Persistent haemorrhagic ascites in generalised haemolymphangiomatosis: a therapeutic dilemma

We report a case of intractable, recurrent intraperitoneal bleeding secondary to visceral lymphangiomatosis, which we do not think has been reported before.

Case report

A woman was well until 1976, when, aged 24, she developed recurrent haemoptyses. Chest radiography showed multiple right cavities in the upper zone. Sputum screening for acid-fast bacilli yielded negative results, and bronchoscopy was unhelpful. Right upper lobectomy was performed, and histological examination showed fibrous cavities lined with alveolar macrophages laden with pigment.

She presented in 1981 with massive splenomegaly. Haemoglobin concentration was 121 g/l, and a peripheral blood film showed macrocytosis and Howell-Jolly bodies. The histological appearance of a bone marrow trephine biopsy specimen was typical of myelofibrosis. At laparotomy a multicystic spleen weighing 4.2 kg was removed. The splenic pulp was replaced by large thin walled vascular spaces filled with proteinaceous material (figure). Postoperative chest radiography showed upper mediastinal widening, and a skeletal survey showed sclerotic deposits in the pelvis, scapula, and femoral heads.

In 1983 she developed severe anaemia (haemoglobin concentration 55 g/l) and gross ascites. Aspiration showed heavily bloodstained fluid with an initial haemoglobin content of 77 g/l. Over the next three years she required abdominal paracentesis on several occasions and received 48 units of blood. In January 1985 a Levine shunt was inserted and drained via the right internal jugular vein; her ascites improved. Scanning after red blood cells had been labelled with technetium-99m showed an unusual vascular pattern in the inferolateral aspect of the liver. At repeat laparotomy the left hepatic lobe contained large bullae filled with blood, which were excised. The venous tip of the Levine shunt became obstructed in October 1985 and required replacement with a similar shunt to the left internal jugular vein. Subsequent scanning showed retrograde flow around this shunt but not down the superior vena cava. In February 1986, aged 34, she collapsed with angiographically proved emboli in both main pulmonary arteries. Streptokinase was thought to be contraindicated and embolotherapy was unsuccessful. Necropsy showed organised thrombus extending through most of both main pulmonary vascular trees as well as lymphangiomas of the liver, mediastinal pleura, kidneys, pancreas, and heart. The right atrium contained friable adherent clot 3 cm long.

Comment

Splenomegaly due to cystic lymphangiomatosis is rare. A review of reports between 1940 and 1952 identified 27 cases.1 Associated ascites is usually chylous and implies coexistent retroperitoneal lymphatic disease. Such ascites has been successfully treated with a Denver peritoneal venous shunt.2 Severe haemorrhagic ascites in a patient with normal coagulation has to our knowledge not been reported before.

The blood in the ascitic fluid may well have obstructed the first shunt in our patient. The second shunt was obstructed by a fibrous sheath. This complication is well recognised,3 and research into non-silicone materials that cause less fibrosis is now in progress. Caval thrombi are a recognised and dangerous complication of Levine shunts and require anticoagulation. Unfortunately, our patient's recurrent emboli were both silent and untreatable as long term treatment was contraindicated by continuing bleeding.

We thank Mr R Norton, Dr K Hollinsrake, and Dr T Ashworth for their help with this case.


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Strongyloides stercoralis infection in Burma Star veterans

Infections with the nematode worm *Strongyloides stercoralis* may persist for many years after exposure, by autoinfection. The condition is well described among former second world war prisoners of the Japanese4 and occurs in over a fifth of those who worked on the infamous Thai-Burma railway.5 Most infections cause a "creeping eruption" or "larva current" eruption—an itchy, serpiginous urticarial rash which occurs intermittently, usually over the trunk. The real danger of strongyloidiasis, however, is the potentially fatal hyperinfection syndrome, which may occur when infected subjects become immuno suppressed—particularly with corticosteroids.6

Soldiers of the second world war Burma campaign fought in areas endemic for strongyloidiasis. They usually had reasonably adequate footwear, however, making them much less liable to infection (which is acquired by skin penetration of free living soil larvae). Nevertheless, we have recently described a case of strongyloidiasis infection in a British Burma Star veteran,7 diagnosed 40 years after tropical exposure. We therefore attempted to determine the prevalence of strongyloidiasis among this group of men.

Patients, methods, and results

We circulated a questionnaire at two reunion meetings of the Burma Star Association in 1985 in Bridlington and London. These questionnaires asked whether members suffered a creeping eruption type of rash, which was described in detail (colour photographs of the rash were also distributed). Of 566 who responded positively were contacted further by post, and if their rash was considered typical investigations were arranged (three stool samples for microscopy and larval culture, blood eosinophil count, and an enzyme linked immunosorbent assay (ELISA) serum test for strongyloides). Of 566 who replied to the questionnaire, three men were finally found to have strongyloidiasis—a prevalence of 0.53%. These were all successfully treated with mebendazole.
Comment

In ex-Far East prisoners of war 84% of those with strongyloides have the typical rash. Given also the accepted limitations of our survey, this suggests that the prevalence of 0-5% for Burma Star veterans is an under-estimate. Nevertheless, even this figure means that some 100 to 200 Burma veterans in Britain today have undiagnosed strongyloides infections. Although the infection rate in ex-Japanese prisoners is much higher, doctors should also be aware of the possibility of strongyloides in veterans of the Burma campaign.


(Accepted 13 February 1987)

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β Endorphin: A factor in "fun run" collapse?

Over the past six years 38 entrants have collapsed near the finish of Tynedale's annual Great North Run (a half marathon "fun run"). Though they represent a very small proportion of the entry, which now exceeds 25 000 a year, we find it surprising that healthy men can run until they become confused, dehydrated, hyperthermic, and hypophosphatemic

without first experiencing intolerable discomfort. Because endogenous opioids suppress pain, they have a possible role in temperature regulation, and may be responsible for "runner's high". The concentration of β endorphin—one of the most potent of these peptides—is worth considering as an important factor facilitating collapse during such runs.

Subjects, methods, and results

Blood samples were obtained from 11 runners who collapsed near the end of the two consecutive Great North Runs and from a control group at both the start and finish of the race. Fortunately, there were also 11 controls. Both groups comprised men of modest but previously undistinguished performance. The collapsed runners were aged 19-43 (median 27) and the controls 27-45 (median 37).

The plasma was separated immediately and stored at -20°C. As soon as was practicable after each race duplicate assays of plasma β endorphin were performed by radioimmunoassay, after immune affinity chromatography, using a method giving a cross reaction with β lipotropin of less than 5%. The figure shows the results.

The control group had a mean β endorphin concentration of 8.4 (4-2) pmol/l before the run and 27.2 (9-7) pmol/l (SD) after the run. Two runners showed slightly increased initial concentrations, perhaps as a result of exercise or anxiety before the race. The highest control concentration after the run was 46.2 pmol/l. The collapsed group at the finish had a median β endorphin concentration of 110 pmol/l (range 66-414 pmol/l). The concentrations in the controls after the run were significantly higher than the corresponding starting concentrations (p<0.005, Wilcoxon signed rank test) and were themselves considerably exceeded by the concentrations found in those who collapsed during or at the finish of the race (p<0.001, Mann-Whitney rank sum test).

Comment

Though we cannot know the β endorphin concentrations of the runners before collapse, and they may reasonably be assumed to increase during the process of collapsing, the evidence from the control group is that the concentration of β endorphin are already high before this happens. Juntal et al showed in a double blind study that long distance runners experience hypoaesthesia and "runner's high" and that the effects are associated with an increase in β endorphin concentrations and are inhibited by naloxone. We suggest that the unusually high concentrations of β endorphin in those who collapsed were probably responsible for the insensitivity to pain, enabling the runner to keep going. The sense of wellbeing produced by opioid peptides may be a factor that determines the competitor's enthusiasm for running. At times this seems to be extraordinary: of the men we studied, one had collapsed during a previous half marathon, and another, who spent considerable time in intensive care after his run, subsequently expressed the intention to continue participating in such events.

Treadmill exercise produces an increase in β endorphin concentration in both trained and untrained subjects. Gambert et al found this to be much greater in men than in women. Perhaps this accounts for the fact that over the past six years all of the runners who collapsed at the end of the Great North Run have been men.

Doubtless the potential collapsing is caught up in the group enthusiasm that surrounds the run and is subjected to the pressures of self esteem and perhaps the knowledge that considerable sums of money given in sponsorship for charities are at risk. The important factor that enables such entrants to run until they collapse, however, is probably the high concentration of circulating endogenous opioids.


(Accepted 28 January 1987)

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Intensive care: a specialty or a branch of anaesthetics?

Sir,—It is remarkable that over two months elapsed before there was a published comment on Professor Hugh Dudley's leading article on intensive care training (21 February, p 459). We have been reticent in replying because our own proposals for a pilot senior registrar training scheme in the South Western region were being considered by the Intercollegiate Committee. Nonetheless, it is of the utmost importance to encourage all anaesthetists to accept the challenge of the liaison group on intensive therapy. We now understand that our scheme has been approved, and we therefore welcome Professor Iain Ledingham's letter (27 April, p 1098), with its emphasis on the importance of encouraging without delay training programmes recommended by the liaison group.

There are two main obstacles to achieving this. The first is funding. The liaison group was set up jointly by the Faculty of Anaesthetists, the Royal Colleges of Surgeons of England and Scotland, and the Royal College of Physicians. Its proposals have been accepted by the conference of royal colleges and their faculties. The agreement achieved is remarkable if not unique. It is difficult to believe that the conference cannot persuade the Department of Health and Social Security of the need to fund a small number of posts, unless those who have supported the liaison group publicly are privately hoping that lack of funds will ensure its premature death.

The second problem is the anxiety among some anaesthetists that the proposals will undermine their role in intensive care and interfere with the training of anaesthetic registrars and senior registrars. For years anaesthetists have borne the main burden of intensive care in this country, and they will continue to do so. Trainees in other disciplines, however, also wish to pursue a career in intensive care. It is essential that they must have completed all three parts of the FFARCS examination before being allowed to do so. We think not. The liaison group's proposals and our own South Western training scheme are designed to improve the training of those doctors who wish to have intensive care as a major component of their consultant career, and who include anaesthetists. Furthermore, the presence of a senior registrar in an intensive care unit may actually improve the training of registrars as he or she will be more readily available to teach than many consultants.

So far as senior registrars in anaesthesia are concerned, it should be possible to maintain the amount of training in intensive care, even though there may already be a senior registrar on the intensive care unit in a hospital to which the new trainee rotates. In many regions, however, training of senior registrars in intensive care is limited by the demand for training in subspecialties within anaesthesia. It is essential that all anaesthetists support these proposals.

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Sue's breast lump

Sir,—Dr Stephen Platt assessed the influence of an attempted suicide in EastEnders on the number of subsequent overdoses (11 April, p 954). Another character in the same programme has recently had a breast lump diagnosed and treated.

John F Searle

30 May 1987

Angie's overdose

Sir,—Dr Stephen Platt's study (11 April, p 954) failed to show a strong imitation effect after televised parasuicide. His data were obtained by letter from accident and emergency departments throughout Britain. Only 41% of the original total provided usable returns for 1985 and 1986. Collecting numbers of overdoses from accident and emergency registers is tedious and time consuming, and there is doubt about the accuracy of responses obtained from poorly motivated administrative staff. For the Hackney Hospital Platt recorded 11 overdoses during the experimental period, while a more careful look at the records reveals 22.

Dr Platt's results do show a significant increase in the number of overdoses for the 18 days after the first screening of Angie's overdose compared with the 18 days before it (p<0·001, sign test). The question remains whether there is a causal relation between the EastEnders programme and this finding.

By choosing the week before the overdose as his control week Platt may be underestimating the initiative effect by including subjects who had been influenced by Angie's plight but who beat her to the inevitable conclusion. Although the overdose was shown on Thursday 27 February 1986, the plot had been building up to it for some time. Angie's overdose was not so much an isolated event but the culmination of a process—an overdose is for many people.

Although the medical and scientific community should understand the significance of a single incident, the Independent Broadcasting Authority and the British Broadcasting Corporation may misinterpret these results as a licence to continue the gratuitous exploitation of parasuicide in soap operas. Since our publication of the initial observed increase in overdoses following Angie's1 the IBA may have thought twice about the portrayal of self harm, but not so the BBC. On 17 October 1986 Amanda in Dynasty took an overdose. She was showered with affection and given a diamond necklace for her pains. The only other way to obtain a diamond necklace in Hackney is to rob a jeweller.

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1 Ellis SJ, Walsh S. Soap may seriously damage your health. Lancet 1986; i:686.

β Endorphin: a factor in “fun run” collapse?

Sir,—Dr G Dale and colleagues (18 April, p 1004) claim that a high concentration of circulating β endorphin is associated with the collapse of some men during “fun runs.” Runners who collapse are usually those who are ill prepared and underetrained, and an increased plasma concentration of β endorphin is a non-specific response to acute stress.1 Plasma β endorphin and adrenocorticotrophic hormone are derived from the pro-opiomelanocortin precursor in the pituitary, pro-opiomelanocortin, which secretes both hormones in response to the same stress.2 Thus cortisol concentrations are also likely to be increased in runners who collapse. The exact role of plasma β endorphin in man is not known, but it may modulate the release of circulating catecholamines and the renin-aldosterone system.3 As plasma β endorphin is fairly impermeable to the blood-brain barrier except at the hypothalamus it cannot be implicated in the “runner’s high” or in driving a runner to collapse.4 Central changes in opioid peptide concentra-
Corticosteroids and bone mass in asthma

**Str—** Dr G J Addis (3 July, p 644) is concerned that we should respond to Dr G K Crompton’s thoughtful criticism (10 January, p 123) of our paper (6 December, p 146).

In his letter Dr Crompton drew attention to the difficulties of obtaining detailed information retrospectively from hospital records and expressed the belief that some of the patients studied who were under his own care may have received additional courses of systemic corticosteroids that were not recorded. As Dr Crompton is a close colleague working in a neighbouring unit that was collaborating in our study, our initial reaction was to ask him to make his own independent assessment of the corticosteroid intake of the patients concerned. Subsequently, however, Dr Addis’s letter has prompted us to review our records, as carefully as possible, the case records of all patients in group 3 who were classified as having received only inhaled steroids, with or without short booster courses of systemic corticosteroids, and to supplement this information with information obtained from the patients themselves and, in some cases, their general practitioners.

Twenty out of 22 case records were traced, and four discrepancies have been ascertained. One man (aged 21) who was classified as having received no oral corticosteroids was found to have received intermittent systemic corticosteroids for several years before total body calcium was measured, although not in the two years immediately before bone mass was estimated. Exclusion of this patient from the group leaves a total of 21 patients (10 men and 11 women), with a mean (SD) age of 57.2 (2.7) years. Of the 40 recorded controls, mean total body calcium was still reduced by 8.4% (p<0.001). Three discrepancies were found in the calculation of the number of booster courses of oral corticosteroids. Two men, previously classified as not having received any, had in fact received two and one 10 day booster courses of steroids. Another man thought to have had only one booster course seems to have had at least five, and five of the patients treated with both inhaled steroids and booster courses had in fact taken higher doses of inhalation steroids for shorter periods. This is impossible in a retrospective study of this kind to determine the relative importance of the inhaled corticosteroids and the intermittent short booster courses of systemic steroids in the associated reduction in total bone mass, but a recent study of bone metabolism, but a recent study of bone metabolism, but a recent study of bone metabolism, but a recent study of bone metabolism, but a recent study of bone metabolism, but a recent study of bone metabolism, but a recent study of bone metabolism, but a recent study of bone metabolism, but a recent study of bone metabolism, but a recent study of bone metabolism, but a recent study of bone metabolism, but a recent study of bone metabolism...