HAEMANGIOMATA OF THE INTESTINE

BY

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The case history of a child who for five years had had repeated attacks of bleeding from the bowel due to multiple haemangiomata of the intestine is presented. It is thought worthy of record because of its unusual nature and the method of treatment employed.

Haemangiomata of the intestine are uncommon causes of intestinal bleeding, and, when multiple, set a difficult problem in treatment. The first recorded case was published by Gascoyen in 1860, his patient having haemangiomata in the bowel wall, liver, parotid gland, and skin. Series of cases collected from the literature and descriptions of new lesions were published by Brown (1924), Kaijser (1937), Lazurus and Marks (1945), and Hansen (1948); and, so far as can be ascertained, the number of reported cases of haemangiomata of the intestine is now 85.

The ages of these 85 patients (54 males, 31 females) ranged from 2 months to 81 years, and 44 had single and 41 multiple lesions, the distribution of which was as follows:

	Small Bowel	Large Bowel	Large and Small Bowel	Not Stated
Single lesions ..	27	16	—	1
Multiple lesions ..	28	3	10	—

Types of Haemangiomata

Kaijser (1937) divides haemangiomata of the intestine into five types: (1) Multiple pin-point varicosities consisting of dark bluish-red nodules which vary in size from a pinhead to a pea and are scattered over a limited portion of the intestine. They are usually situated in the submucosa. Each nodule is made up of communicating cavities, which connect with the smaller veins. They may not be true tumours but the result of venous back pressure, and are usually symptomless. (2) Diffuse infiltrating cavernous haemangioma in which the cavernous tissue usually involves all layers. The bowel wall is thereby thickened and the lumen narrowed. A common site for this type of lesion is the rectum, and haemorrhage from the bowel a frequent occurrence. (3) Circumscribed cavernous haemangioma. This type if often polypoid. (4) Capillary haemangioma, which usually appears as a round submucous tumour, up to a plum in size, protruding into the canal. (5) Multiple haemangiomata, which include tumours varying in structure from the usual cavernous type to one rich in cells and stroma. They may be associated with haemangiomata in other organs.

Symptoms

Though in some instances there are no symptoms referable to the intestinal tract, the most common symptom is that of bleeding from the bowel, the incidence of haemorrhage being higher in those in which the lesion is in the large intestine. The loss may consist of dark altered blood or of bright red blood, which may be profuse enough to endanger life. The bleeding is often undetected at first, the patient being treated for anaemia, perhaps severe and of long standing. A feature of many of the reported cases is that the patients have been several times in hospital for anaemia, sometimes with a haemoglobin level of below 20%, and have required treatment by blood transfusion, abnormality of the gastro-intestinal tract not being discovered until revealed by laparotomy several years later. Other symptoms that may be caused are those of obstruction (as by intussusception) or of inflammation.

The diagnosis is rarely made pre-operatively, but it can be emphasized that if a patient presents with a history of repeated attacks of melaena, often since infancy, if there is marked anaemia, and if no abnormality can be discovered in the gastro-intestinal tract then the possibility of haemangioma of the bowel should always be considered. The presence of other haemangiomata—for example, in the skin—strengthens the suspicion and was helpful in the diagnosis in our case. Other conditions which may give a similar clinical picture are gastric or duodenal ulcer, peptic ulceration in a Meckel's diverticulum, intestinal polyp, haemophilia, and purpura.

Treatment

The most successful method of treatment has been surgical excision, particularly of the single lesion, the involved length of gut being resected if the lesion was not too extensive. So far as can be ascertained from the literature, in only one case of multiple lesions of the small and the large bowel has surgery been attempted. In this case, described by Lazarus and Marks (1945), there were multiple haemangiomata of both small and large intestines. It was decided that most of the bleeding was from lesions in the large bowel, and colectomy was performed in two stages. There had been no further bleeding when the patient was seen about one year after operation.

Other methods of treatment which have been employed are cauterization, partial extirpation, irradiation by *x* rays or by the use of implanted radon seeds, and injection of coagulating substances.

Case Report

The patient, a female child aged 6 years, was first seen by one of us on April 6, 1950, on account of recurrent attacks of pallor. The child's delivery had been normal, and her birth weight was 9¼ lb. (4.2 kg.). It was noticed at the time that she had six toes on both sides, and the supernumerary toes were removed at the age of 2 weeks. At 1 month it was noted that the child had a small lump over the left chest wall resembling a cyst about 1 cm. in diameter: the skin over it was of normal colour. This lump continued to grow until, when the child was 16 months old, it was 5 cm. in diameter. During the next few months three attempts at excision were made, but it was clear that the condition was a haemangioma; bleeding was troublesome, and only partial removal was effected.

When the child was 3 years old the mother observed that she seemed to be very pale, but, in thinking back, remembered that the child had always been rather pale. At the age of 3½ the haemoglobin was 33%, and liver injections were given, the haemoglobin rising to 80%. At about this time a second lump was noticed near the spine, and at the age of 4 a course of *x*-ray therapy was given to both swellings, followed by injections of saline into the lumps. This caused both swellings to shrink. Since that time the lump

near the spine had disappeared, but the tumour over the chest wall remained much the same size. From the age of 3½ onwards there were periods during which the child grew very pale: sometimes a tinge of yellow was noticed in the conjunctivae. After a few weeks the colour gradually returned.

On examination the patient was pale, but there was no icterus. The liver was enlarged to two fingerbreadths and the tip of the spleen could be felt. Over the left chest wall in the mid-axillary line there was a subcutaneous cavernous haemangioma about 8 by 8 cm. Both feet were misshapen, and a radiograph revealed a congenital abnormality of the first metatarsal bones.

Blood investigations showed: haemoglobin, 37%; red cells, 2,110,000; haematocrit, 20%; M.C.V., 95 μ^3; M.C.H.C., 27%; reticulocytes, 20%; white cells, 4,400, with a normal differential. The fragility test was normal and the Coombs reaction was negative.

Progress

The child was admitted to hospital, and examination of the stools consistently showed the presence of occult blood. It was considered that the intermittent attacks of anaemia were due to intestinal bleeding, the possible differential diagnosis being between a peptic ulcer, Meckel's diverticulum, and haemangiomata of the gut. A barium meal showed no abnormality. The child was given a small blood transfusion and was then sent home on an ulcer regime to see whether this would stop any further bleeding.

Within a week she was readmitted because of severe pallor, the haemoglobin having fallen to 25%. A further blood transfusion was given, and a week later the child again returned home. Twice the sequence of pallor, low haemoglobin, and transfusion was repeated, and it was clear that medical treatment for a possible peptic ulcer was having no effect on the disease and that the most likely diagnosis was bleeding from a Meckel's diverticulum or from haemangiomata of the gut. Laparotomy was therefore considered.

On admission to hospital on August 17 the child appeared to be well, the haemoglobin level being 50%. Further blood investigations showed: M.C.V., 80 μ^3; M.C.H., 24.7 $\mu\mu$g.; M.C.H.C., 31.2%. It seemed that the red cells were larger than normal, so a marrow puncture was carried out, but nothing abnormal was found. A repeated barium meal, barium enema, and gastric analysis were normal. All specimens of motions contained a large amount of occult blood.

Suddenly on August 28 the child became very pale and the haemoglobin dropped to 33%. A blood transfusion was given, and in consultation with Professor Bentley it was decided to carry out operative treatment.

Treatment

On September 8 an operation was performed by Professor F. H. Bentley (anaesthetist, Dr. Philip Ayre). A right paramedian incision was made. Multiple small haemangiomata were seen at intervals along the whole length of the small bowel, varying in size from 2–3 mm. up to 1 cm. in diameter. The tumours were dark blue in colour and situated apparently in the submucosa at no regular position in relation to the circumference of the bowel. A few similar lesions were scattered throughout the large intestine.

The only available treatment was to open the bowel in many places and remove the tumours. The procedure was begun at the proximal end of the jejunum: at the site of each haemangioma a short incision was made longitudinally into the antimesenteric border of the bowel, the haemangioma clamped, the base ligated with thread, and the tumour excised. The incision in the bowel was then sutured in two layers.

Sixteen such incisions were made and 19 haemangiomata removed. At this point it was decided to stop, leaving about another dozen haemangiomata to be removed at a later stage from the lower end of the ileum.

Immediate post-operative recovery was uneventful, but a week after this first operation the patient began to bleed again, and occult blood in the stools remained strongly positive. The haemoglobin, which had been as high as 83%, now fell to 49%. It was considered that the next stage of the operation should not be further postponed.

The second operation was performed by Professor F. H. Bentley on September 27 (anaesthetist, Dr. Philip Ayre). A right lower paramedian incision was made. Seven more haemangiomata were removed by a similar method to that used at the previous operation, and two further tumours of larger size plus several smaller angiomata were excised by resection of a 5-cm. length of small bowel, with end-to-end anastomosis. Severe adhesions were noted from the previous operation.

Post-operative recovery was again uneventful and the child's general condition improved, the haemoglobin rising spontaneously to 93%, but the occult blood in the stools still remained positive. The child was discharged home on October 19, with the intention of removing the few tumours from the large bowel at a later date if this proved necessary. Two months later the child was well and the haemoglobin level was 85%.

Summary

The literature on haemangiomata of the bowel is reviewed.

A case of multiple haemangiomata of the small and large bowel in a child is described. For six years the patient had suffered from recurrent severe anaemia due to intestinal bleeding. At operation 30 separate haemangiomata were removed from the small intestine.

Haemangiomata of the bowel should be included with other well-known causes of intestinal bleeding in the differential diagnosis. The presence of subcutaneous haemangiomata, as in this case, strengthens the likelihood of intestinal vascular tumours. Operative removal is the only effective remedy.

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Mr. Hilary Marquand, Minister of Health, recently welcomed teams of medical and social workers who have come from seven European countries to study the treatment and care of disabled children in Britain. Speaking at a luncheon at the Royal National Orthopaedic Hospital, Stanmore, Middlesex, he paid a tribute to the work being done at the hospital, and said that it was being run on the basis that disabled persons asked not for pity but for opportunity. "The whole conception of this and similar hospitals," he added, "is to rehabilitate the patient so that he or she can take their place as far as possible in the normal life of the community. The accent is not on pity or sorrow for suffering, but on rehabilitation—the opportunity to make a new life by overcoming physical handicap." The Minister and the European visitors examined the many ways in which children and adults, after receiving surgical treatment, are helped to overcome disabilities mainly affecting arms and legs. This international study course has been organized by the World Health Organization and the United Nations International Children's Emergency Fund. Its members, who will spend eight weeks in Britain, include orthopaedic surgeons, physiotherapists, occupational therapists, vocational training instructors, technicians, teachers, and social workers.