INTRALUMINAL INTESTINAL CALCIFICATION IN THE NEWBORN*

BY

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Meconium ileus is now a well recognized entity and I do not need to dwell upon it in this paper. It is not only the condition in which an abnormality of the intestinal contents causes obstruction in the neonatal period. In recent years more attention has been paid to meconium plugs as a cause of blockage (Clatworthy, Howard and Lloyd, 1956). The object of this paper is to draw attention to a third condition causing intraluminal obstruction in early infancy. I apologize for reporting a single case. The condition seems to be extremely rare; at any rate I have found very few pertinent references in the course of a careful search through the literature. I wonder, however, whether an increased interest in this syndrome may not result in further cases being reported, as has happened before with many other diseases which were originally thought to be rarities. I apologize also for not being able to throw any light upon the causation of this lesion. In spite of a large volume of clinical and biochemical investigation carried out upon this particular infant, the disease remains surrounded by mystery. Any light shed on the little understood physiology of the neonatal intestinal tract is of value and this is a further reason for the publication of this case.

Case History

Gillian was admitted to the Alder Hey Neonatal Surgical Unit on August 25, 1955, when 37 hours old, a one-month premature infant weighing 5 lb. She was the first child of parents both of whom had been treated for infertility for several years. The infant had vomited repeatedly since birth and the vomitus had become bile-stained; she had not passed any meconium.

On admission her general condition was fair. There was some abdominal distension and a radiograph in the upright position showed numerous fluid levels. On the right side of the abdomen a number of calcified, worm-like-shadows could be seen (Fig. 1). In view of the clinical and radiological findings a provisional diagnosis of low small bowel obstruction associated with meconium peritonitis was made and she was operated upon two hours after admission. At laparotomy the jejunum and

upper two-thirds of ileum were found to be bluish and distended with fluid. The lower ileum and colon were collapsed and contained a little semi-solid material. There was a Meckel's diverticulum, but apart from this no anatomical abnormality could be discovered. The obstruction was now thought to be due to meconium ileus; we were, however, mystified about the calcification in the radiological picture, as there was no evidence of meconium peritonitis. An enterostomy was performed on the most distal loop of distended ileum, through which a rubber catheter was passed proximally. A large amount of greenish fluid material containing many firm

Fig. 1.—Radiograph of the abdomen showing calcified shadows on the right side.

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yellow granules was aspirated. The catheter was then withdrawn and passed with some difficulty down the ileum. Saline was injected with some force and by repeated injections and massaging the gut, it was ultimately possible to evacuate the ileum and to pass the catheter into the caecum and empty the whole colon and rectum. The gut distal to the enterostomy was filled with soft, snowy white material containing similar yellow granules to those found in the proximal intestine. The material did not appear to adhere unduly to the wall of the intestine and was evacuated in the shape of long, worm-like stools. When the proximal and distal intestine was completely cleared, the enterostomy was sutured and the abdomen closed.

Post-operatively the child's condition was quite good; she was placed on intravenous infusions and continuous gastric suction. During the subsequent days large amounts of bile-stained fluid were aspirated and the bowels did not open, although bowel sounds could be heard. By August 31 her condition had deteriorated and on September 1 (a week after the original operation) it was decided to re-open the abdomen.

At the second operation the proximal intestine was found to be distended with thickish fluid identical with that found at the first operation; there was no mechanical obstruction. At about the level of the Meckel's diverticulum the intestinal content became semi-solid and was only with difficulty squeezed along the intestine. A double-barrelled ileostomy was performed at the site of the Meckel's diverticulum and the intestine was completely emptied right up to the duodenum by aspirating its contents through a rubber catheter; the distal ileum and colon were similarly cleared.

Post-operatively the child's condition was poor, the temperature subnormal, but over the next few days she gradually improved and by September 3, many bowel sounds could be heard on auscultation. Feeding with glucose water, and, rather irrationally, pancreatin, was started although the stomach aspirations were still bile-stained and contained trypsin. The baby started to vomit large quantities of bile-stained material and feeds were discontinued on September 4. The ileostomy was washed out with no result. On September 5 a little 'lipiodol' was given by mouth and the child screened. The oil passed rapidly through the small intestine. On the next day (September 6) the child was found to have lost a lot of weight and it was decided that the calory intake must be increased and she was therefore tried on breast milk feeds. Vomiting became again so profuse that the feeds had to be discontinued. Discouraged by this we did not resume oral feeding, but on September 10 a catheter was passed upwards through the ileostomy opening and a continuous drip of protein hydrolysate given. Another catheter was passed downwards through the ileostomy opening and a continuous 10 % glucose drip was administered. The resulting calory intake, small as it was, improved the general condition of the child. Oral feeding was again attempted with glucose water on September 15 and with peptonized milk two days later, but soon had to be discontinued and the child put back on continuous gastric suction because of profuse bile-

stained vomiting. On September 18 we attempted oral feeds with protein hydrolysate, again causing profuse vomiting. On the following day (September 19) we again attempted milk feeds with the same result and thereafter, apart from small feeds of glucose water, no further attempt at oral feeding was made.

The child had by now been on continuous intravenous infusions for three weeks and although her hydration and electrolyte balance had remained relatively constant, we were unable to give her enough calories and wasting was marked. During the fourth week her general condition gradually deteriorated. She developed pneumonia, attacks of cyanosis and Cheyne-Stokes breathing and finally collapsed and died 28 days after the original operation. During this time she had received five blood transfusions and 25 separate infusions of diluted plasma as well as large amounts of crystalloid intravenous solutions. In spite of continuous syphonage of her intestinal contents by gastric suction her blood chemistry was kept more or less within reasonable limits.

Laboratory Investigations. Many biochemical investigations were performed during the four weeks of life. The serial investigations of the blood urea, serum chloride alkali reserve, serum proteins and serum potassium showed no abnormality. Intracellular electrolyte analysis of the red blood cells showed no striking abnormality. The serum calcium, calcium ions, phosphate and alkaline phosphatase of both the infant's and the mother's blood were repeatedly estimated and found to be within normal limits.

The stools evacuated at the first and second operation received special attention. Radiography of the whitish material expressed from the colon showed numerous flakes of calcium corresponding to the opacities which were seen in the pre-operative radiograph of the abdomen (Fig. 2). In these first stools there was no trypsin, but repeated subsequent analysis both of duodenal juices and ileal contents were always strongly positive for trypsin.

Analysis of the meconium obtained at the first operation was carried out and compared with meconium obtained from normal infants of the same age. As will be seen from Table 1, the only significant difference (as was to be expected) was in the calcium content (five times

FIG. 2.—Radiograph of meconium expressed at first operation. There are numerous opacities due to calcification.

normal) and phosphorus content (eight times normal); the iron content of the meconium was definitely increased, but still so small that it was found impossible to estimate it quantitatively.

TABLE 1
ANALYSIS OF NORMAL AND ABNORMAL MECONIUM

Meconium	Gillian	Normal
Dry weight	10.5% of wet weight	22%
Total ash	7.2% of dry weight	4.3%
Na.	0.61%	0.65%
K.	0.12%	0.29%
Ca.	0.62%	0.12%
P as P	0.52%	0.06%
N2.	5.7%	4.7%

Necropsy. A careful necropsy and subsequent histological examination performed by Dr. C. Jones did not throw much light on the aetiology of the condition. There was no mechanical intestinal obstruction, the lower small intestine was dilated, the upper of normal calibre. It was noticed that the intestinal contents were more solid than usual, the upper jejunum containing solid instead of fluid material. The only other abnormal finding was a hydronephrotic left kidney.

Histologically the only positive findings were moderate fatty changes in the liver. The pancreas and lungs were normal. There was no evidence of mucoviscidosis. The intestine was histologically normal; no abnormality of

innervation of the gut could be observed, the ganglion cells appeared normal in size and distribution.

Discussion

Intra-abdominal calcification in the neonatal period is commonly found in meconium peritonitis, calcification occurring in the bowel wall or in meconium extruded into the peritoneal cavity (Neuhauser, 1944). The calcification in the bowel wall is probably the result of intra-uterine perforation of the intestine (Forshall, Hall and Rickham, 1952). That meconium in the peritoneal cavity can calcify within a matter of a few days was observed by Rudnew in 1915. We have recently seen a case with free meconium but no calcification in the peritoneal cavity. At operation we failed to suck the meconium out of the sac of a large complete inguinal hernia. There was radiological evidence of calcification within the scrotum three days after operation (Fig. 3). Calcification of meconium within the bowel lumen has never been demonstrated in meconium peritonitis.

Meconium plugs occasionally show microscopic evidence of calcification, as was first shown by Trump in 1912, but, as in faecoliths, the calcification is localized and removal of the plug is all that is necessary to prevent further calcification and cure the patient.

Intraluminal calcification has only been described twice in the literature, in both cases it was associated with mechanical intestinal obstruction. In a baby girl described by Camp and Roberts (1949) there was an extreme ileal stenosis for which an ileo-transverse colostomy was performed. The intestinal content was microscopically and radiologically very similar to that observed in our case. Unfortunately the child died soon after operation and we do not know whether the subsequent clinical course would have been the same as in our case. At necropsy no abnormality was discovered. The meconium was unfortunately not analysed. No blood chemistry studies were carried out.

A very similar case associated with anal atresia was reported by Khilnani, Wolf and Arnheim (1955). Once the obstruction had been removed this patient had no further trouble, passing normal stools, and made a perfect recovery. The meconium was very similar to that observed in our case; the blood chemistry of patient and mother was normal.

Our case appears therefore to have several unique features; she had no organic obstruction and following complete removal of the calcified meconium the intestinal content re-calcified and re-solidified. These features make one think of mucoviscidosis or long segment Hirschsprung's disease, but there was

FIG. 3.—Meconium peritonitis with complete left inguinal hernia. Radiograph three days after operation shows calcification of meconium in the scrotum.

no evidence of either. We are thus left with the supposition that this case constituted a separate clinical and pathological entity and that the lesion was due to the nature of the meconium, possibly due to some deficiency of gastro-intestinal function. It is unlikely that the enormous increase in the calcium content of the meconium can be derived from swallowed amniotic fluid, which contains very little calcium indeed (Camp and Roberts, 1949). It is more likely that the excess of calcium in our case was excreted into the intestine.

Unfortunately all our biochemical and histological investigations having been inconclusive, we have no explanation to offer, nor can we put forward any helpful therapeutic suggestions. During recent years we have learned a great deal about conditions which I should like to call 'meconium diseases', such as cystic fibrosis of the pancreas, mucoviscidosis, meconium peritonitis and meconium plugs, but the existence of the clinical entity described in this paper

shows that we have still a long way to go before we understand the mysterious processes governing the formation of meconium and intestinal secretions in the foetus.

Summary

A case of intraluminal calcification of meconium, the third to be described in the literature, is reported. The nature of the causative lesion is unknown and in spite of extensive biochemical and histological studies no further light can be thrown upon the aetiology of the condition.

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