

At laparotomy on May 30, 1949, an ileo-ileal intussusception was reduced; no resection. On April 20, 1950, laparotomy revealed an irreducible ileo-ileal intussusception; 12 in. (30 cm.) of ileum was resected.

In November, 1950, and January and July, 1951, he was admitted with intestinal obstruction and intussusceptions were palpated, but spontaneous reduction occurred shortly after admission. He often suffers from both mild and severe attacks of colic, but so far has required no further admission to hospital.

CASE 2

A boy aged 12 attended in February, 1951, with a rectal polyp. He had had an operation for intussusception in 1939. There was no relevant family history. He was a sallow child with typical pigmentation of the lips and oral mucosa. A polyp was removed from the rectum. Barium enema and sigmoidoscopy revealed no further polyps at that time. A second polyp was removed in July 1951, and in May, 1952, examination revealed a third rectal polyp and mass in the ascending colon.

An exploratory laparotomy on July 9, 1952, revealed a polyp in the jejunum, 12 in. (30 cm.) from the duodeno-jejunal flexure, a polypoid mass in the caecum, a polyp in the appendix, a polypoid mass in the descending colon, where a chronic intussusception was present, and a polyp in the sigmoid colon; the mucosa of the terminal ileum was nodular, but no true polyps were present. Resection of 6 in. (15 cm.) of upper jejunum and total colectomy, anastomosing the lower ileum to the rectum, were carried out. The pathological report stated "benign adenomatous polyps."

Post-operative progress was complicated in October, 1952, by intestinal obstruction due to adhesions, which were divided. No further polyps have developed in the 4 in. (10 cm.) of rectum remaining.

I wish to thank Mr. Ridley Thomas and Mr. Norman Townsley for permission to publish the histories of these patients, who are under their care, and for their help and advice.

R. M. T. WALKER-BRASH, B.M., F.R.C.S.,
Registrar, Norfolk and Norwich Hospital.

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Intestinal Polyposis associated with Pigmentation and Intussusception in Triplets

Hunter and Wilson (1953) define two types of true adenomatous polyps of the small intestine: (1) those associated with pigmentation of the lips and buccal mucosa and with familial tendencies; and (2) those not associated with pigmentation and no obvious hereditary factor.

In view of the rarity of this condition the following cases of type 1 occurring in triplets are considered worthy of note. All three presented initially with abdominal colic and all showed marked pigmentation of the lips and buccal mucosa, including the gums; all were of sallow complexion and had dark hair.

CASE 1

The patient was first seen on November 17, 1944, when aged 8. For three years, on and off, she had complained of colicky abdominal pain associated with diarrhoea and vomiting. Marked circumoral and buccal pigmentation was noted along with a patchy brown pigmentation of the abdominal wall. A tentative diagnosis of Addison's disease was made, and she was admitted for further investigation. Full blood examination showed only very slight leucocytosis; blood sodium 307 mg. per 100 ml.; and blood potassium 24 mg. per 100 ml. X-ray examination showed no suprarenal calcification and the Kepler water test was normal.

A diagnosis of mild Addison's disease was made, and she was discharged on January 15, 1945.

She was readmitted on February 10 with symptoms of acute intestinal obstruction. At laparotomy (A. L. L.) 4 feet (122 cm.) of gangrenous ileum was seen and was thought to be due to volvulus. Resection with end-to-end anastomosis was performed. Some calcified tuberculous glands in the mesentery were included in the resection. She was seen again on April 9, complaining of intermittent abdominal pain, but full x-ray examination showed no abnormality.

On March 10, 1953, she was readmitted with very severe abdominal pain and symptoms of intestinal obstruction. Examination revealed a large sausage-shaped swelling in the lower and right abdomen. A diagnosis of small-bowel obstruction was made and laparotomy was performed (W. P. C.). An ileo-ileal intussusception extending through the ileo-caecal valve was found. Reduction was impossible, and a right hemicolectomy was performed. A large, simple adenomatous polyp was found to have initiated the intussusception. Convalescence was uneventful, and since discharge she has been seen several times and has remained perfectly well.

CASE 2

This sister was first seen on April 22, 1948, when aged 12, with symptoms of acute small-bowel obstruction. Circumoral and buccal pigmentation was noted. Laparotomy (A. L. L.) revealed an ileo-ileal intussusception 4 in. (10 cm.) in length. This was reduced and an intestinal polyp $\frac{1}{2}$ in. (1.9 cm.) in diameter was palpated through the bowel wall. The polyp was excised and the bowel sutured transversely. Histological examination showed inflammation of an adenomatous polyp. She was readmitted on May 26, complaining of left lower abdominal pain, but sigmoidoscopy and barium examination revealed no abnormality.

On March 28, 1949, she was again admitted with severe lower left abdominal pain associated with vomiting. At laparotomy a colo-colic intussusception of the sigmoid loop was demonstrated. This was easily reduced, and was found to be initiated by a polyp palpable through the bowel wall. Several other polyps were palpable further up the large bowel as far as the transverse colon. A Paul-Mikulicz resection of the sigmoid colon containing the intussuscepting polyp was performed. The double-barrel colostomy was later successfully closed.

CASE 3

This girl is the only one of the triplets who has not required laparotomy. Nevertheless she was seen on April 9, 1951, aged 15, complaining of colicky abdominal pain. At examination she, too, had marked circumoral and buccal pigmentation. In view of her sisters' history full barium x-ray examination and sigmoidoscopy were performed, but no evidence of polyposis could be demonstrated.

COMMENT

The association of circumoral and buccal pigmentation with polyposis of the small intestine and intussusception, though rare, is by now a well-recognized syndrome. The familial tendency of the condition is well demonstrated in these triplets, but no history of similar pigmentation or intussusception could be elicited from the parents or other members of the family.

Hutchinson (1896) first described pigmentation of the lips and buccal cavity in twins, and Parkes Weber (1919) recorded that one had died of intussusception. Jeghers (1944) and Jeghers *et al.* (1949) described the syndrome more fully and recorded 10 further cases. Tanner (1951) added a further case, and Wolff (1952) reviewed the literature and recorded a case of his own. Kaplan and Feuchtwanger (1953) and Kitchen (1953) added three further cases. Hunter and Wilson (1953) recorded yet another case of adenomatous polyp of the small intestine associated with pigmentation and intussusception, and suggested a classification of intestinal polyps. So far as can be ascertained 31 cases have been recorded.

As stated by Hunter and Wilson (1953) the pigmentation occurs early in childhood on the lips and buccal mucosa. It may also occur on the digits, though not in the cases here described. Patchy brown pigmentation was present in the umbilical region of Case 1. Papillomata may occur in the stomach, duodenum, small intestine, or colon, but the small intestine is most frequently involved. In Case 2 both small and large intestines were involved.

There would appear to be no connexion with congenital polyposis coli, as the finding of polyps in the small intestine has never been described in that condition, nor has buccal pigmentation.

Case 3 has, as yet, not required surgical intervention, but the association of marked pigmentation of the lips and mouth with frequent attacks of abdominal colic is strong presumptive evidence of the presence of intestinal polyps.

A diagnosis of intussusception can be made with confidence when there are symptoms of intestinal obstruction and when buccal pigmentation is present.

W. P. CRONE, M.B., F.R.C.S.,
Surgical Registrar, Wakefield A and B Groups.
A. LAWSON LIGHT, M.D., M.S., F.R.C.S.,
Consultant Surgeon, Wakefield A and B Groups.

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A Case of Methyl Chloride Poisoning

Poisoning from methyl chloride is fortunately of rare occurrence despite the fact that, during recent years, there has been a great increase in the use of this substance as a refrigerant. It is, however, extremely dangerous, and, in order that the risks of exposure should be more widely known than they are, the following case is reported.

CASE REPORT

A foreman electrical engineer aged 52 was admitted to Victoria Hospital, Kirkcaldy, on August 7, 1950. The history was that from August 4 he had been engaged in repairing a faulty commercial refrigerator. On the 5th he experienced a feeling of nausea, and in the evening vomited on several occasions. Next day he felt light-headed and dizzy, and his gait was unsteady. He continued at work, however, but on returning home his wife became alarmed at his condition and called in his doctor. His son, who also worked on refrigerator maintenance, recognized the peculiar odour of methyl chloride from his breath.

When he was admitted to hospital his general appearance was that of a drunken man. He was drowsy, his speech was thick and slurred, his gait unsteady, and pulse 100 but regular in time and force. There was slight cyanosis and his blood pressure was 160/104. Mentally he was confused and unable to give a detailed account of himself. Physical examination revealed no abnormality. Laboratory findings: Urine, S.G. 1005; definite acetonuria; no sugar or albumin. Blood: Hb, 88%; plasma protein 7 g.% (albumin 4.20 g., globulin 2.80 g.); blood urea, 50 mg.

Copious dextrose drinks and sodium bicarbonate, 30 gr. (2 g.), were given four-hourly. There was no further vomiting, and his general condition improved greatly during the next two days.

On August 10, however, he became confused and irrational, and the next day he was vividly hallucinated both aurally and visually, and expressed persecutory ideas. He stated that voices were shouting at him from the ceiling and walls, accusing him of being a spy, that microphones were picking up his thoughts and broadcasting them by means of transmitters in the roof. He accused the nurses and doctors of giving him injections in order to control his

thoughts and interfere with his genitals. He stated that policemen were staring at him through the windows and discussing how to dispose of him. He was extremely agitated and excited, and adopted a threatening attitude towards the nursing staff and refused to co-operate in his treatment. Arrangements were therefore made for admittance to Stratheden Mental Hospital, Cupar, Fife, and while being transferred by ambulance he made a vicious and unprovoked assault on a male escort. At the time of admission he was so aggressive and truculent that a physical examination was impossible, and, following the administration of 10 ml. of paraldehyde by intramuscular injection, he settled down and slept soundly until the next morning.

On awakening he was quite rational and was able to give a lucid account of himself. Although his memory for the preceding two days was rather hazy he realized he had given much trouble and was most apologetic. His persecutory ideas had now completely gone, and, although he remained hallucinated aurally, he realized that the voices were imaginary. He complained of double vision and admitted having had difficulty in focusing for several days. He stated that while working in the refrigerator he was repairing he realized he was inhaling methyl chloride fumes, but liked the peculiar odour of this substance and therefore did not take the precaution of wearing a mask which was available. The space in which he was working measured approximately 8 by 5 ft. (2.4 by 1.5 metres), had no ventilation, and, so far as he was aware, a high concentration of methyl chloride was present. Physical examination revealed no abnormality and his urinary output was satisfactory. Laboratory investigation showed a marked acetonuria but no albumin or sugar; Hb, 85%; red cells, 4,250,000; white cells, 14,000, with a normal differential count. His previous treatment was continued and progress thereafter was uninterrupted. He was discharged from hospital on August 24.

DISCUSSION

Methyl chloride (CH_3Cl) is a colourless gas, but is used as a refrigerant in liquid form. It has a weak but not unpleasant odour, is only faintly irritant, and does not injure the cornea. It is slightly narcotic, but the general toxic action is severe. There are, however, delayed toxic effects which may occur after apparent recovery. Absorption is rapid, but elimination is slow, and it is considered that concentrations of 100 parts per 1,000,000 are extremely dangerous (Jacobs, 1949). Most cases of poisoning due to this substance have occurred in America, where, in all, 50 cases have been reported. Roth described 10 cases in Switzerland in 1923, and Schwarz recorded 9 cases in Germany in 1926. In Britain seven cases have been reported (Lehmann and Flury, 1943).

The toxic symptoms of methyl chloride are dizziness, restlessness, drunkenness, apathy, anorexia, nausea, vomiting, diplopia, tremors, unsteady gait, psychic disturbances, and convulsions. Delayed symptoms and even death can occur two to three weeks after apparent recovery. It is also stated that death can occur without previous symptoms of poisoning. The delayed toxic action is most likely due to a breakdown product of the methyl chloride molecule in the form of phosgene, formaldehyde, or formic acid.

The post-mortem findings which have been reported include cerebral oedema and minute haemorrhages in the brain, lungs, liver, kidneys, and heart. Degenerative changes are often found in the liver and kidneys (McNally, 1937).

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M. M. MACRAE, M.D., D.P.M.
Stratheden Hospital, Cupar, Fife.

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