Neuralgic amyotrophy with bilateral diaphragmatic palsy

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Neuralgic amyotrophy is a well recognised condition of unknown aetiology with characteristic clinical features. These include severe pain in the shoulder region, followed within hours or weeks by motor or sensory abnormalities, or both, in the distribution of the cervical roots or nerves. Unilateral disease is more common clinically but if electromyographic (EMG) studies are performed bilateral disease is often found.

Case report

In late 1981 a previously well 35 year old man presented with two weeks' orthopnoea and increasing exertional breathlessness such that he became breathless after walking 50 m on the level. Four days before the onset of breathlessness severe persisting pain in the right shoulder region occurred. This had developed four days after unaccustomed physical activity. There was no relevant past history, preceding viral illness, recent vaccination, or systemic upset. When he was examined in the supine position the respiratory rate was 40 per minute but it decreased to 24 per minute after he stood up. Breath sounds at the left lung base were decreased. Sensation was diminished on the lateral border of the right upper limb, from shoulder to thumb (corresponding to C5–6 dermatomes). Both triceps, left supinator, and right biceps jerks were diminished. Chest radiography showed collapse and consolidation in the left lower zone. This did not improve with antibiotics and physiotherapy. Bronchoscopy and cervical spine radiographs were normal. Over the next few weeks the dyspnoea remained stable, but the patient became aware of weakness of right shoulder abduction and elbow flexion. During this period paradoxical inspiratory abdominal movement was noted. Diaphragmatic function was evaluated: vital capacity (VC) was 1.0 1 supine and 2.1 1 erect (predicted 4.3 1). The change in transdiaphragmatic pressure measured by oesophageal manometry during a maximum inspiration was zero (predicted >25 mm Hg). Percutaneous stimulation of left and right phrenic nerves produced no electrical response from the diaphragm. Right sural, lateral popliteal nerve motor, median, ulnar, and radial nerve motor and sensory conduction studies all gave normal results. Neither clinical improvement nor change in the VC supine-erect gradient occurred with aminophylline or oral theophylline despite maintenance of serum concentrations of the drug within the therapeutic range for six weeks. The right shoulder pain persisted, but was absent three months later. Five months after the onset of symp-

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as a cause of impaired respiratory function should always be considered.

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References

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