MELAENA DUE TO ECTOPIC PANCREAS IN ENTERIC DUPLICATION A CASE REPORT

By Denis W. King

INTRODUCTION

Melaena has been caused by both ectopic pancreatic tissue (Madeinavita and Loma 1951, Dabadie *et al,* 1953, Vigdoff and Lewis 1961, Chapmen *et al,* 1947, Huddock *et al,* 1956) and small bowel reduplication (Fauara *et al,* 1971, Gross *et al,* 1952, Basu *et al,* 1960, Mellish and Koop 1961). This paper describes a child with recurrent melaena over a period of four years associated with a combination of the two conditions.

HISTORY

Miss S., age 15, an Indian girl, first presented to the surgical unit in 1968 at the age of 11 with epigastric pain and melaena. The melaena spontaneously settled and no cause was found. Sigmoidoscopy to 25 cm. was normal. No radiological investigations were undertaken. In 1969 she presented again with melaena, but no other symptoms. Barium meal and follow-through and barium enema were normal.

On 29.6.72 she presented with melaena, again with no other symptoms. Her hemoglobin on admission was 5.8 gm.%. Nasogastric aspirate contained no blood and there was no abnormality on physical examination. The melaena gradually subsided on conservative treatment. Barium meal and follow-through examination showed an unusual streak of barium in the right iliac fossa, but was otherwise normal.

After transfusion, elective laparotomy was undertaken in view of the recurrent bleeding and the severity of the current episode. At operation the patient was found to have a duplication of the jejunum approximately 30 cm. in length, the duplicated segment being on the mesenteric aspect of the main channel, sharing a common outer muscle coat, and opening into the main channel distally (Fig. 1). At that opening was a roughened, ulcerated area in the mucosa which subsequently proved to be pancreatic tissue, with ulceration.

Excision of the segment with end-to-end two layer anastomosis was performed. The postoperative course was satisfactory.

From the Department of Surgery, University of Singapore. St. Marks Hospital London EC1.

DENIS W. KING,* M.B., B.S., F.R.A.C.S.,

*Present Address:

St. Marks Hospital London EC 1.

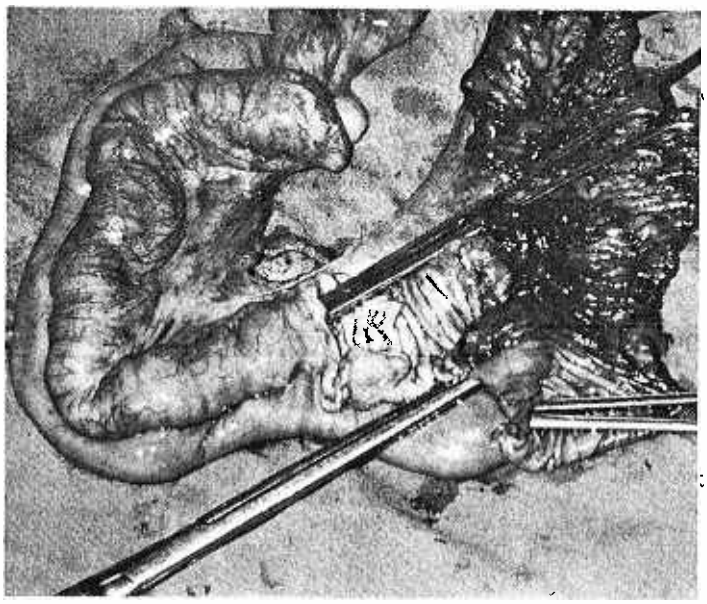

Fig. 1.

DISCUSSION

Duplication anomalies are found most commonly in the ileum, colon, and oesophagus, less frequently in the stomach, and rarely in the jejunum (Rauitch, 1962). About half of the total numbers occur in the ileum (Fauara *et al,* 1971, Mellish and Koop 1961, Gross *et al,* 1952), the majority being cystic, only 7.7% (Fauara *et al,* 1971) being tubular. It is not a common condition, E. L. Potter (1952) finding two cases in more than 9,000 autopsies. Bleeding is not a common presentation, seven out of the 105 cases documented by Fauara *et al* (1971), Mellish and Koop (1961) and Basu *et al* (1960) presenting in this way.

The incidence of ectopic pancreatic tissue varies between 0.6 and 5.6% at autopsy, and is said to be observed in one in every 500 upper abdominal operations. (Barbarossa *et al,* 1946). The ectopic tissue is submucosal in 73%, intramuscular in 17% and subserosal in 10% (Palmer 1951), is in the gastroduodenal region in the majority of cases (Karamandis and Shamnakis 1971, Pearson 1951), and in the ileum in 3% of cases (Pearson 1951).

Radiology is usually unhelpful in the diagnosis of ileal duplication (Gross *et al,* 1952), and in this patient the significance of the abnormal streak of barium in the follow-through films was not appreciated, assuming that it was in the duplicated segment. Radiologically ectopic pancreas is often seen, but usually misdiagnosed (Barbarossa *et al,* 1946), being mistaken for ulcer or neoplasm most often.

Ectopic pancreas usually presents in the fourth to sixth decades (Elfving and Hastbacka 1965), as

either epigastric discomfort or upper gastrointestinal haemorrhage (Chapman *et al*, 1947), and is rarely diagnosed preoperatively (Palmer 1951). Enteric duplication presents in half the cases by the age of five months, and usually by puberty, although it has presented in adults as old as 55 (Slomovitz *et al*, 1943).

The reason for haemorrhage in enteric duplication is unknown, but there are two postulated causes of bleeding from ectopic pancreatic tissue. Ulceration of the pancreatic tissue as in this case, sometimes associated with acute pancreatitis (Vigdoff and Lewis 1961), may cause bleeding. Madeinavita and Loma (1951) have suggested that there may be congestion around the aberrant pancreas leading to haemorrhagic suffusion as the source of blood. Others have suggested enzyme release, possibly causing local vascular damage, as a mechanism (Karamandis and Shamnakis 1971).

It would be interesting to postulate a possible common proposed aetiology for these conditions, as neither can be explained with certainty. Warthin (1904) proposed the most generally accepted theory of pathogenesis of ectopic pancreatic tissue suggesting that it is formed from lateral budding of the rudimentary pancreatic ducts as they penetrate the intestinal wall. The mass of pancreatic tissue is snared off and carried by the longitudinal growth of the intestine. This was supported by Chapmen *et al* (1947) who found Brunners glands overlying ectopic pancreatic tissue in the stomach. They saw also definite anastomoses between pancreatic ducts and Brunners glands in the submucosa.

This theory does not account for those areas of pancreatic tissue found away from the gastrointestinal tract (Crosby and Graham 1932). These may be explained by the development of multipotent cells.

There are a number of theories about the development of enteric duplication, none of which correlates with the theories above.

Bremer (1944) suggested that there may be two methods of duplication. Formation of a traction diverticulum may explain some, but his theory of abnormal fusion of embryonic intercellular vacuoles to produce two channels, the second being cystic or tubular, communicating or not, has gained widest acceptance (Mellish and Koop 1961, Gross *et al*, 1952, Houston and Lynn 1966). According to Mellish and Koop (1961), fragmentary evidence indicates that duplications are a result of environmental stress in the early development of the foetus. That this same stress may produce other anomalies would explain the association of enteric duplication with axial skeletal defects, with intestinal atresia and stenosis, and with a short small bowel.

It has been suggested that the association with axial skeletal defects is in favour of adhesion of neuroectoderm to entoderm producing a traction diverticulum, (Tabnisky *et al*, McLetchie *et al*). The association with intestinal abnormalities is in favour of a common ischaemic aetiology (Fauara *et al*), which would appear to be consistent with Mellish and Koop's theory.

There seems to be general agreement that the treatment of either condition, and presumably therefore of the two together, is excision, although occasionally the pancreatic tissue may be difficult to excise. This patient has progressed well since operation with no recurrent bleeding.

ACKNOWLEDGEMENT

My thanks to Professor S. C. Ong and the Department of Surgery of the University of Singapore for allowing me to report this case.

REFERENCES

1. Barbarossa J. J., Docherty M. B., Waugh J. M.: Pancreatic Heterotopia. S.G. & O., 82, 527-542, 1946.
2. Bartels: Duplications of the Stomach. Amer. Surg., 33, No. 9, p. 747, 1967.
3. Basu R., Forshall I., Rickham P. P.: Duplications of the Alimentary Tract. B.J.S., 47, 477-84, 1960.
4. Bremer J. L.: Diverticula and Duplications of the Intestinal Tract. Arch. Path., 38: 132, 1944.
5. Chapman *et al*: Massive Gastric Haemorrhage Associated with Aberrant Pancreas in the Stomach. G.E., 8: 367-74, 1947.
6. Crosby and Graham: J.A.M.A., 93: 1789, 21.5.32.
7. Dabadie, Berge and Decourt: Bordeaux Chir., 3, 133, 1953.
8. Elfving G. and Hastbacka J.: Pancreatic Heterotopia and Its Clinical Importance. Acta. Chir. Scand., 130: 593-602, 1965.
9. Fauara *et al*: Enteric Duplications—37 Cases: A Vascular Theory of Pathogenesis. Amer. J. Dis. Child, 122: 501-6, Dec., 1971.
10. Fisher H. C.: Duplications of the Intestinal Tract in Infants. Arch. Surg., 61-957-70, 1950.
11. Gross, R. E., Holcomb, G. N., and Farbers: Duplications of the Alimentary Tract. Paediatrics, 9: 449-467, 1952.
12. Houston H. E. and Lynn H. B.: Duplications of the S.I. in Children. Mayo Clinic experience and review of the literature—Mayo. Clin. Proc., 41: 246-256, April, 1966.
13. Huddock J. J., Wanner H. and Reilly C. J,: Ann. Surg., 143, 121, 1956.
14. Karamandis and Shamnakis: Acute Haemorrhage from Aberrant Pancreatic Tissue in the Jejunum. J.R.C.S. Ed. 16,162-5, 1971.
15. McLetchie N. G. B., Purves J. K. and Saunders R. L.: Genesis of Gastric and Certain Intestinal Diverticula and Enterogenous Cysts., 590, 99: 135, 195.
16. Madeinavita J. M. and Loma V.: Revta esp Enferm Apar dig Nutr., 10, 31, 1951.
17. Mellish R. N. P. and Koop C. E.: Clin Manif of Dupl of the Bowel. Paediatrics. 27: 397-407, 1961.
18. Palmer E. D.: Benign Intramural Tumors of the Stomach. Aberrent Pancreatic Tumours. Medicine, 30: 83, 1951.
19. Pearson S: Aberrent Pancreas, Arch. Surg. Chicago, 63, pp. 168-184, 1951.
20. Potter: Pathology of the Foetus and Newborn. Chicago Y.M.P. Inc., p. 314, 1952.
21. Rauitch M. M.: Duplications of the Alimentary Canal—In Paediatric Surgery. Vol. 2, p. 692, Ed. Benson C. D., Mustard W. T., Rauitch M. M., Snyder W. E., Jur and Welch K. J. Chicago. Tear Book Medical Publishers Inc., 1962.
22. Slomovitz Z, Cash I. I. and Enze N.: Gastroent, 11, 528, 1943.
23. Tabnisky *et al*: Duplication of the Stomach: A Cause of Anaemia. Am. J. Gastroent.
24. Vigdoff I. J. and Lewis A.: Calif. Med., 94, 317, 1961.
25. Warthin A. S.: Phys. Surg., 26: 337-351, 1904.