

**Brachial diplegia as a sequel to cardio-respiratory arrest: 'man-in-the-barrel syndrome'**

Sir,

We report the case of an elderly woman who developed man-in-the-barrel syndrome following a myocardial infarction complicated by cardio-respiratory arrest. A previously fit 72 year old woman presented as an emergency with a 6-hour history of severe breathlessness associated with central chest pain. On examination she was found to be in severe left ventricular failure. Shortly after arrival in hospital she suffered a cardio-respiratory arrest from which she was resuscitated after 15 minutes. She required ventilatory support and remained unconscious and mildly hypotensive for a period of 2 hours. By 12 hours her conscious level was normal though she was noted to be moderately confused. Twenty-four hours after the cardiac arrest she complained of weakness in the upper limbs. Neurological examination revealed slow cerebation and a mild confusional state. Language and visuo-spatial functions were normal. Cranial nerve examination was unremarkable and in particular eye movements, visual fields and facial power were all normal. There was a global, flaccid weakness of both upper limbs affecting the right side to a greater extent than the left. The weakness was of a cortical pattern, being more marked distally than proximally, and greater in the extensors than the flexors. There was almost complete paralysis of movements at the wrists and fingers. The upper limb reflexes were retained. The lower limbs and sphincter function were normal and all modalities of sensation were intact. Both plantar responses were flexor. The patient survived for 4 weeks in hospital during which time her neurological deficit showed only minimal improvement. She died following a further cardiac arrest and unfortunately permission for autopsy was refused.

Man-in-the-barrel syndrome produces the clinical picture of bilateral upper limb weakness with essentially normal power in the lower limb and facial muscles. The term was coined by J.P. Mohr because such patients appear as if their arms and trunk are constrained within a barrel while the lower limb and cranial musculature can be moved spontaneously or in response to pain.<sup>1</sup> This syndrome is almost invariably due to bilateral infarction to the upper limb areas of motor cortex which lie in the border zone between the anterior and middle cerebral artery territories.<sup>2</sup> The selective damage is thought to occur because, in the presence of systemic hypotension and global cerebral hypoperfusion, perfusion pressure is lowest in the watershed areas.

Man-in-the-barrel syndrome has been rarely reported and is an entity that tends to be poorly recognized by clinicians. It may, however, be more common than is generally appreciated. One study of 34 patients who became comatose following an episode of systemic hypotension showed that 32% developed features of man-in-the-barrel syndrome.<sup>3</sup> There was a poor prognosis for survival in this group.

We report our case to emphasize this poorly recognized sequel to cardiac arrest. Closer attention to man-in-the-barrel syndrome in the future may help elucidate the reasons for its poor prognosis.

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**Snoring as the presenting feature of hypothyroidism**

Sir,

The medical literature would suggest that snoring as the presenting feature of hypothyroidism is exceptional. However, over one third of the adult population at large snore habitually<sup>1</sup> and many snorers do not seek medical attention.<sup>2</sup>

A 58 year old male civil servant was seen in February 1989 for evaluation of slurred speech. This was detected 6 weeks earlier by a laryngologist to whom the patient had been referred with a 3-month history of nasal discharge and snoring.

Further symptoms were now disclosed. Unsteadiness when walking had developed 7 months ago, while within the last 6 weeks hoarseness of the voice had also become apparent and a numb, tingling sensation in the fingertips occurred transiently on awakening. Direct questioning elicited recent listlessness, dry skin and cold intolerance. The snoring became so conspicuous that he and his wife had for several weeks slept in separate rooms. Theretofore, she had never remarked upon spells of sleep apnoea, recurrent arousals or excessive restlessness. The patient was not himself conscious of any daytime hypersomnolence.

Dysphonia and dysarthria were pronounced. There was striking swelling of soft tissues around the face and jowls and the tongue was notably enlarged. Glossal movements were slow and effortful. The palate elevated well on phonation but poorly on reflex activation. Superficial sensation over the fingers was preserved and Tinel's sign was negative bilaterally. The heel-knee test was deranged slightly on either side. Deep tendon reflexes exhibited prolonged relaxation. The gait was wide based and turning about unsteady.

The free thyroxine was <1.0 pmol/l (reference range 9.0 to 28.0), thyroid stimulating hormone 97.0 mU/l (0.2 to 3.6) and plasma cholesterol 9.2 mmol/l (3.6 to 6.5). Antibody to thyroglobulin was present 1:110 000 and to thyroid microsomes 1:27 000. Neck radiographs showed an excess of soft tissue, posteriorly, in the floor of the mouth with compromise of the post-nasal space.

Spontaneous sleep for 30 minutes during standard electroencephalography was attended by frequent snores; no apnoeic spells occurred.

After 10 weeks of thyroid supplement, the facial appearance resembled that in a photograph taken 18 months earlier and the tongue had shrunk to average dimensions. Snoring continued for a further 1 month, ceasing abruptly 10 days after thyroxine had been increased from 50 to 75 µg daily. Neurological symptoms and signs disappeared *pari passu*,<sup>3</sup> none being present 4 months post-treatment. Radiography showed that the post-nasal space now had a typical configuration.

Snoring signifies incomplete obstruction of the upper airway.<sup>1</sup> Factors predisposing to a critical closing pressure during sleep include altered ventilatory control, a small oropharyngeal lumen and an increase in pharyngeal compliance.<sup>4</sup> Each of these circumstances attends the hypothyroid state.

While the tongue and pharynx are both sites where mucinous oedema is apt to occur,<sup>3,5</sup> genioglossus enlargement has particular significance as this muscle is the key element opposing physiological pharyngeal closure. Its contractile properties, moreover, are deranged as well.<sup>5</sup> Decreased sensitivity of respiratory neurones both to hypoxia and to hypercapnoea has been amply demonstrated in hypothyroidism.<sup>7-9</sup> Hypotheses include a reset chemostat secondary to chronic alveolar hypoventilation<sup>8</sup> and central effects of reduced body temperature and metabolic rate.<sup>6,9</sup>

Apnoea is the counterpart to snoring when upper airway obstruction becomes complete. Episodes of obstructive sleep apnoea (OSA) are common in untreated hypothyroidism and, furthermore, moderate with thyroid supplement.<sup>10</sup> Analogous mechanisms may be invoked when seeking to explain cessation of snoring in the present case.

A reduction in body weight can be readily dismissed, since improvement of OSA has been observed in its absence.<sup>6,10</sup> Besides, our patient was not obese. Concomitant resolution of myxoedema<sup>3</sup> has obvious attractions. However, in at least 2 patients with OSA whose apnoeic episodes significantly decreased when euthyroid, the upper airway and neck pre-treatment were reputedly normal.<sup>6,11</sup> Furthermore, in the present case snoring persisted beyond the time physical stigmata of myxoedema had disappeared. Normalizing of respiratory control<sup>6,10</sup> may therefore be specially relevant. The manner in which thyroid hormone influences neural regulatory drive is unclear. Interestingly, elimination of OSA in a patient with hypothyroidism was effected not only by thyroxine but also by medroxyprogesterone which augments inspiratory effort independently of changes in chemical stimuli, metabolic rate or body temperature.<sup>6</sup>

Mechanisms aside, snoring is a predictable complication of hypothyroidism and thyroid function should be routinely evaluated in all habitual snorers.

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## The need for new therapeutic approaches in visceral leishmaniasis during HIV infection

Sir,

Recently, several reports have emphasized the difficulty in diagnosing visceral leishmaniasis during human immunodeficiency virus (HIV) infection because of atypical clinical pictures.<sup>1-3</sup> In this letter, we would like to underline the therapeutic difficulties which are also frequently encountered in this condition.

A 27 year old drug addict positive for HIV-1 antibodies since 1986 and at the generalized lymphadenopathy stage presented weight loss of 5 kg and voluminous hepatosplenomegaly in February 1989. There was no fever, the differential blood cell count was normal and the lymphocyte T4 count was 230/mm<sup>3</sup>. Leishmaniasis was diagnosed after hepatic needle biopsy and this was confirmed by an examination of the myelogram while an immunofluorescence serotest was negative. *Leishmania donovani infantum* was isolated and found to be sensitive to the usual drugs *in vitro*. The patient was given two successive courses of treatment with *N*-methylglucamine (60 mg/kg/day for 15 days). This was completely ineffective. *N*-methylglucamine (20 mg/kg/day) was then administered for 20 days in combination with intramuscular injections of gamma-interferon (Laboratoires Roussel-UCLAF, France) 2 × 10<sup>6</sup> U/m<sup>2</sup>/day. The hepatosplenomegaly regressed by half and leishmania were no longer isolated from the myelogram.

The patient was then lost to follow-up due to renewed drug addiction and further imprisonment. In October 1989 he was again admitted to hospital with an irregular fever with peaks at 39°C caused by relapsing leishmaniasis. Serological tests for leishmaniasis were negative