Recurrent hemiplegia due to hypoglycaemia

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Summary

A case of recurrent hemiplegia resulting from hypoglycaemia associated with a large thoracic fibrosarcoma, and the satisfactory response to removal of the tumour, is described. The possible causes of the hypoglycaemia are discussed.

Introduction

The association of hypoglycaemia with mesenchymal tumours is well recognized, though ill-understood, and treatment is usually difficult.

Case report

A 74-year-old woman was referred as an emergency with a right hemiplegia of one day's duration. She was found to be semi-comatose, dysphasic and with a mild right hemiplegia. By the evening of the day of admission she had fully recovered and was able to tell us that she had felt dizzy and had staggered from time to time, usually in the morning, for the previous few weeks.

Thereafter, it was noted in the ward that her symptoms of dizziness, slurred speech and right-sided weakness recurred, usually through the night or in the early morning, on several occasions. Blood glucose estimations on these occasions were found to be very low, with figures ranging between 1.2 mmol/litre (22 mg/dl) and 0.9 mmol/litre (17 mg/dl). Having found this, it was possible to prevent the attacks by leaving the patient glucose drinks and carbohydrate snacks at her bedside.

Her previous history showed her to have had a radiological opacity in her left chest occupying the upper half of her left hemithorax, found on X-ray one year previously when she was referred with a cough. Bronchoscopy was negative and surgery was felt unnecessary because of her age, the size of the mass and her general well-being. The provisional diagnosis was of some form of benign tumour.

During the current admission, blood sugar and plasma insulin levels were studied for 180 min after 1 mg intramuscular glucagon but there was no re-

sponse to suggest insulinoma. The blood glucose rose to 5.5 mmol/litre at 90 min but plasma insulin levels were under 3.5 mu./litre. During a glucose tolerance test, plasma insulin levels were also consistently very low (under 6.0 mu./litre). Specimens were then taken with the patient fasting and at two hourly intervals throughout the day. Despite very low fasting glucose levels, the plasma insulin concentration was never raised.

Her chest X-ray showed that the mass had enlarged and she was again referred for an opinion regarding surgery, with the suggestion that the tumour was causing the hypoglycaemia (Fig. 1).

A very large tumour was removed from the left paravertebral region and from the time of the operation no further hypoglycaemia or hemiplegic attacks occurred. The tumour was found to be a fibro-sarcoma. Extraction of tumour tissue showed no significant insulin-like immunoreactivity. Unfortunately, it was not possible to obtain arterio-venous glucose levels across the tumour at the time of operation. Staining of the tumour for glycogen did not reveal this to be present.

Discussion

Recurrent hemiplegia in such a case is presumably due to the effect of hypoglycaemia on a brain possibly rendered susceptible by an already deficient vascular supply (Lawrence, 1967; Silas, Grant and Maddocks, 1981). Characteristically this occurs after fasting and is relieved by food (Payne and Davison, 1979). The tumours which cause hypoglycaemia are most commonly of mesenchymal origin and large (Marks, 1976). The pathogenesis of hypoglycaemia is not clear but the subject has been well reviewed recently (Kahn, 1980).

In this case, one can be certain that the hypoglycaemia was due to the tumour as it has not recurred since the operation 5 years ago. The possible mechanisms in this case are several. Firstly, insulin production by the tumour, or production by the tumour of a substance which stimulated the secretion of insulin

FIG. 1. Pre-operative chest X-ray.

by the pancreas should be considered. Both these possibilities would appear to be excluded by the absence of raised plasma levels of immuno-reactive insulin before the operation, when the patient was hypoglycaemic or had been stimulated with glucose or glucogen. A further possibility is that the tumour was producing a substance which had the properties of insulin, but did not react to the antibody used in the assay. This possibility cannot be excluded as no biological assay for insulin in the tumour could be arranged. Electron microscopy of the tumour failed to show any structures associated with hormone secretion, making it unlikely that either insulin or an insulin-like substance was being formed.

The second possibility that the tumour itself used glucose should be considered. Unfortunately, it proved impossible to collect blood from vessels entering and leaving the tumour in order to measure the arterio-venous difference in glucose concentration, because of dense adhesions between the tumour and the pleura. The absence of glycogen in the tumour tissue excludes the possibility that glucose was being stored in this way, but it is possible that the large mass of the tumour used glucose in its

metabolism. Other cases have been reported in which this was considered to be the most likely explanation for the hypoglycaemia (Payne and Davison, 1979), and this is also the probable explanation in this patient.

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