On the tenth postoperative day the transverse abdominal incision disrupted. At resuture the opportunity was taken to re-examine the strictures, the most distal of which was found to be causing partial intestinal obstruction. This stricture was resected and a primary end-to-end anastomosis performed. Following this procedure he had repeated episodes of partial subacute intestinal obstruction. Barium follow-through examination confirmed the presence of an incomplete low small bowel obstruction. An elective laparotomy performed three weeks after the initial exploration showed that the three remaining strictures were more severe and were causing the obstruction. They were resected and bowel continuity restored by end-to-end anastomoses. The postoperative course was uneventful. Macroscopic examination of the strictures showed well defined circumferential scarring. Microscopically there was ulceration of the mucosa and submucosa extending to the serosa at the antimesenteric border of the bowel, and healing by fibrosis.

Discussion

This case of strictures caused by enteric-coated potassium chloride is unique, firstly in occurring in a child, and, secondly, because there were multiple strictures. In adults there is typically a single stricture which requires resection. At the initial laparotomy we thought that this approach was not justified in the presence of multiple, non-obstructive strictures. Subsequent experience with this patient indicates that in time such strictures become progressively narrower as healing occurs by fibrosis, and they ultimately produce intestinal obstruction.

[1] Schrader, W H, and Hitchcock, C R, *Journal of the American Medical Association*, 1964, **190,** 586.
[2] Boley, S J, *et al*, *Journal of the American Medical Association*, 1965, **192,** 763.
[3] Farquharson-Roberts, M A, Giddings, A E B, and Nunn, A J, *British Medical Journal*, 1975, **3,** 206.
[4] Ball, J R, *Lancet*, 1976, **1,** 495.

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Behaviour of multiple primary neoplasms

It has been established that the existence of one malignant neoplasm implies increased susceptibility to the development of a second.[1] Only three patients have been described with five separate primary malignant growths. We wish to report two patients, one of whom has had six and the other five separate malignant neoplasms.

Case reports

Case 1—In 1962 a 55-year-old woman had a cervical carcinoma treated by radiotherapy. Nine years later she developed two colonic neoplasms, one in the hepatic and the other in the splenic flexure. The excised specimens were locally infiltrating, annular, fungating, columnar cell adenocarcinomas, having invaded the full thickness of the colon and the surrounding fat. Early in 1976 a Bilroth I gastrectomy was performed for a gastric neoplasm, there being no metastases. The specimen showed a large ulcerating, fungating tumour 5 cm in diameter situated 2·5 cm from the pylorus; two small gastric polyps were present on the excised specimen. Histologically the tumour was a mucus-secreting adenocarcinoma with full thickness invasion. One of the gastric polyps completely separate from the former lesion contained a focus of adenocarcinoma.

In 1977 a mass became palpable in the right iliac fossa. This was cystic, 10 cm in diameter, and in close contiguity with several loops of terminal ileum, a length of which was removed with it. Histologically it was a leiomyosarcoma, the patient's sixth primary malignant neoplasm.

Case 2—In 1970 a 48-year-old man presented with a discrete carcinoma of the caecum infiltrating all layers of the bowel wall. Fifteen centimetres from this at the hepatic flexure was a small sessile polyp. Histologically both tumours were well-differentiated adenocarcinomas. Two years later an annular, ulcerating tumour obstructing the small bowel 10 cm (4 in) distal to the duodenojejunal junction was excised; this was a well-differentiated adenocarcinoma. In 1974 a facial basal cell carcinoma was excised. The fifth tumour, a poorly differentiated rectal adenocarcinoma, developed in 1976.

Neither patient had evidence of metastases with any of their tumours. There was no family history of malignant disease and they are both alive and apparently tumour-free.

Discussion

Of 37 580 patients with malignant neoplasms at the Mayo Clinic, 1909 had two, 74 had three consecutive neoplasms, four had four, and one had five.[1] In addition to the two cases described only three other patients with five separate malignancies have been reported. Moertel's[1] patient had lesions in the breast, colon, skin, ureter, and uterus. Ostrowski[2] described a 51-year-old woman with malignancies of the ovary, cervix, uterus, bladder, and caecum, while Jones[3] reported a 47-year-old man with separate lesions in the duodenum, jejunum, renal pelvis, splenic flexure, and pancreas. The 26 tumours in these five cases were often poorly differentiated, were locally invasive, but in none of them were distant blood-borne metastases or lymphatic spread reported.

Given a predisposition to the development of multiple malignancies it would seem unlikely to find 26 consecutive tumours of the nature described in these cases which had not metastasised by the time treatment was carried out.

In our case 1 two of the tumours were gastric; the overall cure rate of gastric cancer is about 8·5%[4,5] and the incidence of lymph node metastasis at the time of surgery is 75%.[5] In addition, two of the tumours were colonic, having invaded the full thickness of the colon and the expected cure rate is about 30% for each of these. It is still early for us to assess the success of treatment of the sixth tumour. It would appear that these multiple malignancies are behaving atypically. In addition to having an inherent predisposition to develop multiple tumours, these patients would also seem to have a resistance to the spread of their tumours, possibly from an altered immunological relationship between the tumour and the patient, though the nature of such an immunological response is unknown.

[1] Moertel, C G, Dockerty, M B, and Bagenstross, A H, *Recent Results in Cancer Research*, vol 7, p 191. New York, Springer, 1966.
[2] Ostrowski, J, *British Medical Journal*, 1975, **4,** 736.
[3] Jones, P, *British Medical Journal* , 1976, **1,** 1533.
[4] Lundh, G, Burn, J I, and Kolig, G, *Annals of the Royal College of Surgeons of England*, 1974, **54,** 219.
[5] Costello, C B, Taylor, T V, and Torrance, B, *British Journal of Surgery*, 1977, **64,** 47.

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Basilar artery migraine with transient atrial fibrillation

Basilar artery migraine with impairment of consciousness is well recognised.[1] We wish to report a case of recurrent transient atrial fibrillation during attacks of basilar artery migraine with loss of consciousness and speculate on the mechanism.

Case report

A 46-year-old man with a family history of travel sickness and paternal migraine had had recurrent headaches with nausea and vomiting since childhood. At the age of 32 the pattern of his attacks changed. A typical attack began with a hot clammy feeling, dizziness, occasional rotatory vertigo, unsteadiness of gait, followed by prostrating bilateral occipital headache, nausea, and vomiting. His only visual disturbance has been photophobia and during attacks he has had parasthesiae of the hands and arms but no dysarthria, dysphasia, or hemiparesis.

On three separate occasions he has been admitted to hospital as an emergency semicomatose in atrial fibrillation confirmed electrocardiographically. Semicoma lasted 1-4 hours, drowsiness up to 20 hours, followed by headache and spontaneous reversion to sinus rhythm. On other occasions he has had similar attacks at home but neither he nor his wife have noticed his pulse irregular at any time.

Apart from mild high tone deafness, the results of investigations including skull *x*-ray films, lumbar puncture, left carotid angiogram, chest *x*-ray film, and protein bound iodine concentration were all normal as was his ECG between attacks. An EEG was reported as showing right-sided slow and sharp waves over the parietal occipital areas.

Treatment at intervals with digoxin, phenytoin, clonidine, amitriptyline, and psychotherapy did not alter the frequency or the nature of the attacks.

Comment

Our patient satisfies the symptoms of basilar artery migraine, as outlined by Bickerstaff.[1] During the migrainous attack he has had three documented episodes of impaired consciousness with atrial fibrillation and no evidence of other precipitating cause for the cardiac arrhythmia. Bickerstaff has speculated that the loss of consciousness in basilar artery migraine may be due to either "epilepsy" or "brain stem ischaemia."[2 3] Mauk *et al* have shown that stimulation of the reticular activating system in dogs leads to cardiac dysrhythmias including atrial fibrillation.[4] These dysrhythmias were mediated by the sympathetic nervous system.[5]

We suggest that ischaemia of the reticular activating system due to basilar artery migraine caused both impairment of consciousness and atrial fibrillation. Therefore neither epilepsy nor atrial fibrillation is likely to be the cause of impairment of consciousness in basilar artery migraine and the logical prophylaxis of the atrial fibrillation would be β-sympathetic-blocking drugs.

[1] Bickerstaff, E R, *Lancet*, 1961, **2,** 1057.
[2] Bickerstaff, E R, *Lancet*, 1961, **1,** 15.
[3] Bickerstaff, E R, *Proceedings of the Royal Society of Medicine*, 1961, **55,** 167.
[4] Mauk, H P, Hockman, C H, and Hoff, E C, *American Heart Journal*, 1964, **68,** 98.
[5] Hockman, C H, Mauk, H P, and Hoff, E C, *American Heart Journal*, 1966, **71,** 695.

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Familial juvenile-onset diabetes

Both hereditary and environmental factors are thought to be important in the aetiology of classical juvenile-onset diabetes. We describe a family in which both parents were diabetics and three of the five children developed overt diabetes within four months.

Patients, methods, and results

The figure shows the HLA genotypes of the family. Mother developed classical insulin-dependent diabetes in 1967 when aged 35. Father was discovered to have glycosuria at a routine medical examination in 1970 when aged 30. His diabetes was well controlled by glibenclamide 5 mg daily. Both parents had diabetic mothers.

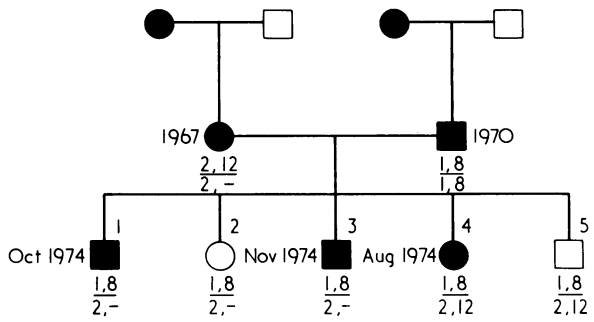
Sib 1—The eldest son, aged 12, was found to have glycosuria in September 1974. Insulin treatment was started three weeks later.

Sib 3—This 10-year-old boy was found to have a trace of glycosuria in August 1974 at the time of his sister's diagnosis. In October he had an illness with severe chest pain, which was identified as Bornholm disease. Diabetes was diagnosed a month later after three weeks of classical symptoms and he was given immediate insulin treatment.

Sib 4—The 9-year-old sister was diagnosed in August 1974 as a case of insulin-dependent diabetes after six months of nocturnal enuresis. Mother had found heavy glycosuria in May but had ignored it as the child appeared to be well.

Sibs 2 and 5 had no diabetic symptoms.

Samples of serum obtained in December 1974 from the three diabetic sibs



HLA genotypes of family and dates of confirmation of diagnosis. Black symbols represent patients with diabetes.

were tested for islet-cell antibodies (ICAs) and antibodies to Coxsackie virus types B1-5, mumps, cytomegalovirus, and *Mycoplasma pneumoniae*. No ICAs were found. Sib 3 had high antibody titres to Coxsackie virus types B1 (1/1024) and B5 (1/256), which were consistent with the recent episode of Bornholm disease. Sib 4 had a low antibody titre to Coxsackie B2 (1/32).

The family was re-examined in April 1975, and HLA typing was carried out. Serum samples from all members of the family again contained no ICAs. Standard oral glucose tolerance tests were performed on the two apparently normal sibs with the following results. Blood glucose concentrations at 0, 30, 60, and 120 minutes were 3·7, 5·2, 7·1, and 5·5 mmol/l (66·7, 93·7, 127·9, and 99·1 mg/100 ml) in sib 2; and 3·0, 4·7, 7·6, and 6·4 mmol/l (54·1, 84·7, 137·0, and 115·3 mg/100 ml) in sib 5. Serum insulin concentrations in sib 2 were 4, 21, 45, and 31 μU/ml at 0, 30, 60, and 120 minutes; and in sib 5, 4, 4, and 10 μU/ml at 0, 30, and 120 minutes. Both sets of results were slightly abnormal. The two-hour blood glucose concentration in sib 5 was more than twice the fasting value, and this was associated with a poor insulin response. The insulin response in sib 2 was delayed and prolonged, and again the two-hour blood glucose concentration greatly exceeded the fasting value.

Comment

Inheritance of diabetes over three generations is rare in families of juvenile-onset diabetics.[1] The abnormalities of glucose tolerance and insulin secretion found in the two apparently normal sibs suggested that all the children were in some way affected by the disease. The diagnosis of juvenile diabetes in three members of the family within a short time suggested the presence of a common initiating factor, possibly—in view of the recent Bornholm-like illness in sib 3—of viral origin. Tests for a limited number of viral antibodies did not, however, suggest recent infection by a virus common to all the children. The absence of circulating ICAs, which are present at onset in many juvenile-onset diabetics,[2] indicated that the diabetes in this family may have been of an unusual kind.

These results support the hypothesis that a complex interplay of genetic and environmental factors may be responsible for juvenile diabetes.

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[1] Tattersall, R B, and Fajans, S S, *Diabetes*, 1975, **24,** 44.
[2] Lendrum, R, *et al*, *Lancet*, 1976, **2,** 1273.

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