Diaphragm weakness as a cause of breathlessness after anatomically distant surgery

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The case histories are presented of two patients in whom breathlessness developed following surgery to an anatomically distant site. Postoperative recovery was uneventful and the right hemidiaphragm to be normally positioned (image available in online supplement at http://www.thoraxjnl.com/supplemental). No postoperative brachial plexus neuropathy may occur in the absence of direct surgical trauma and it is recognised that the phrenic nerves may also be involved. However, isolated phrenic neuropathy following anatomically distant surgery has not been previously described. Here we present two patients in whom breathlessness developed following surgery to an anatomically distant site. Respiratory muscle testing demonstrated diaphragm weakness in both patients.

METHODS
The following common diagnostic techniques were used.

Spirometry
Forced vital capacity (FVC) and forced expiratory volume in 1 second (FEV1) were recorded using a wedge bellows spirometer (Vitalograph, Buckinghamshire, UK).

Respiratory muscle tests
Global inspiratory and expiratory muscle strength was assessed with techniques described in the ATS/ERS joint statement on respiratory muscle testing using standard laboratory apparatus previously described elsewhere. In brief, maximum static inspiratory (MIP) and expiratory (MEP) mouth pressure were measured using a flanged mouthpiece attached to a short rigid tube with a two-way valve incorporating a 2 mm leak to prevent glottic closure. MIP was measured from residual volume (RV) and MEP from total lung capacity (TLC). Diaphragm strength was assessed as transdiaphragmatic pressure measured during a maximal sniff and by anterolateral magnetic phrenic nerve stimulation. This was performed using a Magstim 200 HP stimulator (Magstim Co, Whitland, UK) with a 45 mm coil placed over each phrenic nerve in turn to assess the strength of each hemidiaphragm and then bilaterally. Subjects rested for 20 minutes before stimulation to minimise twitch potentiation, then supramaximal stimulations were delivered at 100% power output while the patient was relaxed at functional residual capacity with the mouth closed and wearing nose clips. At least three satisfactory twitches were recorded for each patient.

Patient 1
A 46 year old man had an uncomplicated sigmoid colectomy for benign adenoma. Postoperative recovery was uneventful but 2 months later he became breathless and wheezy on exertion. The dyspnoea was particularly worse when lying on his back or immersed in water. He had no medical history of note and was a mild smoker. Chest radiography showed a raised right hemidiaphragm (see online supplement available at http://www.thoraxjnl.com/supplemental). No postoperative chest radiograph was available as a baseline for comparison, but a radiograph taken in the immediate postoperative recovery period shows the right hemidiaphragm to be normally positioned (image available in online supplement at http://www.thoraxjnl.com/supplemental).

Patient 2
A 58 year old woman was admitted to hospital for a clinically suspected pulmonary embolus. Ventilation-perfusion scanning showed a high probability for pulmonary embolism. She had had polio as a child but had not required respiratory support and 1 year before her admission she had had a normal chest radiograph. As part of the work-up for her pulmonary embolism she underwent ultrasound scanning of the abdomen. This revealed a tumour in the superior pole of the right kidney. She had a heminephrectomy 3 months later and histological examination confirmed renal cell carcinoma. Postoperatively she complained of breathlessness which was attributed to her anaesthetic. This breathlessness persisted and, as a result, she underwent CT pulmonary angiography and fibreoptic bronchoscopy. Neither of these investigations found a cause for her dyspnoea. One year later she still complained of shortness of breath on exercise, lying flat or bending forward. Physical examination showed paradoxical abdominal motion. A chest radiograph revealed an elevated left hemidiaphragm which was not present preoperatively (see fig 1 and online supplement).

RESULTS
Data for both these patients are shown in the tables in the online supplement available on the Thorax website at http://www.thoraxjnl.com/supplemental. The baseline characteristics and spirometric parameters are shown in table S1 and the results of respiratory muscle testing in table S2.

In addition to the radiograph in fig 1, the online supplement also contains a series of additional images. The first (fig S1) shows the raised hemidiaphragm in patient 1. In order to compare this and the chest radiograph of patient 2 (fig 1) with a “baseline”, three further radiographs are included as control images, one for patient 1 (fig S2) and two for patient 2 (figs S3 and S4).

The respiratory muscle tests for patient 1 showed global inspiratory and expiratory muscle strength within normal limits but sniff transdiaphragmatic pressure (SnPdi) was low at 43.4 cm H2O (normal >80). Magnetic phrenic nerve stimulation demonstrated right hemidiaphragm weakness with a right twitch transdiaphragmatic pressure (TwPdi) of 1.6 cm H2O (normal >7). The response to magnetic stimulation on the left was normal with a TwPdi of 10.5 cm H2O (normal >8).
Diaphragm weakness after surgery

...can become involved in neuralgic amyotrophy, but this is usually in conjunction with the “shoulder girdle” syndrome of pain described above. Rarely, isolated phrenic nerve involvement can occur, but anatomically distant surgery has not been previously described as a trigger for the condition.

In their original series which described brachial plexus neuralgic amyotrophy, Parsonage and Turner noted that nearly half the patients were already in hospital with other conditions. These included infections (such as malaria) and traumatic conditions (such as gunshot wounds). A minority of these, however, were patients recovering from surgery and, of those patients who did have a surgical trigger (12 of 136 cases), all had had anatomically distant surgery (usually hernia repair). As with our patients, neuralgic amyotrophy developed after surgery and diaphragm involvement was not noted in that series.

Patients with isolated phrenic neuropathy often initially present to chest physicians. Recovery of diaphragm strength is more variable than recovery of upper limb function and typically takes 2–5 years. Although no treatment is known to hasten recovery, identification of the condition is worthwhile if only to spare the patient unnecessary investigations. Plain chest radiographic appearances are a poor guide to diaphragm function and, where the diagnosis is suspected or in patients in whom a cause for breathlessness is difficult to discern, we recommend phrenic nerve stimulation be conducted to quantify diaphragm strength.

**DISCUSSION**

Both patients have shown clear evidence of a phrenic neuropathy and diaphragm weakness which was unilateral in patient 1 and bilateral in patient 2. The sites of the surgery were anatomically distant from the diaphragm so direct trauma to the diaphragm or the phrenic nerves was unlikely, and the weakness persisted long after the perioperative period when electrolyte disturbance or the administration of drugs and the weakness persisted long after the perioperative period were anatomically distant from the diaphragm so direct patient 1 and bilateral in patient 2. The sites of the surgery pathy and diaphragm weakness which was unilateral in...