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## Medical Memorandum

### Recurrent Intussusception Due to Polyposis of the Gastro-intestinal Tract Associated with Pigmentation

The intussusception, gastro-intestinal polyposis, and pigmentation of skin and buccal mucosa sign-complex was first described by Jeghers (1944) and further elaborated by Jeghers *et al.* (1949). Tanner (1951) reviewed the literature and added a further case. Since then, so far as we have been able to ascertain, only two more cases have been reported (Schaffer and Sachs, 1952; Wolff, 1952), bringing the total number of recorded cases to 27. The following case, to the best of our knowledge, is the first to be reported from the Middle East.

The salient features of the syndrome, as borne out by all the reported cases, are: (1) The patients are usually of dark complexion. (2) Pigmented spots are present on the buccal mucosa, lips, and face, and sometimes on the digits. The spots vary in colour from "light brown on the face to dark blue-black on the mucosa" and have been shown to contain melanin. (3) The presence of gastro-intestinal polyposis, predominantly in the small intestine. The polyps are usually benign, but in one case malignant change has been reported. (4) The condition usually presents as recurrent intussusception in the second decade of life. (5) It is often familial, and is believed to be transmitted as a Mendelian dominant.

#### CASE REPORT

A boy aged 16, who was born in Turkey and had come to Israel in 1950, was admitted to the department of surgery on October 1, 1952, complaining of generalized abdominal pain associated with anorexia and vomiting, following the passage of a hard stool some four to five hours previously. The pain was colicky in nature, maximal in the epigastrium, and radiated to the left flank. Nausea was persistent and he had vomited several times after the onset of the pain. The vomitus contained undigested food. No blood was present in the stools and there was no history of recent diarrhoea. He had had an almost identical attack two weeks previously, when he was admitted to another hospital and discharged 48 hours later, his symptoms having subsided.

He had been well until 18 months earlier (March 8, 1951), when he was admitted to this hospital complaining of abdominal pain, constipation, vomiting, and the passage of red blood in his stools. On laparotomy an "ileo-ileal intussusception with a polyp about the size of an apricot" was found. The pathological report was that of a "benign polyp of the small gut." On discharge from hospital he continued to have attacks of abdominal pain with occasional bloody stools at approximately weekly intervals.

On August 9, 1952, he was readmitted with abdominal pain, constipation, vomiting, and blood in his stools. A laparotomy was again performed the day after admission, and "ileus due to adhesions related to the ileum and herniation of bowel through a deficiency in the omentum" was reported. After his discharge (nine days later) he was relatively well until the attack two weeks before his present admission to hospital. His mother, father, two brothers, and two sisters are all healthy and have no history of abdominal symptoms or operation.

Examination revealed a dark-complexioned youth, physically underdeveloped for his age—weight 73 lb. 11 oz. (33.4 kg.), height 4 ft. 10 in. (147 cm.). No pubic or axillary hair was present. The genitalia were small and undeveloped and the voice was high-pitched. Small pigmented spots about the size of a pinhead were present over the bridge of his nose and around his mouth. Larger bluish-black macular spots varying in size from 1/16 to 1/4 in. (1.6 to 6.4 mm.) in diameter were present on his lips and buccal mucosa, particularly the lower lip and the inside of the left cheek. No other pigmented spots or papillomata were found on the rest of the body surface. In the abdomen a sausage-shaped mass extending from the left flank to the left of the umbilicus was clearly palpable. Generalized abdominal tenderness was present, but no guarding or rigidity. No free fluid was detected, and the liver and spleen were not palpable. Rectal examination was negative.

A diagnosis of intussusception was made. At laparotomy a jejuno-jejunal triple intussusception was found involving about 1.5 m. of small bowel, with a large polyp (about 1/2 in. (1.9 cm.) in diameter) at the tip of the intussusception. Resection of 10 cm. of bowel, including the polyp, was performed and the specimen sent for section. The pathological report was of a "benign adenomatous papilloma."

The patient made an uneventful recovery. A barium meal and follow-through, and a barium enema at a later date, showed no radiological evidence of further polyposis. On rectoscopy, however, a cluster of four sessile polypi, each about 1/4 in. (6 mm.) in diameter, was seen on the left lateral rectal wall 9 cm. from the anal verge.

A subsequent examination of the patient's parents and siblings revealed no pigmentation of the buccal mucosa or skin in any of them, nor signs of polyposis of the bowel.

We wish to express our thanks to Dr. N. Rabinowitz, of the department of surgery, Hadassah University Hospital, for permission to report this case, and to Dr. Charlotte Kaplan for drawing our attention to this interesting but rare syndrome.

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Since its inception in 1947 the National Corporation for the Care of Old People has devoted its resources principally to assisting voluntary organizations by means of grants-in-aid to provide homes, clubs, and welfare centres. Its annual report for 1951-2 records that £116,515 was allocated in this way during the year, making a total of £790,000 during the last five years. The Corporation opened a rest home at Stanmore to admit patients discharged from the geriatric unit of West Middlesex Hospital and from Edgware General Hospital. Another home was opened at Greenisland, near Belfast, and a third is planned for Oxford. An experiment in Bristol was the provision in 1951 of a laundry service for old people. The Corporation, which has power to accept trusteeship of charitable bequests made to benefit old people, pleads that they "should not be regarded as a class apart, as objects either of pity or charity, but as people who with help can retain their independence and make their own contribution to the general welfare." The welfare of old people is also the concern of a special committee of the National Council of Social Service, whose annual report for 1951-2 records its activities. It makes available to Government departments, voluntary societies, students, and others up-to-date information and experience affecting the welfare of old people. At its national conference at Scarborough in 1952, 400 delegates heard Sir Walter Monckton outline the steps taken by the Government to ensure a new and sympathetic approach to the employment of the elderly on the part of employers, trade unions, and the public.