REMNANTS OF THE VITELLO-INTESTINAL DUCT

A CLINICAL ANALYSIS OF 88 CASES

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Remnants of the vitello-intestinal duct are said to be present in 2 to 4% of all routine post-mortem examinations, but presumably many people live their allotted span of life despite their presence and at no time have symptoms referable to them. On the other hand, these vestigial structures may make their presence known dramatically in the first few years of life, or, more rarely, in adult life. The complications to which they are subject are serious and are commonest in infants and young children of the male sex.

This paper is based on 88 patients under the age of 12 years who were operated upon in the Royal Hospital for Sick Children, Glasgow, during the past 20 years. In 73 cases there was a definite lesion involving vitelline duct remnants and in 15 the structures were discovered incidentally during laparotomy for some other condition.

Embryology

In the early weeks of foetal life the apex of the mid-gut loop has a wide communication with the yolk-sac, the vitello-intestinal duct. This structure, with its accompanying vitelline artery and veins, gradually becomes reduced in size until complete obliteration occurs at about the seventh week. The obliterative process begins at the umbilical end of the duct and extends towards the intestine and thus it is that the intestinal end persists most frequently in the form of a Meckel's diverticulum. This finger-shaped sac projects from the ante-mesenteric border of the ileum within 36 in. (76 cm.) of the ileo-caecal valve. It generally has a small mesentery in which run the patent remains of the vitelline artery and veins and its distal end is frequently free in the peritoneal cavity. In some instances, however, the diverticulum terminates in a thin cord which may remain attached to the umbilicus or acquire a new attachment elsewhere. This cord is the partially obliterated remnant of the vitelline duct and/or its vessels. More rarely the umbilical portion of the

duct remains, giving rise to one type of umbilical polyp (entero-teratoma) which may or may not be associated with an underlying diverticulum. Still more uncommon is the formation of an omphalo-mesenteric cyst (entero-cystocoele) in the partially obliterated duct. In some few instances the whole length of the duct remains patent.

Occasionally an uncomplicated diverticulum is found apparently within the leaves of the ileal mesentery. This anomaly was explained by Moll (1926) who suggested that the diverticulum, having been rotated through 180° by traction of the slowly obliterating vitelline vessels, was pulled against the mesentery and later acquired a filmy covering from it. Various writers have described as 'Meckel's' or 'giant' diverticula structures with a lumen communicating with that of the ileum but truly within the ileal mesentery as proved by their being crossed by the mesenteric vessels. Such structures are more rationally regarded as bowel duplications.

Although the vitelline duct is embryologically related to the ileum, its remnants, where a lumen exists, are not necessarily lined throughout with ileal mucous membrane, and heterotopic tissue is said to be found in 25% of cases (Matt and Timpone, 1940). Ladd and Gross (1941) in their series of 73 cases found ileal mucosa, combined with gastric mucosa in 40, with duodenal in four, with colonic in four and with pancreatic cells in one. In the present series the findings were: ileal mucosa alone, 33 cases; ileal combined with gastric mucosa, 15; and ileal combined with duodenal mucosa, one. In a number of specimens the mucosa had been too extensively destroyed by inflammatory changes for accurate identification of the microscopic structure to be possible.

Numerous explanations have been put forward to account for the presence of aberrant elements. The most acceptable is that of Greenblatt, Pund and Chaney (1936) who suggest that the endothelial lining of the embryo forms cell groups which function as a primitive digestive system and that, while

normally this system regresses as soon as its function ceases, occasionally 'a vestige of heterotopic tissue remains as a consequence of retarded retrogression of the vitello-intestinal duct'.

Clinical and Pathological Findings

In analysing the 73 cases in which the lesion of the vitelline duct was responsible for the clinical picture there were found to be 47 males and 26 females with an age incidence as follows:

Years	0-1	1-2	2-3	3-4	4-5	5-6	6-7	7-8	8-9	9-10	10-11	11-12
Number	21	4	4	3	11	4	7	6	4	4	4	1

The whole series of 88 cases has been grouped according to the preoperative diagnosis or operative findings thus:

Simulation of appendicitis (acute or chronic)	28 cases
Intestinal obstruction	18 ..
Intestinal haemorrhage	14 ..
Intussusception	8 ..
Umbilical polyp or fistula	5 ..
Incidental finding at operation	15 ..

Simulation of Appendicitis. In the whole group there were 28 cases and no deaths.

ACUTE. In this group there were 22 cases, the sex distribution was equal, and the ages ranged from 2 years to 11 years with an average of $7\frac{1}{2}$ years. The duration of symptoms varied between six hours and eight days and three children gave a history of similar but milder attacks. The symptoms and signs in 20 instances were indistinguishable from those of acute appendicitis in that the illness began with central abdominal colic and vomiting progressing to pyrexia and constant pain in the lower abdomen or to the right of the mid-line. Two cases only varied from this pattern; in one, pain and tenderness were confined to the upper left abdominal quadrant, and in the other the symptoms and signs were of a wandering character, the only case in the series showing this peculiarity although it is sometimes said to be a common feature diagnostic of the condition.

At operation all these children were found to have acute Meckel's diverticulitis. In 13 instances there was perforation of the diverticulum with peritonitis, which was generalized in six. In the remaining nine there was acute inflammation of the diverticulum only. The high incidence of perforation of the diverticulum (60%) is notable. In 17 cases the diverticulum alone was resected, but in four a resection, including the neighbouring ileum, had to be performed. The inflammation had been set up in one case by the presence of a large fish bone which had perforated the diverticular wall, and in another

a gangrenous and twisted omphalo-mesenteric cyst attached by one pole to an uninfamed diverticulum was found; histology showed ileal mucosa lining both diverticulum and cyst.

The fact that there were no deaths in this group is mainly attributable to two factors, namely, the greater average age and the similarity of the symptoms and signs to those of acute appendicitis, a clinical picture so well known that most cases were sent to hospital at a relatively early date. In spite of this, however, perforation and peritonitis were

present in 13 instances. Ladd and Gross (1941) state that peritonitis resulting from perforation of an inflamed diverticulum is of a rapidly spreading and often fatal type but this has not been our experience; in seven of the 13 cases a localized abscess only had formed.

In the young child pain, tenderness and rigidity due to an acutely inflamed appendix may be situated anywhere to the right of the midline; it is only in the occasional case in which the signs are to the left of the midline or of a wandering character that one can make even a tentative diagnosis of inflamed Meckel's diverticulum. However, when a diagnosis of acute appendicitis has been made and when at operation the gross appearance of the appendix is not in keeping with the clinical picture, search for a diverticulum should always be carried out.

CHRONIC. In this group of six cases there were three males and three females aged from 5 to 9 years. The average duration of symptoms was six months and these consisted of vague peri-umbilical colic and vomiting attacks of varying duration. Diarrhoea was present during the attacks in two instances. Although in all cases a normal appendix and an uninfamed diverticulum were removed, no patient had a recurrence of the symptoms which, according to Ladd and Gross, are due to an upset in peristalsis caused by the presence of the diverticulum.

Intestinal Obstruction. In this group (18 cases, nine deaths) there were 11 males and seven females with ages varying from 3 days to 11 years, the majority being between 4 and 5 years old. The duration of symptoms varied from six hours to six days and only one case had had a similar but milder attack. All these children were extremely ill and their symptoms and signs were in keeping with a diagnosis of severe intestinal obstruction. Dehydration was marked, especially in the younger children, and in 14 instances abdominal distension with ladder

pattern and visible peristalsis were evident. Only in one patient, in whom the umbilicus was markedly indrawn and fixed, could a tentative diagnosis of intestinal obstruction by a vitelline remnant be made.

The operative findings, the varied attachment of the obstructing cord and the associated mortality are shown in Fig. 1. Two patients died pre-operatively and necropsy revealed a volvulus of the diverticulum and associated ileum in both. Haber (1947) and others state that intestinal obstruction is the most common complication but in the present series it accounts for only 25%. Regarding its seriousness all are agreed. Intestinal obstruction by a vitelline remnant is an extremely lethal condition as is evidenced by the 50% mortality. This high figure can be attributed to several factors. The majority of patients are in the 4-5-year age group when the child is uncooperative, making early diagnosis difficult and delaying admission to hospital. Further, the effects of fluid and salt loss which accompany intestinal obstruction are much more serious in the young child than in the adult. In spite of replacement therapy, the cumulative effect is that the child comes to operation ill-fitted to stand a surgical procedure which by the very nature of the obstructing agent must be a lengthy one involving loss of blood and prolonged anaesthesia.

The fibrous cord from the diverticulum, which was the cause of obstruction in 15 cases, is of the consistency of thin string and consequently early strangulation of the snared loop or loops occurs; for this reason resection of varying lengths of bowel and anastomosis was necessary in four cases. The thinness of the cord and the tightness of the constriction may result in early development of transverse lines of gangrene on each limb of the strangulated loop where it underlies the cord. Such areas were present in three cases and had to be oversewn.

Apart from causing strangulation of loops of bowel the fibrous cord attaching the ileum to the umbilicus also provides an ideal axis around which rotation can take place. Volvulus at any age, whatever its cause, is a lethal condition and the two cases in which it occurred were no exception to the rule. Their symptoms of sudden onset and short duration (six and nine hours respectively) were those of fulminating intestinal obstruction with strangulation, and the rapid collapse and death before operation could be performed were due to severe surgical shock.

Intestinal Haemorrhage. In this group of 14 cases with two deaths (12 male and two female) the ages varied from 7 weeks to 10 years. Ten patients were

less than 18 months old and the duration of symptoms of eight of these was from 28 hours to two days; the remaining six patients had had similar previous attacks. In 12 instances bleeding was from a peptic ulcer secondary to the presence of gastric mucosa in the diverticulum, and in two cases, although the diverticulum was the source of the haemorrhage, no ulcer or gastric mucosa could be found on pathological examination.

Peptic ulceration in the diverticulum is due to the secretion of hydrochloric acid and pepsin by a gastric mucosal rest which may vary in size from a complete lining to a mere vestige. Those reported cases of ulcers involving gastric mucosa are of doubtful validity, and it is suggested by Aschner and Karelitz (1930) that the actual ulceration occurs in an island of ileal mucosa within the gastric rest.

According to Cobb (1936) the neck of the diverticulum is the commonest site for the ulcer, thereafter the body, the tip and lastly the immediately neighbouring ileum. Maddock and Coventry (1941) report a case in which the ulcer was in the ileum several inches distal to the diverticular orifice. In the present series six ulcers were in the diverticular body, four in the neck, one at the tip and one in the ileum at the diverticular orifice.

The history of the children in this group was that of attacks of melaena, which, in some cases, had culminated in the passage of unaltered blood, or of one haemorrhage sufficiently alarming to cause the patient to be brought to hospital. Some abdominal pain was usually present but was never severe unless perforation had occurred. Examination of the abdomen was not helpful except in three children under 2 years of age who showed signs of peritonitis and at operation were found to have perforated ulcers. Rectal examination in all 14 revealed the presence of blood, either bright red or altered to a varying extent.

Pre-operative blood transfusion was necessary in eight of the 10 cases below 18 months, their average haemoglobin level being under 50%.

The operative findings and associated mortality were as follows:

No peptic ulcer, diverticulum lined ileal mucosa	2 cases, 0 deaths
Unperforated peptic ulcer, gastric mucosa present	6 cases, 0 deaths
Perforated peptic ulcer plugged by omentum, gastric mucosa present	1 case, 0 deaths
Perforated peptic ulcer and localized peritonitis (abscess), gastric mucosa present	2 cases, 2 deaths
Perforated peptic ulcer and generalized peritonitis, gastric mucosa present	1 case, 0 deaths
Peptic ulcer penetrating a neighbouring structure, gastric mucosa present	2 cases, 0 deaths

The last two cases are unusual as penetration of such a peptic ulcer into another structure has not been noted previously in the literature. In one

INTESTINAL OBSTRUCTION

FINDINGS IN 18 CASES

FIG. 1.

instance the ulcer had deeply penetrated the root of the mesentery exposing a large artery and in the other a loop of distal ileum had been penetrated down to the mucosa. In six patients, of whom five were less than 18 months old, a resection of the diverticulum and three inches of ileum on either side, followed by side-to-side anastomosis, was performed and all recovered. The only fatalities were in the two cases with perforation and localized peritonitis.

Because the signs and symptoms vary according to age it is helpful to divide the cases into those under 2 years and those over 2 years. The condition is more common and more severe in the infant, 71% being under 18 months in the present series as compared with 80% under 2 years in that of Ladd and Gross.

CASES UNDER 2 YEARS. The passage of a varying amount of only slightly altered blood in a thriving infant in whom there have been no previous symptoms is the cardinal sign. Occasionally the first stool is black or dark but subsequent ones are bright red. Following the passage of the first abnormal stool the signs of haemorrhage—pallor, lethargy and collapse—may rapidly supervene. The 10 infants of this series had passed but little blood before admission and yet presented the picture of internal haemorrhage, the true state of affairs only being revealed on rectal examination when withdrawal of the examining finger resulted in an actual jet of fluid blood and clots projected to some distance. Higgons and Gundy (1937) suggest that this disparity between the clinical condition and the amount of blood known to have been passed is due to the slow entry of blood into the colon and its retention there, the bowel having lost its tone following blood loss. Some degree of spasm of the sphincter must also be present.

The duration of bleeding in the infant is usually short; it was less than 48 hours in eight of the ten cases and in the other two there was a history of bleeding for 24 hours about six weeks previously resulting in admission to hospital from which they were discharged when the haemorrhage stopped. On admission to this hospital both children were found to have perforated ulcers with peritonitis.

Abdominal pain was not a feature in this age group although some parents stated that the infant had had 'wind pains'. There were some slight abdominal distension and possibly some periumbilical tenderness. The three cases with perforated ulcers were recognizable by the features of peritonitis and the finding of blood per rectum.

CASES OVER 2 YEARS. The patients of this group had much longer histories than the preceding ones, the shortest being three weeks and the longest

four and a half years; three had previously been in hospital with the same complaint. The principal symptoms, recurring in attacks of varying duration and frequency, were the passage of black stools and occasionally of small amounts of red blood often accompanied by mild abdominal colic. Examination of the abdomen revealed no abnormality. Haemoglobin levels, however, were somewhat low and the faeces contained blood, altered to a varying degree.

That early diagnosis and active treatment are essential in the infant is underlined by three findings in the series. In the first place, in three cases out of 14 (23%) the ulcer had perforated and of these, two patients died despite operation. The high mortality of operation once perforation has occurred is agreed upon by all writers. Secondly, once haemorrhage from the ulcer has begun, a sudden increase may occur at any moment and the haemoglobin level may fall below 50% in a few hours. Thirdly, when the ulcer is of a penetrating nature, as was the case in two patients, a sudden massive and probably fatal haemorrhage from an eroded mesenteric vessel may take place.

The disease in the older child is a much less severe one and its manifestations therefore may recur over a period of years without the child ever becoming acutely ill. Greenwald and Steiner (1931) in their study of 34 cases of different ages came to similar conclusions.

Intussusception. Eight patients (five male and three female) presented with an intussusception. This is 11·2% of the total, slightly less than the percentage of 17 reported by Harkins (1933). None had had a previous attack and the average duration of symptoms before admission was 14 hours. The symptoms were no different from the usually accepted ones, namely spasms of acute abdominal colic and vomiting. In three cases the intussusception mass was palpable only by rectal examination, being situated entirely in the pelvis anterior to the rectum: in several, normal faeces were present in the rectum at first, blood and mucus only appearing some time after the administration of a small enema. At operation, four intussusceptions were of the ileo-ileal type, the remainder, having progressed further, were of the ileo-ileo-colic type. Seven were reducible and had been caused by invagination of the diverticular base; the irreducible one had been started by inversion of a diverticulum lined with oedematous gastric mucosa. Resection was carried out and the infant made an uneventful recovery (Aitken, 1950a).

Intussusception started by a diverticulum is seen more frequently in children and adolescents than in infants. Hertzler and Gibson (1913) in their review of 34 cases record 13 years as the average age, and

Bailey (1938) is of the opinion that it is commonest in adolescence. The average age in this series, however, was only $4\frac{1}{2}$ years, one patient being but 5 months old. The difference in these average figures may in part be due to the upper age limit of admission to this hospital being 13 years.

Umbilical Polyp or Fistula. All patients in this group (five, with one death) were males. In two, aged respectively 2 weeks and 8 months, an umbilical polyp with purulent discharge and excoriation of the skin had been present since separation of the cord. Caution and ligation had been tried without success. At operation the polyps were found to be connected by a fibrous band to a small Meckel's diverticulum; polyps, bands and diverticula were removed. In both cases the diverticulum was lined with ileal mucosa, but in one the polyp was composed of duodenal and in the other, of gastric mucosa.

Two patients, aged respectively 2 weeks and 4 months, had similar histories and appearances, and on probing the polyp a track running deeply was found. These were visualized by the injection of barium mixture and found to communicate with the small bowel. Polyps and tracks were removed at operation and found to be lined with ileal mucosa.

One child, aged 12 days, had a fistula in the umbilicus through which faeces and flatus were passed dating from separation of the cord. Umbilicus and fistula were removed and found to be lined with ileal mucosa. There was one death (bronchopneumonia) in the group; the others made good recoveries.

In the case of an infant with an umbilical polyp, especially when there is much excoriation of skin or when the usual treatments of cautery or ligation have failed, the possibility of the condition being a vitelline remnant should be considered. Careful probing of many such polyps reveals a small opening in the centre which will accommodate a fine catheter, and, following injection of a barium mixture or lipiodol, a skiagram will confirm its connexion with the small bowel. In many polyps where no fistula exists biopsy and microscopical examination will show the presence of alimentary mucosa, and in such cases exploration of the abdomen is sound practice owing to the probable association of a Meckel's diverticulum and cord. In the type of case with a large umbilical fistula through which faeces is passed the pathology and diagnosis are not likely to be in doubt. Although no case of prolapse of the ileum through such a fistula occurs in the series this complication is not so rare as one might expect. Kirtland (1951), reporting one case, states that of the 131 cases of patent vitello-intestinal duct so far recorded in the litera-

ture, prolapse had occurred in 28 instances. He therefore deprecates the practice of 'waiting until the baby is older' to operate on an umbilical fistula, and advises early and prompt operative treatment.

Incidental Finding at Operation. In the hospital in which these patients were treated it has been the practice to search for a Meckel's diverticulum in each case when an abdomen has been opened at operation unless there is a definite contraindication, such as the danger of spreading infection. In 13 cases of this group of 15, with no deaths, an inflamed but not grossly pathological appendix had been found and removed. The diverticulum was found on further search and removed.

In two babies, aged 1 day and 12 hours respectively, operation was performed for cure of an exomphalos, and in both the diverticulum was found firmly adherent to the fundus of the sac. In one, the diverticulum was separated but not otherwise interfered with until it was removed when the child was 8 years old, following attacks of abdominal pain and vomiting. In the second case, one loop of ileum entered the diverticulum, while two loops with separate mesenteries emerged; these continued into a duplicated caecum, colon and rectum (Aitken, 1950b).

Discussion

Complications associated with vitello-intestinal remnants are varied and may be serious, especially in children under 5 years of age. In the 88 cases reported in this paper there were 15 deaths, a mortality rate of 17%. When the non-pathological cases are excluded the mortality is 20.5%. Six of the deaths were in the first year of life, and six in the first to fifth years.

Cases resembling acute appendicitis and those in which the duct was found incidentally, all recovered. The one death in the umbilical polyp group was fortuitous. The most serious complication is intestinal obstruction; of the 18 patients presenting with this condition the majority were in the 4-5-years age group and nine died. On the other hand, intestinal haemorrhage (two deaths in 12 cases) and intussusception (three deaths in eight cases) occurred mainly in the infants and are the next most serious complications. It seems obvious from these figures that early recognition and active treatment of these three types of complication is of the utmost importance.

A number of patients in this series were sent to hospital unduly late. The delay in recognition of the child's serious condition was especially noticeable in the intestinal obstruction group where in several instances the possibility of obstruction was not considered until constipation and abdominal distension

had become marked. It cannot be too strongly emphasized that these are late signs; the two early signs of value in diagnosis are splashing on succussion and increased peristalsis, both of which can easily be elicited by auscultation of the abdomen. If radiography is available the finding of fluid levels in a straight skiagram of the abdomen with the child upright confirms the diagnosis. When these signs are present and when there has been no previous intra-abdominal disease, a vitelline remnant is one of the likeliest causes of intestinal obstruction in the 4-5 years age group.

In the intestinal haemorrhage group especially, not a few children had been discharged from hospital after a few days having had no treatment because bleeding had ceased, the child appeared perfectly well, and investigations, which included blood examination, sigmoidoscopy and barium meal skiagrams, had been consistently negative. With regard to the last, the lack of radiographic evidence of gastro-intestinal abnormality in these cases merely imparts a false sense of security as the possibility of demonstrating a Meckel's diverticulum by this means is remote.

The diagnosis of an ulcerated Meckel's diverticulum should always be considered in the case of a thriving infant who suddenly becomes pale and listless although it may have passed but one melaena stool or a small quantity of blood. Should the haemorrhage cease, as sometimes happens, especially in the older child, the condition should not be lightly dismissed and the patient discharged from hospital but, failing a reasonable explanation of the bleeding, a laparotomy should be performed. There is no place for expectant treatment in the management of these cases if the risk of further and possibly fatal haemorrhage or perforation and peritonitis is to be avoided.

As regards the group presenting with intussusception, the signs in the early case may be misleading. The intussusception, being initially an ileo-ileal one, is at a more proximal level in the bowel than the common ileo-colic type and consequently normal stools may be passed before blood and mucus appear in the rectum. Moreover, the intussusception mass being mobile at first may gravitate into the pelvis and lie concealed therein. Time may be saved in such doubtful cases by the administration of a small enema and by careful rectal examination with the

intent not only of obtaining blood and mucus but also of examining the pelvic contents.

Summary and Conclusions

A series of 88 children, in whom remnants of the vitello-intestinal duct were present, is analysed. The cases are discussed under six headings according to the pre-operative diagnosis and operative findings.

The overall mortality of the pathological cases in the series was 20.5%. The mortality is greatest at ages below 5 years and especially in the first year of life.

Of all the complications, acute diverticulitis simulating acute appendicitis was found to be the commonest, and intestinal obstruction to have the highest mortality.

Intestinal haemorrhage due to peptic ulceration was seen most frequently in the first 18 months of life, intussusception and intestinal obstruction between 4 and 5 years, and acute diverticulitis between 7 and 8 years.

Intestinal haemorrhage and intestinal obstruction cases, on the whole, were diagnosed and admitted to hospital late. Reasons and remedies for this have been suggested and the importance of early recognition and active treatment of these two complications have been stressed.

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