

The factors which may alter the results obtained are discussed.

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Medical Memoranda

Pancreatic Cyst in Infancy : Recovery after Marsupialization

Pancreatic cyst is a very rare condition: Aldis, quoted by Aird (1957), found only 2 in 75,000 patients. Most cysts have occurred in adult females. The condition is exceedingly rare in childhood and infancy. Miles (1959), in a review of the literature, found only eight cases in infants under 2 years of age, and, of these, cysts were demonstrated at birth in only two, and none was operated upon in the neonatal period. His own case was that of a full-term female baby who was operated upon when 8 days old. There had been frequent regurgitation of the baby's feeds with increasing abdominal distension. Examination of the abdomen and chest by *x* rays, intravenous pyelogram, barium meal, and barium enema showed a homogeneous shadow over most of the abdomen, with upward and medial displacement of the stomach and downward displacement of the large and small intestines. Laparotomy revealed a large thin-walled multilocular cyst arising from the head and body of the pancreas: the cyst was excised and the baby made a satisfactory recovery.

CASE REPORT

The patient, a full-term first-born male child, was seen in the Waterford Maternity Hospital when 23 days old. He had been admitted with a history of intermittent vomiting since birth. The child was healthy-looking when first seen and weighed 7 lb. (3.2 kg.). Abdominal examination revealed a firm round swelling the size of a grapefruit in the hypogastrium: it could be freely moved over the greater part of the abdomen.

Operation was performed on the same day, the abdomen being opened through a supraumbilical midline incision. The cyst was seen to present between the stomach and the transverse colon. The gastrocolic omentum was incised and the cyst exposed. It was fairly thin-walled and was seen to arise from the neck and adjacent portion of the body of the pancreas. The cyst could easily be separated from the stomach and colon but was intimately blended with the pancreas over a distance of several centimetres. The splenic vessels were closely attached to the base of the cyst. It was decided not to attempt complete extirpation. The fluid was removed as completely as possible by aspiration, after which the greater part of the cyst wall was excised. The margin of the remaining portion of the cyst was then

sutured to the peritoneum of the middle third of the abdominal incision, using No. 1 chromic gut. The linea alba and the skin in the remainder of the incision were repaired with interrupted nylon sutures.

Over the course of the next 12 hours 120 ml. of half-normal saline, with added hyalase, was given subcutaneously, and 24 hours after the operation oral feeding with half-strength expressed breast milk was begun. Streptomycin 0.25 g. and penicillin 250,000 units were given twice daily for eight days. The child vomited approximately half of each feed on the day after operation and vomited once on the second post-operative day, otherwise the post-operative course was uneventful. Drainage from the wound continued for over two months after the operation. During this time the child's general condition was satisfactory in every respect. A tendency to excoriation of the edges of the wound was immediately checked by the use of I.C.I. barrier cream. On January 4, 1960, the wound dressing was completely dry for the first time, and on January 7 the wound was healed and remained so. On January 23 the baby's weight was 12 lb. 4 oz. (5.6 kg.) and he was taking full milk, "farex," rusks, and egg yolks. No swelling could be felt on abdominal palpation.

On May 6, over six months after the operation, the mother stated that he seemed to be healthy in every way, that he was feeding satisfactorily, and that his bowels were regular with normal-coloured stools. On examination he looked very well. The wound was soundly healed and no abnormality was found on palpation of the abdomen. His weight was 19 lb. 7½ oz. (8.8 kg.). In December Dr. Evelyn S. Harris, of Seattle, Washington, kindly sent me a report on the patient: he was well in every way and weighed 24 lb. 7½ oz. (11.1 kg.).

Microscopical examination of the cyst wall showed that it consisted of fibrous tissue lined by a very flattened epithelium. Diastase could not be demonstrated in the fluid; trypsin was present to a titre of 1/40.

COMMENT

More than one classification of cysts of the pancreas has been proposed. McPherson and Heersma (1948) classify pancreatic cysts as follows: (1) retention cysts; (2) proliferation cysts (tumour); (3) congenital cystic disease; (4) parasitic cysts; (5) pseudocysts; and (6) dermoid cysts. Fibrocystic disease of the pancreas is not usually characterized by gross cyst formation. In the present case the origin of the cyst from the pancreas could be clearly demonstrated at operation: one can only speculate on whether the cyst was a true cyst or a so-called pseudocyst. Pseudocysts arise as a result of previous inflammation or injury, and there was no evidence that either of these had occurred in post-natal life, and presumably the opportunities for the occurrence of either condition in antenatal life are limited.

The case described here showed no external evidence of congenital abnormality, and, with one exception, this is in accordance with the cases traced in the literature. The exception is the case described by de Lange and Janssen (1948). Necropsy showed a cyst about the size of a grapefruit lying below the stomach and liver. The right foot showed six toes, the right lung had only two lobes, and there was marked enlargement of the cisterna magna.

In most of the reported cases in childhood and in adult life abdominal distension has been evident and abdominal palpation has shown this to be due to the presence of a swelling in the upper part or left side of the abdomen. Vomiting has been a cardinal symptom in the cases occurring in childhood and infancy. In the adult the swelling is usually central and fixed. In the above case the cyst at the first examination was

in the site usually occupied by a distended urinary bladder, and on being moved manually from its position it slipped upwards to lie above the umbilicus and tended to remain there. Its extreme mobility was presumably attributable to a mobility of the retroperitoneal structures as a whole, since the cyst did not possess a long pedicle but was attached to the pancreas by a short and broad base.

In the reported cases the investigations carried out have included "straight" x-ray film of the abdomen, x-ray film of chest, barium meal, barium enema, and intravenous pyelogram. The necessity for these procedures, involving considerable exposure of the infant to radiation, is doubtful, since vomiting in infancy which persists in spite of conservative measures raises the question of surgical exploration, which is put beyond doubt by the finding of an abdominal tumour. There is unlikely to be any difficulty in distinguishing the tumour from the "tumour" of hypertrophic pyloric stenosis.

There is no general agreement on the form surgical treatment should take, though it is commonly stated that excision of the cyst is the ideal treatment but that it is not always feasible owing to close adhesion to surrounding structures. Where excision is regarded as impracticable, the choice of treatment lies between marsupialization and anastomosis of the cyst to the stomach, duodenum, or jejunum. In the present case it was felt that complex excision should not be attempted and that the wall of the cyst was so thin as to render an anastomosis with any part of the gastro-intestinal tract unsafe. Marsupialization, a less extensive procedure than either excision or anastomosis, was therefore carried out with satisfactory results.

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Idiopathic Myoglobinuria

Idiopathic myoglobinuria is a rare disease in which myoglobin is liberated from muscle tissue and excreted by the kidney. We encountered an example of this condition in a previously healthy young man, and have investigated methods for the detection of the urine pigments.

CASE REPORT

The patient, a youth of 19, had always led a sedentary existence; on the afternoon of September 27, 1959, he played football for the first time for two years. During the game he sustained a kick on the left thigh which, although it did not prevent his finishing the game, left a small bruise. Later that evening he noticed pain and stiffness in the muscles of the lower back and thighs, and weakness of both legs. The pain prevented sleep that night, and by the next morning the weakness of the legs and back had increased to an alarming extent, so that he was not able to sit up unaided in bed. He then noticed for the first time that his urine was dark red in colour.

He was admitted to hospital 18 hours after the onset of symptoms. His general condition was satisfactory, tempera-

ture, pulse, and blood-pressure being normal. Because of profound weakness of the extensor muscles of the back and thighs he was unable to lift himself unaided from the supine to the sitting position, or to raise either leg off the bed. Although the muscles of both thighs were tender on palpation, there was no swelling of the affected muscle groups. Knee reflexes, although diminished, were equal. There was no abnormality of other muscle groups, and examination of other systems revealed nothing abnormal.

Investigations.—The urine passed on admission—that is, 24 hours after the game of football—contained no albumin, sugar, red blood cells, or casts, but was port wine in colour, and contained myoglobin and metmyoglobin. The blood count, serum bilirubin, liver-function tests, and electrolytes were normal, and no excess urinary porphyrins were detected. The plasma creatine and creatinine were normal, but the excretion in the urine showed characteristic changes:

Urine creatinine	.. 1.8 g./litre	} 24 hours after onset of symptoms
„ creatine	.. 0.2 g./litre	
Urine creatinine	.. 0.7 g./litre	} 36 hours after onset of symptoms
„ creatine	.. 0.2 g./litre	

Muscle biopsy from an affected muscle group in the thigh, performed five days after admission to hospital, showed normal voluntary muscle, with no evidence of breakdown of fibres or loss of striation (Dr. K. A. D. Turk).

After admission there was a steady improvement with return of power to the affected muscles; within 24 hours there was no detectable weakness or muscle tenderness, and the knee reflexes had returned to normal. Urine passed on the morning after admission still contained myoglobin although only in about one-twentieth of the concentration in the first specimen. At no time during the illness did he develop oliguria or urea retention.

This type of episode had never occurred previously in the patient or other members of his family. The youth had never been very energetic or taken strenuous exercise. He subsequently admitted to having had similar pain in the back and thighs, and slight weakness of the legs, lasting 24 hours, on two previous occasions, both associated with an infection of the upper respiratory tract. He had not noticed any alteration in the colour of the urine during these episodes.

IDENTIFICATION OF MYOGLOBIN

The identification of the urine pigment is usually made spectroscopically, although Whisnant, Owings, Cantrell, and Cooper (1959) used the ultracentrifuge and paper electrophoresis. The latter test takes several hours and it is advisable to have an authentic sample of human myoglobin for comparison. Both haemoglobin (Hb) and myoglobin (Mb) are rapidly oxidized in urine to their met-derivatives, the spectral characteristics of which are not very different. Direct examination of the urine may therefore show, as in this case, both MbO₂ and metMb, which may not be easy to identify. Berenbaum, Birch, and Moreland (1955) showed that the absorption maxima of HbCO and MbCO were sufficiently separated to permit this method to be used to identify the pigment present in the urine of their case. The spectra of a number of derivatives were therefore compared to see which would provide the best distinction between Hb and Mb.

A Hb solution was used as a control, and absorption spectra were measured with Unicam SP 500 spectrophotometer. The absorption maxima found, and those previously reported, are given in the Table.

The acid met-derivatives (urine, buffered to pH 6, plus 2 drops dil. K₃Fe(CN)₆) did not give sharp maxima in the visible region of the spectrum, possibly because of "background" absorption by other urine pigments. These spectra would be of little use as a diagnostic test, although the Soret bands (406 and 410 mμ) were clearer. The absorption spectra of the met-cyanide derivatives