CASE REPORTS

Recurrent spontaneous hypothermia

I. N. SLOTKI*

B.Sc.(Hons), M.B., M.R.C.P.

M. H. OELBAUM

M.D., F.R.C.P.

Department of Medicine, North Manchester General Hospital, Manchester M8 6RB

Summary

A patient with recurrent attacks of hypothermia is presented. Despite intensive investigation, no other manifestation of hypothalamic or autonomic dysfunction has been found. Treatment of concurrent biochemical hypothyroidism has not altered the frequency or severity of the hypothermic attacks.

Introduction

Spontaneous hypothermia is rare. It occurs in metabolic diseases such as myxoedema and as a result of certain focal lesions of the hypothalamus and adjacent areas of the brain. There are also about a dozen case reports of recurrent spontaneous hypothermia of undetermined cause (for review see Maclean and Emslie-Smith, 1977). A further case is now reported which is of interest in that treatment of concurrent sub-clinical hypothyroidism has not prevented the hypothermic attacks.

Case report

A 60-year-old spinster was admitted to hospital in January 1974 with a 5-week history of increasing confusion and lethargy and 2 to 3 days' wandering naked around her centrally heated house.

Examination

The patient was lethargic and uncommunicative; skin was dry. Rectal temperature 27°C. She had neck stiffness, rigidity, hyperreflexia and extensor plantars. Pulse was 44/min, sinus rhythm, BP 90/60 mmHg. Despite attempts to rewarm her she remained hypothermic for 2 weeks. Her conscious level and abnormal neurological signs fluctuated until 5 days before the end of the attack, when steady recovery occurred.

From 1974 to July 1979 she suffered 11 similar attacks necessitating admission. All except 2 (May 1978 and May 1979) were between November and

February. There were no precipitating factors and recovery time varied from 3 to 20 days. During 3 admissions she developed urinary infections, associated with pyrexia (38°C). From 1975 she had post-menopausal osteoporosis.

Investigations (during and between attacks)

Blood counts showed intermittent microcytic hypochromic anaemia, due to iron deficiency; no bleeding site was identified and oral iron corrected the abnormalities. Serum urea, electrolytes, creatinine, liver function tests, protein electrophoresis, amylase, urate, Wasserman reaction and cerebrospinal fluid were normal. Electrocardiograms showed sinus bradycardia or slow atrial fibrillation during attacks, but were otherwise normal. Electroencephalograms showed bilateral theta and delta waves during and between attacks.

Skull X-rays, isotope brain scans, carotid angiography and computer tomography were normal.

Endocrine studies

Serum T₄ and T₃ normal during and between attacks. Serum TSH varied from 6 u./ml to 84 u./ml (normal < 6 u./ml). Thyroxine therapy sufficient to maintain normal TSH did not alter the frequency nor severity of the attacks.

Fasting blood sugars, prolonged glucose tolerance tests, urinary 5 hydroxy indole acetic acid, urinary 17 oxo-steroids and 17 oxogenic steroids normal. Plasma cortisols showed loss of diurnal rhythm in attacks, but were otherwise normal. Synacthen test and growth hormone and cortisol responses to insulin were normal. Urinary gonadotrophin 41 i.u./24 hr (normal 50-400 i.u./24 hr for post-menopausal females). Plasma prolactin was increased on one occasion to 1195 mu./l (normal < 360 mu./l) but was otherwise normal. Responses to water load and deprivation normal.

Autonomic studies (performed between attacks)

Valsalva's manoeuvre, noise, mental arithmetic, i.v. noradrenaline 5 µg/min and atropine 1.2 mg

*Present address: K.R.U.F. Institute of Renal Disease, Welsh National School of Medicine, Royal Infirmary, Cardiff.

caused normal pulse and BP responses (recorded by intra-arterial measurement).

Thermoregulatory studies (performed between attacks)

Raising ambient temperature to 40°C by radiant heat produced sweating, a rise of rectal temperature to 38°C (normals show a slight fall) and skin temperature to 39°C. Lowering ambient temperature by immersion in water at 15°C caused a fall in rectal temperature to 36°C (normals show a slight rise) and in skin temperature to 27°C.

Discussion

Although there are about a dozen case reports of recurrent spontaneous hypothermia the clinical presentation has varied and no aetiology has so far been identified. Prodromal sweating was a feature of most cases, a few have been associated with agenesis of the corpus callosum (Shapiro, Williams and Plum, 1969) and occasionally patients have had grand mal epilepsy (Thomas and Green, 1973). It has been suggested that the condition is a form of diencephalic epilepsy (Shapiro *et al.*, 1969) but although anticonvulsants abolished the attacks in 2 cases (Hines and Bannick, 1934; Hoffman and Pobirs, 1942) and improved them in one (Maehle, 1973) these drugs are generally ineffective.

The attacks in the present patient have been prolonged and mainly in winter, and prodromal sweating has not been observed. Investigation showed no structural abnormality of the brain and hypothalamic and autonomic function were normal. Temperature response to urinary infection was appropriate. However, thermoregulatory studies,

indicated minor defects of heat conservation and dissipation. In this respect she resembles case 2 of Duff *et al.*, 1961. Although the present patient was biochemically hypothyroid, thyroxine therapy sufficient to maintain a normal serum TSH did not prevent or modify the hypothermic attacks. Therefore it seems unlikely that the hypothyroidism and hypothermia are related. Duff *et al.*, (1961) suggested an extremely localized focal lesion as a possible cause of hypothermia in their 2 patients. The present authors suggest that their patient may have a structural or functional defect of the thermoregulatory centre, probably degenerative, but at present no firm conclusion can be drawn.

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