Medical Memoranda

A Case of Familial Incidence of Ectopia Vesicae

Ectopia vesicae is a congenital anomaly of the urinary tract characterized by failure of development of the anterior part of the bladder opening on to the infra-umbilical anterior abdominal wall, which is deficient. In males this anomaly is associated with a rudimentary penis which shows an epispadias deformity of the urethra. The pubic bones are separated at the symphysis by a fibrous band and there is commonly an associated inguinal hernia.

This distressing condition occurs approximately once in every 30,000–40,000 births, and is more frequent in males than in females, the ratio being approximately four to one. It is estimated that in the United States each year a minimum of 100 children are born with this anomaly.

The following case is of interest because it illustrates the occurrence of this condition in two successive male children born to the same parents. We have been able to trace only three other instances of the familial incidence of this condition in the literature. These cases were reported by Higgins (1943, 1958), in the United States of America, and included one case in twins.

CASE REPORT

The mother, aged 28, has been healthy throughout her life and, apart from childish complaints, has had no serious illnesses. She has had no radiological examinations other than mass radiography of the chest some few years before her marriage, and has had no obvious contact with ionizing radiation or radioactive isotopes. The father, also aged 28, is a healthy man and has had no serious illnesses. He has had no radiological examinations other than routine chest x-ray films in 1951 and 1953. He is employed as an aircraft maintenance engineer at London Airport, and has had no contact with radioactive materials. There is no family history of congenital anomalies occurring in either side of the family.

The mother first became pregnant in 1957 after 18 months of married life. The pregnancy proceeded uneventfully and she was delivered of a full-term living male infant weighing 6½ lb. (3.060 g.) on February 14, 1958, after a labour lasting three and half hours. The child showed typical ectopia vesicae, epispadias, and right inguinal hernia. There was no evidence of any other congenital anomaly and the infant thrived. There was no evidence of maternal rubella during pregnancy. The mother became pregnant for the second time in 1959, and the pregnancy proceeded uneventfully except that the foetus presented by the breech at 30 weeks but had undergone spontaneous version by 32 weeks. She was delivered of a living male infant at full term weighing 6½ lb. (3.060 g.) on February 29, 1960, after a labour lasting 12 hours. This child, like the first, showed typical ectopia vesicae. There was no evidence of any other congenital anomaly, and the child thrived. The mother did not suffer from maternal rubella during pregnancy.

The first child was operated on at Great Ormond Street Hospital at the age of 6 months, when the bladder was turned in and the abdominal wall deficiency repaired. At the age of 2½ years he was thriving and was passing urine through the epispadias, but had little or no bladder control.

The second child was operated on at the age of 3 months at Great Ormond Street. It was found that the bladder

was too small to be turned in and implantation of the ureters into an ileal loop and uretero-ileostomy was performed. At the age of 6 months he was in good health. It is hoped at some later date to perform a plastic repair of the bladder, utilizing the ileal loop to enlarge the bladder capacity (ileocystoplasty).

COMMENT

Ectopia vesicae occurring in two successive male children of the same parents is reported. This occurrence is of extreme rarity.

None of the known predisposing factors to the development of congenital anomalies such as maternal rubella, radiological pelvimetry, or exposure to ionizing radiations appear to exist in this case. It is probable that genetic factors play an important causative part in this instance.

The occurrence of more than one case in the same family, although rare, is significant, as a question often posed by the parents is—Doctor, if I have another child, is there a possibility of its having a similar defect? Although familial occurrence is rare, the mother must be told that it is a remote possibility.

LEONARD H. GLASER, M.B., B.S., D.Obst.R.C.O.G.
A. P. ROSSITER LEWIS,* M.B., B.S., D.P.M., D.P.H.

REFERENCES

- Higgins, C. C. (1943). *Cleveland Clin. Quart.*, **10**, 138.
— (1958). *J. Urol. (Baltimore)*, **80**, 279.

Fascioliasis Presenting as a Localized Hepatic Mass

Human infection with the trematode *Fasciola hepatica* is becoming increasingly recognized. It was the first trematode found to be the cause of disease and the first to have its life-cycle described (by Leuckart in 1883). From this description the life-cycle of all digenetic trematodes was elucidated.

Paul (1927) found only 47 cases of human infection. Bürgi (1936) found 186 cases. Facey and Marsden (1960), however, reviewed reports covering several hundred cases throughout the world. They traced 16 cases in Britain and added six cases occurring as an outbreak.

A further case is reported because of the interesting way in which the disease presented. It produced a discrete hepatic mass for which laparotomy was performed, with surgical removal of the mass. Only after repeated examinations of the specimen was an immature fluke found, allowing the diagnosis to be made.

CASE REPORT

An Egyptian physician aged 48 was admitted to Charing Cross Hospital in October, 1959. He had had right upper abdominal discomfort for five months, occurring irregularly and unrelated to meals, with nausea, anorexia, and gradual but marked loss of weight. The bowels were regular. A vague mass was found in the right upper abdomen one month before his arrival in England, and a low-grade pyrexia was noted. Except for four years in Edinburgh, the patient had lived all his life in Egypt.

He was much underweight, and his temperature was 99.5° F. (37.5° C.). In the right upper quadrant of the abdomen was a rounded mass, approximately 7.5 cm. in diameter, lying anteriorly, moving on respiration, dull to percussion, and not tender. The spleen was not felt.

*Since this report was written Dr. A. P. Rossiter Lewis has died.

In four specimens of stool, no pus cells, red cells, ova, cysts, amoebae, or worms were seen. The white blood cells numbered 14,300/c.mm., with 20% eosinophils. The Casoni skin test for hydatid disease was positive on three occasions in Egypt. At Charing Cross Hospital a weakly positive reaction was obtained. Chest x-ray and barium-meal examination, cholecystogram, and excretion urogram were normal, but barium-enema films showed depression of the hepatic flexure of the colon.

At laparotomy there were two partially cystic masses in the right lobe of the liver, the larger being 7.5 cm. in diameter. Partial hepatectomy was performed, including both masses in the specimen. The rest of the liver appeared pale and firm, and the spleen was enlarged; no other abnormality was found in the abdomen.

Depression of the hepatic flexure of the colon, seen on barium-enema radiograph, produced by the hepatic mass containing fasciolae.

Histological section of the specimen showed multiple foci of granulomatous reaction, tuberculoid in type, with a necrotic core, partially coagulative, partially caseous, and liquefying. Some foci contained eosinophils and Charcot-Leyden crystals. Subsequent search revealed a fluke in one of the dilated bile-ducts which, though a good deal smaller than the mature *fasciola*, was confirmed as an immature example of this fluke.

The patient made a rapid recovery and after three weeks was transferred to the Hospital for Tropical Diseases. Repeated stool and rectal snip examinations revealed no ova or cysts, but the fasciola complement-fixation test was strongly positive.

Treatment with chloroquine, 300 mg. twice daily for three weeks, was well tolerated by the patient, his temperature settled almost to normal, and the eosinophil count fell from 20% to 2%.

COMMENT

Although the specimen of fluke was too immature for the species to be identified, it was most likely to be *F. hepatica*. The immaturity explains the absence of a fully developed syndrome and the fact that ova were not found in the faeces, since it takes a minimum of three to four months for the fluke to reach sexual maturity. Had the operation been delayed for a few weeks the patient might have developed higher fever and abdominal pain and have had ova in the faeces.

The localized mass in the liver, depressing the hepatic flexure of the colon, was the first physical sign in this patient. Arenas *et al.* (1948) found hepatomegaly in 38 out of 52 cases.

I thank Mr. Norman C. Tanner, who operated upon the patient, for permission to report this case. Dr. H. I. Winner and Dr. Sidney Shaw performed the bacteriological and haematological investigations at Charing Cross Hospital. Professor W. St. C. Symmers reported upon the specimen

of liver and found the fluke, which was examined by Mr. Pester, of the London School of Hygiene and Tropical Medicine. Professor A. W. Woodruff investigated and treated the patient at the Hospital for Tropical Diseases.

R. M. KIRK, M.B., F.R.C.S.,
Senior Surgical Registrar,
Hampstead General Hospital, London.

REFERENCES

- Arenas, R., Espinosa, A., Padron Ruiz, E., and Andreu, R. M. (1948). *Rev. Kuba Med. trop.*, 4, 92.
Bürgi, K. (1936). *Mitt. Grenzgeb. Med. Chir.*, 44, 488.
Facey, R. V., and Marsden, P. D. (1960). *Brit. med. J.*, 2, 619.
Paul, F. (1927). *Med. Klin.*, 23, 829.

Recurrent Thrombosis of Mesenteric Veins Complicating Cirrhosis of Liver

Although thrombosis of the portal vein occurs in approximately 11% of cases of hepatic cirrhosis (Hunt, 1958; Walker, 1959), thrombosis of the mesenteric veins is uncommon in such cases (Hunt, 1958). When thrombosis of the portal vein occurs in the absence of cirrhosis, portal hypertension results and is not infrequently accompanied by cavernous replacement of the vein. In this condition, which may be difficult to distinguish clinically from cirrhosis, mesenteric venous thrombosis is common; it is given as the cause of death in almost half the reported cases (Klemperer, 1928; Simonds, 1936; Gibson and Richards, 1955; Parker and Seal, 1955). Most of these reported cases had experienced several attacks of abdominal pain during periods varying from a week to several years before death. Various suggestions have been made to account for these attacks.

The following case is one of portal hypertension due to hepatic cirrhosis, in which recurrent attacks of severe abdominal pain had occurred for three months before death from intestinal infarction. The necropsy findings in this case indicate that these attacks were due to recurrent bouts of thrombosis in the mesenteric veins.

CASE REPORT

A man aged 39 was first admitted to hospital on January 7, 1959, with a 24-hour history of jaundice and intermittent attacks, for two months, of low back pain, anorexia, and nausea with some vomiting. He had had vague abdominal discomfort for several years but no previous episodes of jaundice. There was gross hepatosplenomegaly, and this, together with the laboratory findings, suggested a diagnosis of virus hepatitis superimposed on hepatic cirrhosis of obscure origin. The jaundice gradually cleared, but one week after discharge he was readmitted with pneumonia; this responded well to treatment.

During the next three months he was subject to attacks of abdominal pain occurring at intervals of about a week. It came on suddenly in the epigastrium, radiated through to the back, and was severe and continuous, lasting about two hours; he vomited frequently. Between episodes he had vague intermittent discomfort in the right hypochondrium.

On May 15 he had an attack of pain and vomiting which persisted. It was identical with previous attacks except that it was more severe; after several days he passed blood per rectum, and was readmitted to hospital five days after the onset.

At laparotomy (Mr. J. Hadfield), 18 in. (46 cm.) of the lower ileum was gangrenous, and was resected. The veins in the excised mesentery were thrombosed, but the arteries were patent. Twenty-four hours after operation severe abdominal pain recurred; large amounts of blood-stained