Short report

Combined medical and surgical treatment of spinal hydatid disease: a case report

AGTW FIENNES,* DGT THOMAS

From the Gough-Cooper Department of Neurological Surgery, Institute of Neurology, The National Hospital, London

SUMMARY A case of chronic hydatid disease of the dorsal spine is described. The patient had undergone four previous operations over a 26 year period and had suffered recent further deterioration leading to complete paraplegia. On this occasion she was treated by a combination of drug therapy and surgery. She showed some neurological recovery after a two and a half month interval, without evidence of recurrence. The surgery would not have been feasible without concurrent drug therapy.

Spinal hydatid disease has hitherto carried a prognosis of inevitable paraplegia¹ and surgical treatment by laminectomy has at most postponed the time when advanced cases become irrecoverable. The authors report treatment of a severe long standing case with high dose mebendazole as well as anterolateral spinal decompression in an effort to gain more permanent improvement.

Case report

A 39-year-old Iranian woman was admitted from a rehabilitation centre unable to walk, with increased left-sided sensory loss and one week's sudden increase in thoracic back pain. Previously she had walked with a frame. For some years her attacks of bronchospasm had required broncho-dilators. Twenty-six years previously, following the onset of mild paraparesis, upper dorsal laminectomy was performed with removal of several hydatid cysts. Thirteen, 18 and 24 years later she underwent further upper dorsal spinal surgery for similar symptoms. Following the last intervention she became paraplegic and was expected to remain so, but nonetheless gradually improved over the ensuing two years, so as to be mobile with some bladder sensation one week before the current admission. During this rehabilitation she received mebendazole 400 mg twice daily, increasing to 500 mg four times daily. Her general condition was good, without obvious respiratory embarrassment but with marked upper

thoracic kyphoscoliosis. The neurological picture was that of a severe paraparesis and superimposed Brown-Sequard syndrome with dense unilateral global sensory loss below D4 anteriorly and D10 posteriorly. There was a 9% eosinophilia and plain radiographs showed fracture dislocation with forward and right-sided shift of the D3 vertebra on D4. Lumbar metrizamide myelography (Fig 1 a, b) showed substantial angulation of the cord in both planes. Immediate subsequent CT scan of the region (fig 2) further demonstrated the extent of bony destruction, revealing also multiple smooth filling defects impinging on the cord. Bilateral soft tissue masses bulged into the pleura, but chest radiographs showed the lung fields were clear.

By right D3-4 costotransversectomy the theca was exposed and found running horizontally in an anteroposterior and right to left direction. Several large cysts were encountered during the operation and evacuated. Two smaller cysts were also removed from the spinal canal, and the dura contained no cysts. The posterior part of the kyphus formed by the vertebral bodies was resected to allow a less acute angulation of the cord, which was only 5 mm in diameter. It was necessary to open the pleura. Histology showed degenerate hydatid material only. The patient spent six weeks after operation on a Stryker frame and first sat three weeks after that. At no time was she subject to broncho-spasm, but there was continuous pyrexia up to 38°C for six days, unresponsive to antibiotics and with negative bacteriology. Eosinophilia persisted up to 18% for a month and mebendazole therapy at the higher dose was continued until, 2 months after operation, she developed an acute psychosis.

Initially neurological recovery was slow, but when she was reviewed 13 weeks post operatively she had a suspended global sensory depression in the right D4 dermatome and loss of fine touch and joint position sense in the left foot. The spasticity had substantially decreased

Address for reprint requests: Mr DGT Thomas, Gough-Cooper Department of Neurological Surgery, Institute of Neurology, The National Hospital, London WC1E 3BG, UK

Received 10 June 1980 and in revised form 15 April 1982.
Accepted 24 April 1982

Fig 1 Metrizamide myelogram of the upper dorsal region (a) oblique, (b) right lateral: the theca is compressed by a smooth mass as well as by the fracture dislocation. The lower theca is seen in the vertical plane, whilst above it runs horizontally.

Fig 2 Metrizamide contrast CT scans of the lower border of (a) and through the body (b) of the D3 vertebra: Note the soft tissue masses bulging into the pleural space and the disorganisation of the posterior elements.

with assisted voluntary movements at both knees and ankles.

Discussion

With a world-wide incidence far lower than hepatic or pulmonary involvement,³ bony hydatid disease is uniformly due to *Echinococcus granulosus*.² Spinal deposits, mainly dorsal, account for half of this^{1 3 5} because of porto-vertebral venous shunts, although there is wide variation in the frequency of cord compression.¹ Exogenous vesiculation¹ leads to a characteristic process of indolent bony destruction, the vertebral discs representing relative barriers to spread.^{1 3 5} Treatment has hitherto been entirely surgical as there was not considered to be any effective medical alternative.⁴ For soft tissue disease, surgical strategy varies widely between radical clearance with a risk of spread and anaphylaxis, followed by lavage to counter spillage,⁴⁻⁶ and simple instillation of scolicide.⁷ Diffuse bony disease of the dorsal spine frequently requires piece-meal surgical removal since there are no large cysts such as those seen in the soft tissues. Relapse is common,^{3 4} or the rule¹ after a variable interval. A purely palliative stance is that of Bettaieb and colleagues who regard the disease as "practiquement maligne." They regard surgical intervention as temporising only and paraplegia inevitable.¹

The benzimidazole derivative mebendazole (Vermox JANSEN), marketed for intestinal worm infestations, was noted by Heath and Chevis⁸ to have a lethal effect on secondary *E granulosus* cysts in mice, suggesting a potential as an adjunct to surgery. Bekhti *et al* were the first to give a full report on its employ in human hepatic hydatidosis, using short high dose courses,¹⁰ although relapse prevention requires other regimes,^{6 11 12} because of poor absorption and diffusion.¹¹ Despite both theoretical doubts of its efficacy¹³ and recovery of intact but sterile cysts from mebendazole treated patients,¹⁴ the drug may in man only prevent evolution rather than eradicate the disease.^{6 15} Few side effects are reported over periods of treatment up to three years at the currently recommended dose.¹² Initial pyrexia is reported in several patients and may be due to transient toxicity or systemic passage of necrotic hydatid material.^{6 18} Definitely allergic phenomena during treatment have not been documented in man. Some are hopeful that mebendazole will in time replace surgery.^{15 18} Others believe it is only useful in cases of surgical inaccessibility,⁷ and its benefit species dependant.¹¹

Our patient's exacerbation of chronic recurrent disease presented with the classical triad⁴ of pain, deformity and neurologic deficit and we believe the

rapid deterioration represented further displacement due to continuing active bony destruction, not controlled by the initial low dose of mebendazole. She was comparable to other reported cases. Werczeberger *et al*⁶ reported cough and pyrexia as features of allergy to hydatid antigen. Whilst on mebendazole therapy our patient showed no bronchospasm and whilst her pyrexia may be of similar origin to that reported by others^{6 13 16 17} she did not exhibit florid anaphylaxis following piecemeal surgery to remove large quantities of hydatid material.

In conclusion, we believe drug therapy and surgery to occupy truly complementary roles in the management of such a case. Mebendazole had nothing to offer for the chief symptom, paraplegia. On the other hand, surgery alone carried an unacceptable risk of acute allergic phenomena and early recurrence in an already severely handicapped patient. The prevention of recurrence can only be judged in long term retrospect but we see no reason why mebendazole should be less successful in this type of patient than in others.^{12 19 20}

References

- Bettaieb A., Khaldi M, Ben Rhouma T, Touibi S. L'echinococcose vertebro-médullaire: a propos de 32 cas. *Neurochirurgie* 1978;**24**:205-10.
- Vasal PC, Sharma VP, Agarwal RF. Jugular foramen syndrome due to hydatid cyst. A case report. *Neurology (India)* 1978;**26**:74-5.
- Morshed AA. Hydatid disease of the spine. *Neurochirurgia* 1977;**20**:211-5.
- Savini R, Capelli A. Vertebral localization of echinococcosis. *Ital J Orthop Traumatol* 1978;**4**:99-104.
- Fenyés G, Ladvaszky C. Echinokokken im Wirbelkanal. *Zbl Neurochirurgie* 1977;**38**:157-64.
- Werczeberger A, Golhman J, Wertheim G, Gunders AE, Chowars, I., *et al*. Disseminated Echinococcosis with Repeated Anaphylactic Shock treated with mebendazole. *Chest*, 1979;**76**:482-4.
- Nott DB. Hydatid disease in man—past, present and future. *Aust Vet J* 1979;**55**:136-9.
- Heath DD, Chevis RAF. Mebendazole and hydatid cysts. *Lancet* 1974;**2**:218-9.
- Goodman HT. Mebendazole. *Med J Aust* 1976;**2**:662.
- Bekhti A, Schaaps JP, Capron M, Dessaint JP, Santoro F, Capron A. Treatment of hepatic hydatid disease with Mebendazole: preliminary results in four cases. *Br Med J* 1977;**2**:1047-51.
- Keystone JS, Murdoch JK. Mebendazole. *Ann Int Med* 1979;**91**:582-6.
- Wilson JF, Davidson M, Raush RL. A clinical trial of mebendazole in the treatment of alveolar hydatid disease. *Am Rev Resp Dis* 1978;**118**:747-57.
- Osborne DR. Mebendazole and hydatid disease. *Br Med J* 1980;**1**:183.

- ¹⁴ Al-Moslih, MI. Treatment of hydatidosis. *Br Med J* 1978;**2**:1435.
- ¹⁵ Beard TC, Rickard MD, Goodman HT. Medical treatment for hydatids. *Med J Aust* 1978;**1**:633-5.
- ¹⁶ Murray-Lyon IM, Reynolds KW. Complications of mebendazole treatment for hydatid disease. *Br Med J* 1979;**2**:1111.
- ¹⁷ Harris A. Pyrexia and mebendazole. *Br Med J* 1979;**2**:1365.
- ¹⁸ Anon. Medical treatment for hydatid disease? *Br Med J* 1979;**2**:563.
- ¹⁹ Gamble WG, Segal M, Schantz PM, Rausch RL. Alveolar hydatid disease in Minnesota: first human case acquired in the contiguous United States. *JAMA* 1979;**241**:904-7.
- ²⁰ Ludin CE, Gyr K, Karoussos K. Therapy of alveococcosis in man. *J Int Med Res* 1977;**5**:367-8.
- ²¹ Macnair AL. Mebendazole and hydatid disease. *Br Med J* 1980;**1**:1055.