten infants with the anomaly weighed more than 2500 g at birth.

The different forms of remnant vitelline duct show a characteristic age distribution [9]. A patent duct was

found mainly immediately after birth, or in early infancy. If Meckel's diverticulum gives rise to symptoms, it will do so at any age but it occurs in over 50% of children before the second year of life [1, 5, 11]. Asymptomatic diverticula are inconsistently found in later childhood. In contrast, in our material Meckel's diverticulum requiring surgery displayed a nearly equal frequency up to 14 years of age.

It is easy to recognize a patent vitelline duct if there is a wide connection between intestine and umbilicus, especially in the early postnatal period. In a quarter of these patients a partial or total T-shaped prolapse of the small bowel developed, which is a life-threatening complication. Apart from two emergency cases with volvulus, all the patients were admitted with a diagnosis of omphalitis, exomphalos, umbilical granuloma, umbilical sepsis, or umbilical hernia.

In the manifest cases, a mainly painless rectal bleeding, sometimes intestinal obstruction or abdominal inflammation were the symptoms. These patients had a long history of repeated paraumbilical pains or of occult intestinal haemorrhage. This could have led to the diagnosis earlier. The most frequent preoperative diagnosis was intussusception or appendicitis.

Fistulography with contrast material will mostly demonstrate or disprove the connection between umbilicus and intestine. The diverticulum is not revealed by radiological examination but if intussusception occurs, it will be demonstrated by a barium enema [11]. Serious and painless rectal bleeding points to Meckel's diverticulum if sigmoidoscopy excludes rectal and sigmoid polyps [12]. Helpful in diagnosis is an abdominal scintigram with ^{99m}Tc [7, 8] owing to the affinity of technetium to the parietal cells of the gastric mucosa.

The therapy of choice of the patent vitelline duct is its removal. The operation is urgent if the ileum is prolapsed. Efforts should be made to limit the surgery to removal of the duct, avoiding to resect an ileal segment.

The diagnosis of Meckel's diverticulum is complicated by the variability of the symptoms [4, 14]. When no cause for the bleeding can be identified, an exploratory laparotomy is indicated; it often reveals a diverticulum [6]. Intestinal obstruction, secondary to intussusception of the diverticulum, is easily overlooked. Perforation of the diverticulum is an uncommon but most serious complication. In our material there was a single such case and we could not diagnose it preoperatively. Canty et al. [3] reported nine infants with perforated Meckel's diverticulum of which none was diagnosed preoperatively. For treatment, simple diverticulotomy with wedge resection of the ileum is usually sufficient. Cases with advanced obstruction resulting from intussusception or volvulus require a segmental resection of the small bowel with end-to-end anastomosis [2, 3].

The high incidence of ectopic mucosa, mostly gastric, is responsible for the majority of characteristic clinical

features such as peptic ulceration with bleeding or inflammation and probably for intussusception, too [9]. There is uncertainty as to how to proceed if a Meckel's diverticulum is discovered at surgery carried out for some other condition. Although many authors recommend diverticulotomy in such cases [2, 4, 9, 13] even in neonates [10], it is contraindicated in the presence of acute inflammation or severe disease of other abdominal organs. On the other hand, the fact that pathological changes (intussusception, torsion, inflammation, gangrene, peritonitis, adhesive stricture, etc.) develop in about 20% of all Meckel's diverticula [2, 13] encourages every surgeon to remove it, even if it is found incidentally. The death of an infant with Meckel's diverticulum in our material is a warning event. Our policy now is as follows. Under the age of one year we do not remove an incidentally found Meckel's diverticulum if other changes may endanger the suture line since in infants the greater omentum is not sufficiently developed to afford protection in the case of leakage After one year of age, intestinal obstruction due to some cause other than Meckel's diverticulum contraindicates the emergency resection of an incidentally found diverticulum in the dangerously ill patient. In these cases a second operation is indicated to remove the diverticulum.

Overall mortality amounted to 7% in the 66 patients with different forms of remnant vitelline duct. Three of the deaths were in connection with patent vitelline duct in neonates. No death occurred in the group of manifest Meckel's diverticula. One infant with asymptomatic Meckel's diverticulum with incarcerated hernia may have lived had we not removed the incidentally found diverticulum.

Thus, the prognosis is favourable in all forms of remnant vitelline duct. After due treatment these patients recover and develop well both physically and mentally.

In summary, the mortality risk associated with the various types of remnant vitelline duct must be borne in mind together with the experience that early diagnosis and proper surgical treatment can only prevent the life-threatening complications. Dr. Charles W. Mayo's statement "Meckel's diverticulum is frequently suspected, often looked for, and seldom found" is still and will remain actual probably forever.

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