

# Trichobezoar, gastric polyposis, protein-losing gastroenteropathy and steatorrhoea

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**SUMMARY** A mentally subnormal patient presenting with oedema was found at gastroscopy to have a large trichobezoar and multiple gastric polyps. The serum concentrations of albumin and IgG were low in the absence of proteinuria, and the gastrointestinal clearance of radiochromium after the intravenous administration of radiochromic chloride was increased. These findings are compatible with increased gastrointestinal loss of plasma proteins. In addition, the patient had steatorrhoea. All of these abnormal findings were markedly improved after operative removal of the bezoar.

A trichobezoar is a mass of hair, wool, and similar material found in the stomach, and less commonly in the intestine, of some patients with abnormal appetites. The adolescent girl with long, flowing hair and the mentally subnormal feature prominently among those afflicted.

Complications are frequent. Gastric ulceration, haemorrhage, perforation, and intestinal obstruction are among the better known. The association between bezoar and gastric polyposis, though well documented, is not so widely appreciated (Davies, 1921; Charache, Polayes, Behr, Murata, and Dimetriades, 1957; Valberg, McCorriston, and Partington, 1966). In one case, one of the polyps had become malignant (Charache *et al.*, 1957).

Ankle swelling as a presenting symptom was first mentioned by Davies in 1921. Plasma proteins were not measured. Wine, in 1957, described a patient with gastric trichobezoar, oedema, and hypoproteinaemia. An insight into the pathophysiology of this complication was obtained by Valberg and his colleagues in 1966. Using <sup>131</sup>I-labelled human albumin and polyvinylpyrrolidone, they demonstrated excessive gastrointestinal protein loss in one patient, and their results, though incomplete, suggested that it occurred in another patient as well. Gastrointestinal protein loss returned to normal after operative removal of the bezoar.

The patient reported here presented with oedema only. He was found on investigation to have a large trichobezoar associated with gastric polyposis, protein-losing gastroenteropathy, and steatorrhoea.

## Method

Gastrointestinal protein loss was estimated by the clearance technique of Van Tongeren and Reichert (1966). Forty microcuries of <sup>51</sup>CrCl<sub>3</sub>, diluted in physiological saline acidified to pH2, was injected intravenously. Faeces were collected for seven days, each specimen being kept separately. Blood samples were taken 15 minutes, 30 minutes, and 12 hours after the injection, and thereafter at daily intervals. Plasma radioactivity was measured with a well counter (Packard Auto-Gamma spectrometer—model 300B) and expressed as a fraction of the dose per litre. The radioactivity of each specimen of faeces was measured in a sensitive large-volume gamma counter (J. & P. Engineering, Reading, Limited) and expressed as a fraction of the dose per specimen. The rate of clearance of plasma into the gastrointestinal tract, in litres per day, is obtained by dividing the cumulative faecal radioactivity by the area under the curve of plasma radioactivity against time.

Serum immunoglobulin levels were measured by the method of Mancini, Carbonara, and Heremans (1965). Urinary indican was estimated by the method of Curzon and Walsh (1962).

## Case Report

The patient was a 20-year-old male, mentally subnormal as a result of the congenital rubella syndrome. Swelling of the ankles was first noticed in March 1972 in the institution where he has been resident for several years. By the end of September 1972, he had pitting oedema of both legs up to the

knees. There was a mass in the epigastrium and right hypochondrium extending down to the umbilicus. Minimal ascites and a small pleural effusion on the right were the only other positive findings. The cardiovascular system, in particular, was normal.

Urine testing on several occasions showed no significant albuminuria. Blood urea was 36 mg/100 ml. Liver function tests were normal. Haemoglobin level was 10.9 g/100 ml. Serum iron was 66 µg/100 ml. Serum vitamin B<sub>12</sub> and red cell folate were normal.

The diagnosis was made at gastroscopy. Apart from the trichobezoar, a large number of polyps were seen in the stomach. They were most numerous in the antrum, where the surface of some of them was ulcerated. Two smaller polyps were seen in the duodenum. Multiple biopsies of the gastric polyps showed that they were benign. They were histologically different from adenomata. Although it was not possible to exclude a congenital origin, an inflammatory one was considered more likely (Dr A. McIver).

The results of other investigations are shown in table I, and the barium meal appearance in figure 1.

Before Operation		Eight Weeks after Operation	
Total serum proteins	g/ 3.1		6.3
Serum albumin	100 1.3		3.2
Serum globulin	ml 1.8		2.9
IgG	590		1400 (normal 900-1300)
IgA	220		280 (normal 160-400)
IgM	180		310 (normal 60-180)
Gastrointestinal clearance of <sup>51</sup> Cr-labelled protein	180 ml plasma/day		34 (normal <20)
Five-day faecal fats	14.1 g/day		5.7
Urinary indican	100 mg/day		—

Table I Results of investigations carried out before and after removal of the trichobezoar

The patient started vomiting on 17 October 1972. Two days later, after parenteral fluid and electrolyte replacement, laparotomy was carried out by Mr T. Rowntree. The presence of the polyps was confirmed (fig 2). The bezoar removed was a perfect cast of the stomach, with short tails protruding into the duodenum and the oesophagus (fig 3). The extension into the oesophagus is a very unusual feature. The liver appeared normal.

Recovery was uneventful. The parents consented to the readmission of the patient eight weeks after the operation. Oedema had by then disappeared completely. The results of investigations are compared with the preoperative ones in table I. A repeat gastroscopy showed the polyps to be still present (fig 4). They were somewhat smaller and no longer ulcerated.

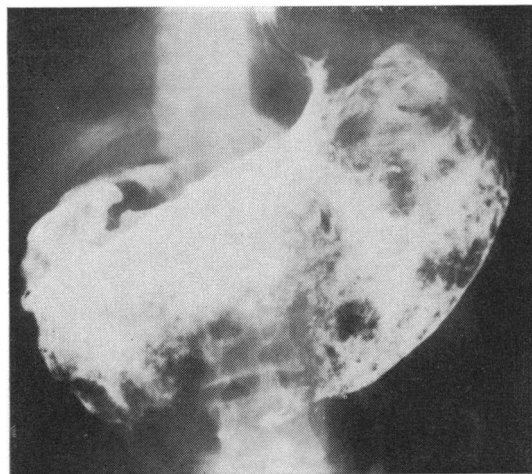


Fig 1 Barium meal showing the trichobezoar in the stomach.



Fig 2 Trichobezoar being removed from the stomach. One large and several smaller polyps can be seen on the exposed mucosa.

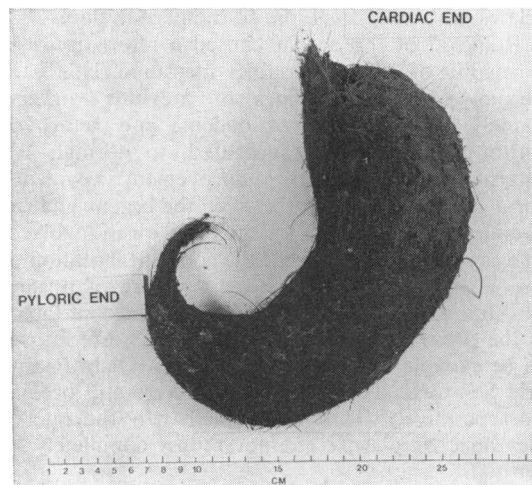


Fig 3 Trichobezoar removed from the stomach of the patient.

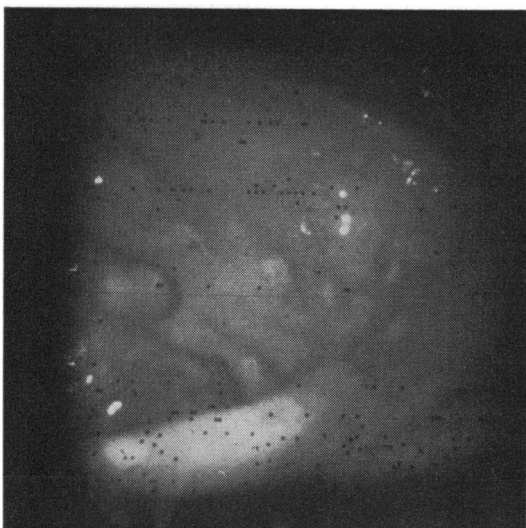


Fig 4 Polyps as seen at gastroscopy eight weeks postoperatively.

### Discussion

Oedema and hypoproteinaemia in the absence of a grossly deficient diet, proteinuria, or overt liver disease should automatically raise the suspicion of protein-losing gastroenteropathy. In this condition the IgG level is usually low, presumably because this protein, like albumin, has a low fractional turnover rate (Waldmann, 1970). A low IgG level was found in our patient, making a diagnosis of protein-losing gastroenteropathy all the more likely. The high rate of clearance of plasma into the gastrointestinal tract confirmed the diagnosis.

The superiority of a technique measuring the clearance of plasma proteins has already been pointed out by Waldmann (1970). Unlike other methods, it relates gastrointestinal loss to the plasma pool.

Removal of the bezoar caused a return towards normality of all the variables measured. This is in keeping with the findings of previous workers, namely, disappearance of oedema and return of serum proteins, where measured, to normal. We interpret the postoperative improvement as evidence for a causal relationship between the bezoar and the protein-losing state. As for the mechanism involved, one can only speculate that the constant irritation of the gastric mucosa leads to the oozing of plasma into the gastric lumen. A possible additional factor is the presence of gastric polyps, which are known to be capable of causing protein loss (Dich, Paaby, and Schwartz, 1961). The polyps were still present postoperatively. This may explain why the rate of clearance of plasma did not return completely to normal.

The patient of Davies (1921), who presented with oedema and was found to have a gastric trichobezoar, also suffered from persistent diarrhoea. Faecal fats were not measured. Steatorrhoea does not appear to have ever been considered as a complication of gastric bezoars. It was present in our patient before, but not after, the removal of the bezoar. A duodenal biopsy taken at gastroduodenoscopy showed the presence of normal villi, making coincidental coeliac disease very unlikely. Culture of duodenal contents aspirated eight weeks postoperatively showed no evidence of bacterial overgrowth. Unfortunately, this was not performed preoperatively, and the bezoar was not cultured. It is of interest that steatorrhoea has been observed in other causes of protein loss in the stomach. In his series of 10 such patients, Jarnum (1963) found three with raised faecal fats. Two of those three were achlorhydric, and bacterial overgrowth in the small intestine as a cause of the steatorrhoea is a distinct possibility (Sherwood, Goldstein, Haurani, and Wirts, 1964). The trichobezoar of our patient had the usual highly offensive smell. It is possible that bacterial contamination of the bezoar resulted in bacterial overgrowth in the small intestine and caused his steatorrhoea. The raised urinary indican supports this hypothesis.

We are grateful to Dr P. Todd, to Mr T. Rowntree, and to the Nuclear Medicine Department, and in particular to Mr R. Mardell, and to the nursing staff of the Metabolic Ward for their cooperation.

### References

- Charache, H., Polayes, S. H., Behr, I. S., Murata, M. Y., and Dimetriades, A. D. (1957). Report of a case of gastric trichobezoar complicated by multiple polyps with malignant degeneration of one of the polyps. *Ann. Surg.*, **145**, 282-286.
- Curzon, G., and Walsh, J. (1962). A method for the determination of urinary indoxyl sulphate (indican). *Clin. chim. Acta.*, **7**, 657-663.
- Davies, I. J. (1921). Hair-balls or hair-casts of the stomach and gastrointestinal tract. *Lancet*, **2**, 791-795.
- Dich, J., Paaby, H., and Schwartz, M. (1961). Protein-secreting tumour of stomach: severe hypoproteinaemia cured by removal of gastric polyp. *Brit. med. J.*, **2**, 686-688.
- Jarnum, S. (1963). *Protein-Losing Gastroenteropathy*. Blackwell, Oxford.
- Mancini, G., Carbonara, A. O., and Heremans, J. F. (1965). Immunochemical quantitation of antigens by single radial immunodiffusion. *Immunochemistry*, **2**, 235-254.
- Sherwood, W. C., Goldstein, F., Haurani, F. I., and Wirts, C. W. (1964). Studies of the small-intestinal bacterial flora and of intestinal absorption in pernicious anaemia. *Amer. J. dig. Dis.*, **9**, 416-425.
- Valberg, L. S., McCorrison, J. R., and Partington, M. W. (1966). Bezoar: an unusual cause of protein-losing gastroenteropathy. *Canad. med. Ass. J.*, **94**, 388-391.
- Van Tongeren, J. H. M., and Reichert, W. J. (1966). The quantitative estimation of gastrointestinal protein loss, using <sup>51</sup>Cr-labelled plasma proteins. *Clin. chim. Acta*, **14**, 42-48.
- Waldmann, T. A. (1970). Protein-losing enteropathy. In *Modern Trends in Gastroenterology*, edited by W. I. Card and B. Creamer, **4**, pp. 125-142. Butterworths, London.
- Wine, P. M. (1957). Trichobezoar presenting as nutritional oedema. *Brit. med. J.*, **2**, 590.