

Haemothorax via an intact diaphragm

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When haemothorax follows trauma, it usually results from damage to chest wall vessels, pulmonary parenchymal laceration or great vessel injury¹. We describe a case in which blood seems to have passed into the chest through an intact diaphragm.

CASE HISTORY

A woman aged 70 was admitted after collapsing with sudden left-sided weakness. On admission her Glasgow coma score (GCS) was 12 and she had a mild left hemiparesis, though deep tendon reflexes were symmetrical and plantar reflexes downgoing. Examination of the cardiovascular and respiratory systems and abdomen revealed nothing of note. Her haemoglobin was 13.8 g/dL and platelet count, clotting screen and blood chemistry were normal. A computerized tomographic (CT) scan of her head revealed a subarachnoid haemorrhage. Thirty hours after admission her GCS fell to 10 and she became haemodynamically unstable with a heart rate of 120 beats per minute and an unrecordable blood pressure. A right internal jugular central venous line was inserted and she was resuscitated with colloid transfusion. Subsequently she developed recurrent generalized seizures which were terminated with intravenous diazepam. No clinical cause was found for her cardiovascular collapse, and, in view of the onset of seizures, she was thought to have had a cerebral aneurysmal rebleed. A chest radiograph performed four hours post-resuscitation was unremarkable and showed satisfactory placement of the right internal jugular line (Figure 1). She was loaded with intravenous phenytoin, received two units of packed cells and two units of fresh frozen plasma, and was transferred to the regional neurosurgical centre.

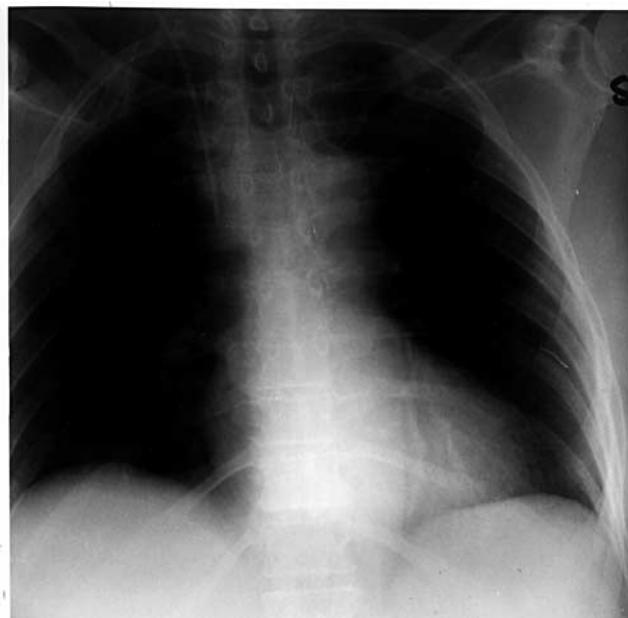


Figure 1 Chest radiograph 4 hours after central line insertion

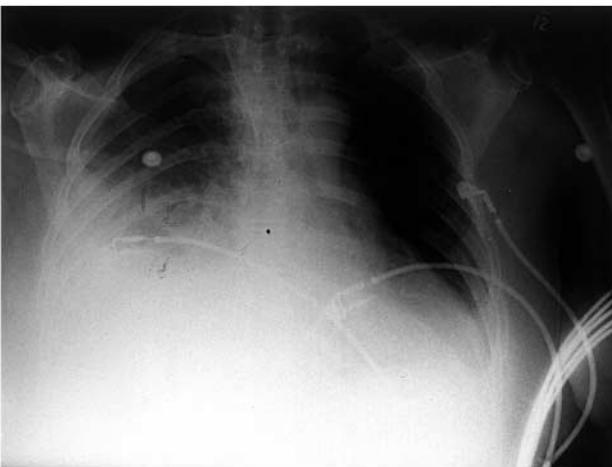


Figure 2 Chest radiograph showing fluid within the right chest cavity

On arrival her neurological state was unchanged and she was haemodynamically stable. There was clinical evidence of a large right-sided pleural effusion and her abdomen was diffusely tender. Haemoglobin was now 8.5 g/dL, and a chest radiograph (Figure 2) confirmed the presence of a fluid collection in the right pleural cavity. Ultrasound guided aspiration yielded heavily bloodstained serous fluid (haemoglobin 6.5 g/dL). A chest drain was inserted and 950 mL was drained. Chest radiographs before and after insertion of the pleural drain showed no rib fractures. An ultrasound examination of the abdomen revealed free fluid in the abdomen and an 8 cm laceration in the right lobe of the liver; on aspiration the fluid proved to be blood (haemoglobin 12.8 g/dL). Since she had remained haemodynamically stable without further transfusion, she was

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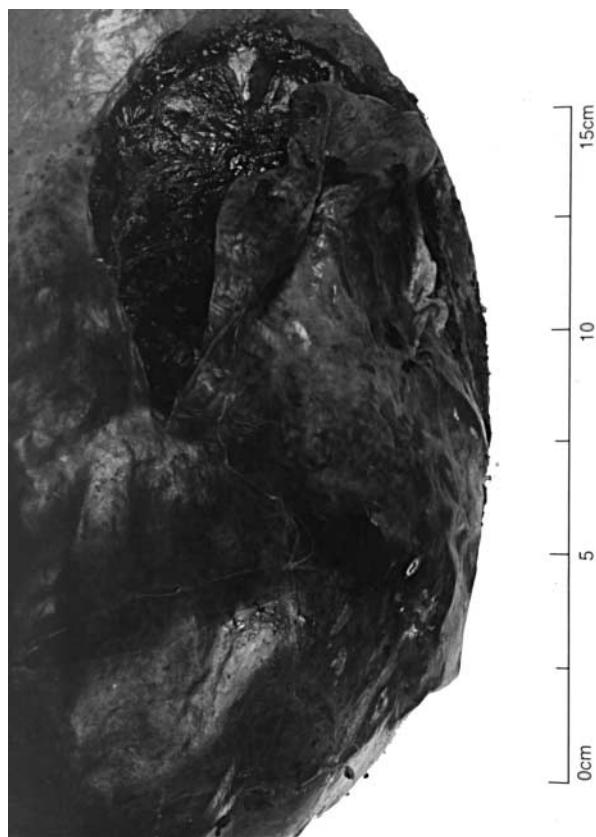


Figure 3 Tear in liver capsule seen at necropsy

managed conservatively. Fourteen days after the initial subarachnoid haemorrhage she had an asystolic cardio-respiratory arrest from which she could not be resuscitated.

Necropsy confirmed the presence of subarachnoid haemorrhage and a saccular aneurysm of the anterior communicating artery. The aorta was fully intact. The central veins were normal; injuries to the vessels of the mediastinum and neck were sought and not found. 500 mL of bloodstained fluid was present in the right chest cavity with some collapse of the right lung. The abdominal cavity contained some 600 mL of fresh blood and there was a 10 cm capsular tear of the liver anteriorly (Figure 3). The diaphragm was intact, with blood adherent to its undersurface.

COMMENT

Presumably this patient sustained the liver laceration when she originally collapsed. How can we account for the haemothorax, when the diaphragm was intact?

No intrathoracic trauma was evident. Another cause to be excluded was the central venous cannulation². In this patient, a right internal jugular central venous catheter had been inserted during resuscitation but the chest radiograph after insertion was normal and no vascular injury was found at necropsy. Furthermore, the chest fluid had a much lower haemoglobin concentration than the abdominal fluid, so the abdominal cavity was the likely source of the blood.

We have found a single report of splenic rupture presenting with a haemothorax³. The patient had suffered blunt trauma to the left side of the chest and abdomen and surgical exploration revealed a large diaphragmatic tear and rupture of the spleen. In the case we report here, in which the diaphragm was intact, we think that blood from the abdomen must have found its way into the pleural cavity by pleuroperitoneal connections. There is a well documented association between ascites and pleural effusions in patients with liver disease and pelvic neoplasms^{4,5}. In such instances an exudative or transudative pleural effusion may be the consequence of passage of peritoneal fluid through an intact diaphragm⁵. Meig's syndrome, initially described by Salmon in 1934⁷ and subsequently by Meig and Cass in 1937⁸, describes the presence of ascites with concomitant pleural effusions in patients with benign ovarian fibromas. The syndrome is now known to be associated with several primary pelvic neoplasms, both benign and malignant. Johnstone and Loo studied the phenomenon in patients with cirrhosis and ascites⁵ and concluded that absorption of ascitic fluid through diaphragmatic lymphatics was the mechanism of formation of hepatic hydrothorax. However, Nakamura *et al.*⁹ showed that technetium-99-labelled albumin injected into the abdominal cavity accumulated in pleural fluid within 7 minutes⁹. Since lymphatic flow rate in the thoracic duct is only 1 mL/h per kg bodyweight, lymphatic transfer does not adequately explain this phenomenon. Ascitic fluid appears to pass directly through defects in the diaphragm into the pleural space. Microscopic (0.03–1.2 mm) and larger (up to 5 mm) defects, some with epithelialized ducts, have since been demonstrated at necropsy in patients with ascites and cirrhosis¹⁰. These are believed to result from thinning and subsequent spontaneous microrupture of the tendinous part of the diaphragm¹¹. Hydrothoraces have also been noted in patients undergoing peritoneal dialysis¹² and are thought to occur by the same mechanism.

The concept that blood may cross from the peritoneum to the pleural cavity in the absence of diaphragmatic injury is of interest since it extends the mechanisms thought to be involved in the production of Meig's syndrome to the development of haemothorax after intra-abdominal bleeding. In addition, these findings have implications in the assessment of patients with haemothorax following trauma. Whilst investigation for intra-abdominal injury would always be mandatory¹³, our experience provides another mechanism by which confounding clinical findings may be observed.

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Acromegaly, primary thyrotoxicosis, a dense left hemiparesis and a right internal carotid aneurysm

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When multiple pathology exists, the individual components can be hard to manage.

CASE HISTORY

A man aged 57 was admitted having been found collapsed on the street. On examination, he was unconscious with a Glasgow coma scale of 5. He had a dense left-sided hemiparesis. Bony features of acromegaly were noted, together with soft-tissue wasting and evidence of a proximal myopathy. He had no goitre or dysthyroid eye signs. Blood pressure was 200/110 mmHg.

His initial endocrine investigations (see Box 1 for key to abbreviations) showed a raised serum GH of 238 mU/L and a low-normal serum IGF-I of 13.7 nmol/L (normal range 11–43.2). He was thyrotoxic (TSH undetectable, free thyroxine 59.9 pmol/L, free tri-iodothyronine 13.4 pmol/L). On further investigations he proved to have secondary hypogonadism (testosterone 0.4 nmol/L, LH < 0.5 U/L, FSH 0.7 U/L). Prolactin (583 mU/L) and cortisol

(691 nmol/L) were normal. Thyroid microsomal and thyroglobulin antibodies were strongly positive.

We subsequently learned that one month earlier his general practitioner had suspected that he was acromegalic and noticed signs of cardiac failure. He had arranged an IGF-I (18.2 nmol/L), TSH (0.3 mU/L) and thyroxine (16.9 pmol/L), all of which were normal, and tri-iodothyronine (3.0 pmol/L), which was low. Since the initial specimen for GH was haemolysed, the blood tests were repeated one week before admission, by which time he had become thyrotoxic (TSH < 0.1, free thyroxine 59.3, free tri-iodothyronine 11.4). His random serum GH was raised (208 mU/L).

Box 1 Key to abbreviations

GH=growth hormone
IGF-1=insulin-like growth factor-1
TSH=thyroid stimulating hormone
LH=luteinizing hormone
FSH=follicle stimulating hormone
ACE=angiotensin converting enzyme
IGFBP=IGF-binding protein

A magnetic resonance scan of his brain revealed a dense right-sided cerebral infarct and a 25 mm pituitary tumour, which was adjacent to a 12 mm calcified thrombosed left internal carotid artery aneurysm (Figures 1 and 2). There was cardiomegaly on the chest X-ray and an echocardiogram showed a dilated left atrium and ventricle with reduced ejection fraction.

He was transferred to the intensive therapy unit for ventilation, where he received carbimazole and testosterone replacement. Despite the poor ejection fraction, he maintained a satisfactory cardiac and urinary output throughout his admission. However, in view of his echocardiogram findings and his hypertension, he was treated with diuretics and ACE inhibition. His conscious level returned to normal over the next week and he began stroke rehabilitation. After five weeks of carbimazole treatment his thyroid hormones fell into the normal range. A repeat 0900 h cortisol was 455 nmol/L indicating

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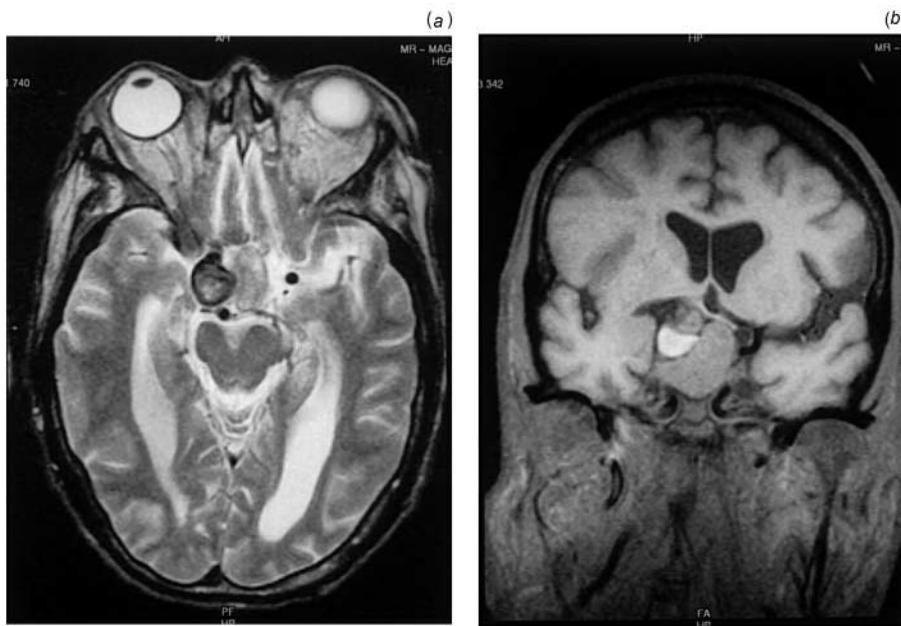


Figure 1 Magnetic resonance transverse (a) and coronal (b) views of the pituitary gland showing a large pituitary tumour adjacent to a right internal carotid artery aneurysm

adequate adrenal reserve. Despite nasogastric feeding and an improvement in his nutritional status, his IGF-I levels remained between 6.9 and 18.4 nmol/L. GH day profile showed levels persistently in excess of 300 mU/L. The diagnosis of GH resistance was substantiated by further investigation of his GH-IGF axis which revealed low levels of free IGF-I at 0.013 nmol/L (normal range 0.11–0.98), IGF binding protein-3 (IGFBP-3) 1.36 mg/L (2.08–4.31) and acid labile subunit 5.7 mg/L (11.8–26.9). In contrast, IGFBP-2 levels were raised at 986 µg/L (224–454) while IGFBP-1 levels were normal at 68.8 µg/L (25.2–129.3).

He had a further stroke eight weeks after admission and died.

COMMENT

The combination of stroke and endocrine disorders made the management of both challenging. The most likely cause of his stroke was an embolism from the internal carotid artery aneurysm. However, we suggest that the stroke was related to his acromegaly and thyrotoxicosis.

The stroke may have been linked to his acromegaly through the rare association between acromegaly and intracerebral aneurysms^{1–3}. The aetiology linking these two conditions is not established but may relate to turnover of collagen. This is increased in acromegaly and results in a deficiency in type III collagen, which has been linked with the development of intracerebral aneurysms¹.

The thyrotoxicosis may have contributed to the cause of his stroke. Systemic emboli occur in 10–40% of patients with thyrotoxicosis and atrial fibrillation; however, even in the absence of atrial fibrillation, thyrotoxicosis is a risk factor for the development of emboli. Therefore anticoagulation is recommended in the absence of atrial fibrillation if there is a

history of previous emboli, valvular heart disease or left atrial enlargement (as in our patient).

Although there is an association between acromegaly and thyrotoxicosis, the two conditions are unlikely to have been linked in our patient. Acromegaly is not associated with an increase in the prevalence of thyroid autoantibodies and so we believe that the Graves' disease was unconnected. His immediate management, in terms of improving cardiorespiratory function, was made more difficult by his thyrotoxicosis, since this has adverse cardiac effects which can lead to high-output cardiac failure. Thyrotoxicosis could also have been responsible for the myopathy, which possibly impeded recovery from the stroke. Finally, the risks of a subsequent stroke were further increased by the thyrotoxicosis. Though his acromegaly was not relevant to immediate management, it may have increased the risks of a further stroke.



Figure 2 Magnetic resonance angiogram showing a large right internal carotid artery aneurysm

The presence of pronounced GH resistance, as defined by low serum IGF-I in association with raised GH levels, was curious and its aetiology remains unclear. The patient was a recluse and showed signs of malnutrition on admission. Chronic undernutrition has profound effects on the GH-IGF axis, which results in GH resistance. However, despite nasogastric feeding and improvements in nutritional status, IGF-I remained below or at the lower end of the reference range. Catabolic illness results in acquired GH resistance and may explain the low IGF-I levels on admission⁴. However, his IGF-I levels were low a month before admission and remained so despite an initial recovery from his stroke. The effect of thyrotoxicosis on the GH-IGF axis is controversial, but seemingly thyrotoxicosis does not result in a fall in serum IGF-I⁵. Ultimately, this patient had one pathology too many.

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Rupture of the ankle extensor retinaculum in a dancer

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Ligament and tendon injuries around the foot and ankle are common in dancers and ballerinas^{1,2}, but rupture of the ankle extensor retinaculum has not previously been reported.

CASE HISTORY

A professional ice dancer aged 17 had a sudden pain in her right ankle while skating and was unable to continue. When seen in the orthopaedic clinic two weeks later the foot was painful and she was worried by an abnormal 'band' that appeared over the anterior aspect of her right ankle when she pulled her foot up. On examination there was a palpable defect in the inferior part of the ankle extensor retinaculum, through which the tendon of tibialis anterior seemed to bowstring. This was accentuated by ankle

dorsiflexion (Figure 1). The ankle was therefore explored surgically.

At operation the superior limb of the inferior part of the extensor retinaculum was found to have ruptured, allowing a normal-appearing tibialis anterior tendon to bowstring through. The rest of the extensor retinaculum was intact. The superior limb of the inferior extensor retinaculum was then reconstructed with a segment of fascia lata harvested



Figure 1 View of both ankles demonstrating the tibialis anterior tendon 'bowstringing' over the anterior medial aspect on the right

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from the thigh. Postoperatively the patient's ankle was initially immobilized in a below-knee equinus plaster-cast for 3 weeks and then in a neutral cast for a further 3 weeks. Thereafter the patient underwent 6 weeks of physiotherapy. At review five months after surgery she was pain free and had returned to her preinjury level of skating. On examination the tibialis anterior tendon was well contained.

COMMENT

Although rupture of the tendons that pass under the ankle extensor retinaculum has previously been reported³, rupture of the extensor retinaculum itself has not. We have been unable to find a specific cause for the retinacular injury described and suspect that an element of attrition was involved. However, no predisposing repetitive manoeuvre or underlying disease was identified.

We are also unsure why a partial retinacular rupture should have been so disabling. The wrist extensor retinaculum is often left partly deficient after reconstruction

procedures for rheumatoid arthritis without any troublesome bowstringing of the wrist extensor tendons⁴. Perhaps the greater change in direction taken by the extensor tendons as they pass over the front of the ankle to the foot or the short distance to the insertion of tibialis anterior is of relevance.

This unusual case offers a reminder that a sports-related ankle injury may occasionally be due to a tendinous or retinacular lesion rather than to the more common ligamentous or bony damage.

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Malign anterior knee pain

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Anterior knee pain is common and usually of benign origin; but malignant disease should not be forgotten.

CASE HISTORY

A woman aged 32 consulted her general practitioner because of bilateral anterior knee pain, worse on the right. Nothing of note was found on examination apart from obesity, and radiographs of the right knee were reported normal. She was advised to lose weight, modify activity and use simple analgesia as required. 2 years later she was seen with the same complaint by an orthopaedic surgeon who referred her to a rheumatologist. Again history, examination and radiographs were unremarkable and the working diagnosis was patellofemoral osteoarthritis with infrapatellar bursitis. Similar advice regarding weight loss, activity and analgesia was given. During the next six months swelling developed on the inferolateral aspect of the right knee with tenderness on the medial joint line and over the lateral and medial poles of the patella. There was a full range of

movement with tenderness at extremes. Her erythrocyte sedimentation rate (ESR) was 71 mm/h but rheumatoid factor was absent, and the diagnosis was revised to synovitis, possibly due to an inflammatory arthritis. Injection of the knee joint with 80 mg methylprednisolone gave no relief but two months later her ESR had declined to 31 mm/h. Over the subsequent five months two further steroid injections were given, again without symptomatic improvement. One month later—3 years after the onset of symptoms—the patient suffered a trivial hyperextension injury to the right knee resulting in pronounced local tenderness and an inability to straight-leg-raise. A radiograph revealed a lytic lesion with associated fracture through the inferior pole of the patella (Figure 1).

At operation 10 mL of lipohaemarthrosis fluid was drained from the joint and a 1 × 1 cm cavity in the inferior pole of the patella was identified. This contained yellow jelly-like material extending into the ligamentum patellae and surrounding synovium. On histological examination this proved to be a grade 2 myxoid chondrosarcoma with incomplete excision margins, and at a further operation the patella and distal quadriceps were excised. Again the excision margins were found to be incomplete and an above-knee amputation was performed. Seeding of the tumour was found histologically throughout the joint. Six years postoperatively there is no evidence of local or metastatic disease.

COMMENT

A Medline search, a hand search and discussion with specialists did not yield any previous reports of primary

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Figure 1 Lateral radiograph of right knee

chondrosarcoma at this site. We believe that this is the first time this tumour has been reported arising from the patella.

Skeletal myxoid chondrosarcomas are variants of conventional chondrosarcomas. They are usually low-grade malignant neoplasms with lobulation and sparse cellularity; composed of round stellate cells with abundant mucoid matrix, they have the histochemical characteristics of cartilage¹, and they usually occur in the axial skeleton. Skeletal chondrosarcomas, whether myxoid or not, are graded 1 to 3, with metastatic potential rising with the grade. Myxoid degeneration is a purely descriptive term indicating less differentiation. However, chondrosarcomas have the ability to 'dedifferentiate' over time. In a series of 20 patients with skeletal chondrosarcoma reported from the Memorial Sloan-Kettering Cancer Center², New York, the most common sites of skeletal myxoid chondrosarcoma were the pelvis (6 cases), the femur (5) and the tibia (2);

the other 7 cases were distributed through the axial skeleton. 5 of these patients had developed metastases.

Whether a primary patella tumour is benign cannot be decided radiologically³ but most of them are: in a published series of 42 patellar tumours⁴, 90% were benign, the commonest diagnosis being chondroblastoma. In addition to chondrosarcomas, malignant primary tumours arising from the patella include haemangioendothelioma, haemangiopericytoma, lymphoma, fibrous histiocytoma, osteoblastoma, osteosarcoma and plasmacytoma. Metastasis to the patella is rare but well described⁵.

In malignant tumours of the patella, pathological fracture is often the presenting complaint. Interruption of articular cartilage signifies contamination of the joint and the slightest bone damage may lead to metastasis⁶, notably to the lungs. Diagnosis before fracture is therefore greatly preferable.

Since clinical and radiological diagnosis is unreliable, the safest option may be fine needle aspiration, possibly under computed tomographic guidance, with care not to cross into the joint. Open biopsy of sarcomatous tumours is ill-advised³. The diagnosis of primary bone malignancy should prompt referral to a specialist bone oncology centre. Surgical treatment of a primary patella tumour usually involves total patellectomy with muscle-tendon transfer if the extensor mechanism has to be resected.

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Group A streptococcal meningitis with an infected BCG scar

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Despite the recent increase in severe invasive Group A streptococcal disease, *Streptococcus pyogenes* remains a rare cause of meningitis.

CASE HISTORY

A 3-month-old boy presented with two days of fever, irritability, diarrhoea, vomiting and a widespread violaceous macular rash. Generalized seizures and cardiovascular collapse ensued and he was admitted to paediatric intensive care for sedation, mechanical ventilation and resuscitation with intravenous colloid, dopamine and noradrenaline. White cell count was $12.3 \times 10^9/L$, predominantly neutrophils, and C-reactive protein was 190 mg/L. Intravenous cefotaxime was started. Computed tomography revealed bilateral subdural collections which required repeated drainage. In cerebrospinal fluid the white cell count was $4 \times 10^9/L$, predominantly lymphocytes, protein 1.5 g/L and glucose 2.4 mmol/L. Plasma glucose was 7.2 mmol/L. No organisms were seen on Gram and auramine stains, nor cultured from cerebrospinal fluid taken after the start of antibiotics. Blood cultures grew *S. pyogenes*. The colonial appearance was mucoid and the organism had non-typable M and T antigens. Sensitivity testing revealed tetracycline resistance only. The same organism was isolated from his subdural collections and the site of his previous BCG immunization, though this did not appear infected (Figure 1). There was no family history of sore throats or handling by relatives with infected skin lesions or eczema. Throat swabs from the child and mother grew commensals. His cardiovascular system stabilized, seizure activity abated and he was weaned off inotropes. Antimicrobial therapy was discontinued after two weeks. At 3 months' follow-up he was seizure-free and neurodevelopmentally normal.

COMMENT

A 'Medline' search of published work since 1966, using the key words *S. pyogenes* and meningitis, yielded one review¹ and



Figure 1 The patient in intensive care after bilateral craniotomies to drain subdural collections. A normal-appearing BCG scar is visible on his left arm

several case reports. In most cases *S. pyogenes* meningitis was community acquired and affected immunocompetent children.

The initial presenting features were lethargy, irritability, nausea, vomiting, headache and sore throat, and the most common signs were fever and meningism. Rash, not typically scarlatiniform, was described twice^{1,2} but without desquamation and toxic shock. Most patients recovered fully; severe neurological impairment, sensorineural hearing loss, cranial nerve palsies, subdural collections and brain abscesses were rare complications. In cases where *S. pyogenes* was cultured from the cerebrospinal fluid blood cultures were normally positive. The white cell count in cerebrospinal fluid was always high, and on three occasions lymphocytes predominated^{1,3,4}. Three cases are reported where the organism was isolated from skin; two of these lesions did not appear infected^{1,5}.

We think that our patient's slow healing BCG scar provided a portal of entry for a highly virulent skin organism to seed to the meninges via the blood. We are not suggesting vaccine contamination. Trivial skin lesions are known to predispose to severe invasive disease caused by *S. pyogenes*. This, to our knowledge, is the first report of Group A streptococcal meningitis complicating recent immunization.

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