Intestinal and Cutaneous Hemangiomatosis

Report of a Case

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HEMANGIOMAS, benign intestinal tumors, are rare, their incidence being 1 in 14,000 admissions.1 Bleeding of these tumors may cause a prolonged and unexplained chronic hypochromic anemia.

In some rare instances intestinal hemangiomas are associated with similar tumors of the skin. Only 19 such associations have been reported in the literature.1-14

The rarity of the condition and diagnostic difficulties associated with it prompted us to report the following case.

CASE REPORT

A.Z., an 18-year-old male, was admitted because of severe anemia. At the age of 12 years a cavernous hemangioma was removed from the skin of the right anterior chest. From the age of 14 years, weakness, pallor, and pica appeared and hypochromic anemia was detected. Irregular therapy with iron, Vitamin B12, and liver extract was administered. In spite of this the hemoglobin decreased and the patient was admitted to another hospital. Except for pallor, no pathologic findings were revealed. Hemoglobin was 8.7 gm.%, R.B.C., 3.6 million/cu. mm.; W.B.C., 6400/cu. mm., with normal differential count; and reticulocytes, 0.8%. E.S.R. and liver function tests, urine examination. E.C.G., X-ray of the chest, and I.V.P. were normal. One benzidine test of the feces was positive (without previous diet). The patient was discharged with a diagnosis of hypochromic anemia of unknown origin.

At home weakness and pica persisted and, after 8 months, the patient was re-admitted. Again the physical examination was negative: Hgb., 9.3 gm.%; reticulocytes, 3%; stomach juice, normal acidity; bone marrow, no iron pigment in macrophages. The diagnosis was iron deficiency anemia of unknown cause.

The patient continued to complain of weakness and pica. He noted subjective improvement during periods of extensive iron and liver extract therapy, and a change for the worse when the therapy was stopped. There was no bleeding and he had no abdominal complaints. The color of the feces was always normal.

A few weeks prior to admission severe weakness and pallor appeared. The site of the removed skin tumor swelled and was painful on palpation. The patient was very pale and in good general condition. On the skin of the right anterior lower chest a soft tumor was felt, exactly in the region of the previous operation. The spleen was not palpable and the liver edge was felt 2 cm. below the costal margin. There were no other findings.

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Rectal examination and sigmoidoscopy were normal. Blood urea and glucose, proteins, and electrophoresis, urine, and general feces examinations were normal. Stomach juice showed free and total acidity of 32 and 41 mEq./L., respectively. Examinations for occult blood in the feces (with the patient on a meat-free diet) were positive on five of seven occasions. X-ray examination of the chest, skull, pelvis, extremities, I.V.P. and barium enema examinations were normal. An upper G.I. series revealed a round filling defect in the first part of the duodenum, with polyp or aberrant pancreas suspected (Fig. 1).

Hematologic study showed: Hgb., 7.0 gm.%; R.B.C., 2.5 million/cu. mm.; and W.B.C., 7500/cu. mm., with normal differential count. Reticulocytes were 1.7%; platelets, 112,000/cu. mm.; and PT, 100%. Bleeding and clotting time, clot retraction, and prothrombin consumption were normal. Serum iron was 47 µg.%; iron saturation, 41%; and iron-binding capacity, 67 µg.%. Total iron binding capacity was 114 µg.%, and serum vitamin B₁₂, 450 µg./ml. A blood smear revealed hypochromic and faintly polychromatophilic anisocytosis; bone marrow-hypercellular bone marrow with inverted M/E ratio; and extremely low iron content. Mechanical and osmotic fragility were normal and Coomb's tests negative. Glutathione was 46 mg.%. In hemoglobinophoresis (agar, starch, and paper), normal A and F and 2% of A₂ Hgb. were detected. Blood volume was 62 cc./kg, and red cells volume 24 cc./kg.

Investigation of G.I. tract blood loss with Cr³⁺-labeled red cells revealed a blood loss of 6 cc. in a period of 3 successive days (2-3 times normal in this laboratory).

The parents of the patient and a 21-year-old brother are Jews born in Poland and healthy. There was no consanguinity.

Iron dextran, ferrous sulfate, and folic acid were administered. The Hgb. rose to 15.3

Fig. 1. A cavernous hemangioma; note round lesion in first part of duodenum.