Case Report

BILHARZIAL GRANULOMA OF THE SPINAL CORD

A. M. BARNETT,* M.B., B.CH. (RAND), F.C.P. (S.A.), *Department of Medicine, University of Natal, Durban*

Bilharzia is endemic in many parts of Southern Africa and most commonly causes disease of the genito-urinary system. Involvement of the spinal cord is relatively uncommon and it is thought worth while to report the following case.

CASE REPORT

History

A 35-year-old White male, a sheet metal worker, was admitted to Addington Hospital on 23 January 1961. Eight days previously he had developed a feeling of severe discomfort over his lower back, abdomen, scrotum and penis. He described this as being similar to severe sunburn and he could not tolerate any pressure over the affected areas. This feeling became less intense after 3 or 4 days. Seven days before admission he began experiencing a tingling pain in the lumbosacral region. This pain radiated through the buttocks into the legs and feet and had gradually become worse. For 2 days he had noticed mild stiffness of the legs and slight hesitancy and intermittency during micturition.

Fifteen months previously he had been swimming in a lagoon known to contain infected snails and after leaving the water he had noted itching. A few weeks later he developed a vague illness characterized by muscle pains and recurrent bouts of fever. After investigation by his doctor he was given 10 intravenous injections for bilharzia. He then remained well until the present admission.

*Present address: Department of Neurology, Royal Victoria Infirmary, Newcastle-upon-Tyne, England.

Signs and Symptoms

Examination showed a well-nourished, extremely anxious young man. The heart rate was 80/min. and the blood pressure was 150/95 mm.Hg. He was apyrexial and there were no abnormal findings in the heart, lungs or abdomen. The cranial nerves and fundi were normal. There was slight weakness of all movements at the hips, knees and ankles. The tendon reflexes in the legs were hyperactive and the plantar responses were flexor. There was hyperaesthesia to light touch and pinprick over a large area corresponding to segments D8 to L3.

Investigations gave the following results: haemoglobin 17·3 G/100 ml.; haematocrit 49 vols. %; ESR (Wintrobe) 10 mm. in 1 hour; leucocytes 13,000 per cu.mm. with 47% polymorphs, 18% lymphocytes, 1% monocytes and 34% eosinophils; blood urea 18 mg./100 ml. Urine examination was normal. The cerebrospinal fluid was clear and colourless and under a pressure of 120 mm. of water and there was no block. The fluid contained 2 polymorphs/cu.mm.; 18 lymphocytes/cu.mm.; protein 60 mg./100 ml.; sugar 47 mg./100 ml.; chloride 709 mg./100 ml. The fluid was sterile on direct examination and culture and no virus was isolated in tissue culture. X-ray examination

of the spine showed loss of the normal lumbar lordosis and slight degenerative changes at D12 - L1.

Course

During the next 3 days his signs gradually progressed until there was complete paralysis of the legs and bladder and loss of all forms of sensation below D10 with a band of hyperaesthesia to touch and pinprick at D8.

Lumbar myelography showed a partial hold-up and a filling defect extending from the disc space between D12 and L1 to the disc space between L1 and L2. The cord was displaced to the left side (Fig. 1). Repeated stool examinations and 24-hour

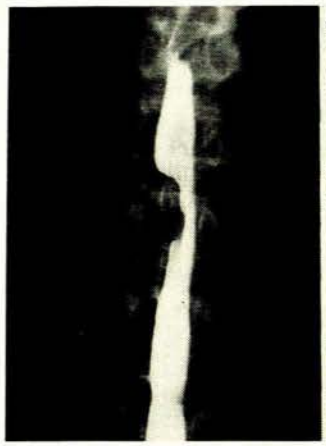

Fig. 1

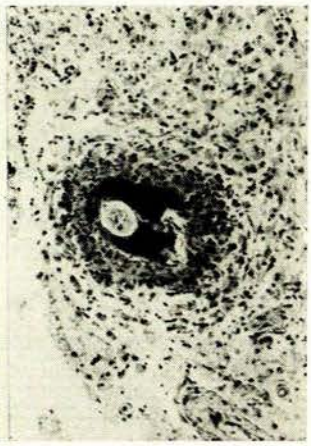

Fig. 2

Fig. 1. Lumbar myelogram showing indentation of Myodil column at the level of L 1, and slight displacement of the spinal cord to the left. *Fig. 2.* Section of granulation tissue removed at operation, showing bilharzia ovum surrounded by necrotic tissue, lymphocytes, plasma cells and macrophages.

specimens of urine failed to show any parasitic ova. Biopsy of the rectal mucosa was normal.

On 4 February 1961 an exploratory laminectomy was performed by Mr. M. J. Joubert. The spinal cord at L1 was found to be involved by a large mass which macroscopically appeared to be malignant. In view of this a wide excision was made. Histological examination of the specimen showed the presence of ova of bilharzia (Fig. 2).

Postoperatively he has remained with a complete paraplegia with a sensory level at D10. When the nature of the condition was known he received a course of sodium antimony tartrate. He has a chronic urinary infection but has shown great determination and has returned to his original employment.

DISCUSSION

Although infection with *S. haematobium* and *S. mansoni* is very common in Natal, serious clinical manifestations are comparatively unusual. Spinal cord involvement is extremely rare and only 12 cases with histological proof have been described.[2-13]

Spinal cord involvement may occur at any age but only 3 patients were more than 35 years of age. Ten of the patients were males and 2 were females. In all reports where details have been given the lesion has been in the lower dorsal or lumbar segments of the cord. All cases had paraplegia and in the majority this had developed within a few days of the initial symptoms. The CSF was abnormal in 8 of the 10 patients of whom details are given. A slight pleocytosis and a small rise in protein were the commonest abnormalities, but Froin's syndrome has been described.[4,12,13]

Gelfand[1] examined the brains of 50 patients known to have had bilharzia, by means of digestion with potash and was able to demonstrate ova in 28 cases. Ova were found in only 1 of 25 spinal cords subjected to the same examination. The adult worm is believed to gain access to the spinal canal through branches of the pelvic veins which anastomose with the vertebral venous plexus. A variety of lesions have been described and include oedema, demyelinization, venous occlusion and granulomata.

In this patient the course of events can be traced from the time of the initial infection. Exposure was followed by an illness suggestive of the Katayama syndrome. After a course of treatment he remained asymptomatic although uncured.

It is important to remember that the larval stage of schistosomiasis is not affected by antimony preparations. If the larval stage of schistosomiasis is suspected, treatment should be withheld for 4 - 6 weeks in order that the mature worms may be attacked. Although bilharzia was considered as the cause of this patient's spinal cord lesion he was referred for neurosurgical treatment because of the findings at myelography. The macroscopic appearance suggested malignancy and a wide excision was made.

In view of the essentially benign nature of this disease, patients with spinal cord compression in whom there is a history of bilharzia or who have an unexplained eosinophilia should be intensively investigated before radical surgery is undertaken. This should include rectal biopsy and urine and stool examination. If a granulomatous lesion is found at operation a biopsy may save the patient from an extensive surgical procedure. In otherwise unexplained cases of myelitis it is worth while treating the patient with antimony if any suspicion of bilharzia exists. Antimony is effective against the adult worm only but this will prevent further deposition of ova. Spontaneous clinical improvement may occur as the inflammatory process subsides. In this patient it is impossible to know what the outcome would have been with conservative treatment, but the final result may have been less crippling.

SUMMARY

A case of bilharzial involvement of the spinal cord is described. Patients suspected of having bilharzial involvement of the spinal cord should be intensively investigated before undergoing radical surgery.

I should like to express my thanks to Dr. J. V. Tanchel, Superintendent of Addington Hospital, for permission to publish this report.

ADDENDUM

Since this paper was submitted for publication a further report on the spinal cord complications of bilharzia has appeared.[14] The author describes 9 cases, 6 of radiculitis and 3 of transverse myelitis. Histological proof was obtained in 3 cases. The author emphasizes the importance of early diagnosis and treatment.

REFERENCES

1. Gelfand, M. (1950): *Schistosomiasis*, pp. 197-201. Cape Town: Juta.
2. *Idem* (1950): *Ibid.*, p. 196.
3. Hutton, P. W. and Holland, J. T. (1960): Brit. Med. J., **2**, 1931.
4. Raper, A. B. (1948): E. Afr. Med. J., **25**, 262.
5. Gama, C. and De Sa, J. M. (1946): Trop. Dis. Bull., **43**, 936.
6. Ross, G. L. *et al.* (1952): New Engl. J. Med., **246**, 823.
7. Day, H. B. and Kenawy, M. R. (1936): Trans. Roy. Soc. Trop. Med. Hyg., **30**, 223.
8. Muller, H. R. and Stender, A. (1930): *Op cit.*[7]
9. Bayoumi, M. L. (1939): *Op. cit.*[1]
10. Espin, J. (1941): *Op. cit.*[6]
11. Abbott, P. H. and Spencer, H. (1953): Trans. Roy. Soc. Trop. Med. Hyg., **47**, 221.
12. Maciel, Z. *et al.* (1954): Rev. neurol., **91**, 241.
13. Pepler, W. J. and Lombard, C. M. (1958): J. Neuropath. Exp. Neurol., **17**, 656.
14. Bird, A. V. (1965): S. Afr. Med. J., **39**, 158.