CASE REPORTS

HYDATID CYST OF THE SPINAL CANAL SUCCESSFULLY TREATED BY OPERATION

BY

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This case of solitary hydatid cyst of the spinal canal is reported not only for its rarity but also because it shows that the results of operative treatment of this condition, of which the records in this country are comparatively few, may be dramatically successful.

Clinical record

A girl, aged nine years, the daughter of poor parents, began to complain in October, 1933, of pains in the thighs and, later, in the lower part of the spine. Early in 1934 she attended the surgical out-patient department and in February was admitted to a medical ward for observation. Radiological examination of the spine and of the hip-joints revealed no abnormality; the electrical reactions were normal except for a slight weakness of some of the muscles of the right leg. As nothing was found to account for her symptoms, she was discharged after eight days with the diagnosis of subacute rheumatism and resumed her attendance at the surgical department. During the next ten weeks her condition steadily became worse, and on May 1, 1934, she was examined by one of us (J.S.Y.R.). The following is a brief account of the history and clinical signs then elicited:

In the family history there was nothing noteworthy and the patient herself had always been strong and healthy until the onset of her present illness. The pains, of which she had now been complaining for about seven months, occurred first of all in the left thigh and knee and later in the right thigh; they were not severe to begin with, but had gradually become worse. Early in 1934 bouts of pain, localized in character and always in the same spot, developed in the lower part of the back. This pain was not present when she was walking, but came on when she was sitting in a chair or lying in bed, and was then so sharp and agonizing that it kept her awake and crying most of the night. She had also some pain in the left hip, which made her walk with a limp. In the early part

of the year, in her father's words, 'She could run a hundred yards like a hare and then collapse'; now she could walk only a short distance, and when going upstairs had to haul herself up by the hand-rail.

Examination did not reveal any spinal deformity or any sign of rigidity. Bending the body backwards did not cause pain, and if pain were already present did not increase it. The site of the pain was in the region of the three upper lumbar vertebrae; but pressure over that area did not evoke it, although there was a slight hyperaesthesia of the skin. She had a peculiar waddling walk, with a limp on the left side. Passive movements showed no limitation of movements at hips or knees, but there was some pain on movement at the left hip-joint. The muscles of both legs were soft, flaccid and somewhat atrophied. The power to resist movements of the legs and the freedom and rapidity of active movement were all diminished. The knee-jerks were absent and there was anaesthesia of the legs. There was no disturbance of function of bladder or bowel. An examination of the blood showed haemoglobin 83 per cent., red cells 5,000,000 and white cells 7,300 per c.mm. No abnormal cells were found and no increase in eosinophils.

On May 7, an x-ray examination was again made, and the radiologist reported that he failed to find evidence of bone disease either in the hips or in the lumbar spine.

From the persistent recurrence of localized pain in the upper lumbar spine, the pain in the thighs, pointing to a referred pain from pressure in the same region of the spine, and the flaccid paresis of the legs such as occurs in a compression myelitis, the tentative diagnosis was made of intraspinal tumour and the case was referred for a surgical opinion to the late Professor John Anderson. Considering her peculiar walk and limp and the pain on movement at the left hip, Professor Anderson inclined to the view that there might be an early tuberculosis of the hip-joint, even although x-ray examination had shown nothing abnormal, and arranged to admit her to his ward when a bed was available. By June 7, however, her condition was changing so rapidly for the worse that she was admitted as an urgent case. On the following day cisternal puncture was performed under light anaesthesia and lipiodol injected; the skiagraph showed spinal blockage at the level of the first lumbar vertebra (fig. 1).

On June 28, under rectal ether, Professor Anderson exposed the spines from the twelfth dorsal to the fourth lumbar, and performed laminectomy of the first, second and third lumbar vertebrae. A greyish-white cystic mass was found lying extradurally in the canal and was easily shelled out. The cyst extended from the lower border of the first lumbar to the upper border of the third lumbar vertebra, measuring about 2 inches in length by $\frac{3}{4}$ inch in greatest breadth. The results of the operation were excellent. By August 4 the patient was up and walking with support; by August 21 the limp had gone, and although still unsteady she was walking much better; by August 30 sensation was normal, and, except for a little difficulty in balancing, she was walking very well. She was discharged from hospital on September 12, 1934, completely cured. An x-ray examination made later showed that the lipiodol now passed well down to the lower end of the canal (fig. 2). When last seen in April, 1938, she looked strong and fit and showed no abnormality in her walk.

Pathological examination

The specimen consisted of the two halves of an ovoid unilocular cyst measuring approximately $1\frac{1}{2}$ inches in length by $\frac{3}{4}$ inch in greatest breadth. The wall of the cyst was composed of a whitish, gelatinous, translucent tissue resembling coagulated egg-white, and was apparently homogeneous throughout.

Both the external and internal surfaces were perfectly smooth and there were no ingrowths. On the inner surface one minute pinkish spot was just visible to the naked eye, and on microscopic examination of a scraping from the wall at this spot, several typical hydatid hooklets were found amongst amorphous debris (fig. 3). From the characters of the cyst wall and the size of the hooklets it was clear that the mass from the spinal canal was the cystic stage of *taenia echinococcus*. A section from the cyst wall showed the laminated cuticula and lining proliferative layer of a hydatid ; no scolices were found. The cyst

FIG 1.—X-ray five hours after injection of lipiodol, showing spinal blockage at the level of the first lumbar vertebra.

having ruptured, no fluid was available for examination. The source of the infection was not discovered ; the patient was not known to have been in contact with dogs.

Commentary

The spinal canal is one of the more unusual sites of hydatid cyst.

Neisser, in his collection of 986 cases of echinococcus disease in the literature, found only thirteen cases with cysts in the spinal canal (quoted by Leuckart, 1886). Lyon (1902) in an exhaustive review of echinococcosis in North America

Fig. 2.—X-ray showing lipiodol passing freely down to lower end of spinal canal.

FIG. 3.—Hooklets from hydatid cyst of spinal canal.

did not record a single instance of intraspinal cyst in the 241 cases of hydatid disease which he collected from the literature up to 1901.

In this country, where hydatid disease is uncommon, a cyst in the spinal canal is, at the present time, an extremely rare occurrence ; nevertheless records of a considerable number of cases are to be found in the English literature, especially in the latter part of the past century and in the early years of the present. In most cases the diagnosis was made post mortem and the records of successful operative treatment are few.

Ogle (1860) refers to a case of hydatid cysts within the spinous process of the seventh cervical vertebra and causing compression of the cord. Fagge (1871) mentions a case observed at post mortem, and Murchison (1885) a specimen in the Middlesex Hospital from a case of paraplegia caused by compression of the cord by secondary hydatids. Ransome and Anderson (1891) record a case in which laminectomy, performed for a supposed spinal tumour, failed to reveal the intraspinal hydatid cyst found later at autopsy. Colman (1899) quotes Gowers for a case of spinal hydatid in which 'Mr. Horsley trephined the spine and a quantity of cysts the size of peas were found compressing the cord.' Colman collected from the literature thirty-six records of spinal hydatids and added a case observed by himself of paraplegia produced by an extradural cyst in the spinal canal of a boy aged ten years ; the diagnosis was made post mortem. In a case recorded by Barrs and Trevelyan (1899) multiple hydatids were found in the spinal canal ; they had produced symptoms of acute myelitis. Tytler and Williamson (1903), in reporting successful operative treatment by laminectomy in a case of spinal hydatid cysts with severe compression myelitis, pointed out that a study of the recorded cases showed that spinal hydatids were usually external to the dura, and being mostly on the posterior aspect were usually favourably situated for operative removal. In a survey of hydatid disease in children including cases published since Lyon's paper of 1902 Mills (1926) found few in the spinal canal ; in addition to Colman's case, he mentions one reported by Fournier (1918) in a boy aged twelve, with successful operation ; the cyst had produced a paraplegia. He also quotes a record by Owen of Melbourne (1905) of five cases of spinal hydatids treated by operation. Another case of successful operation is reported by Gill and Bullock (1919). These authors comment on the small number of records of successful operations for hydatids in the spinal column, and consider that by the time a diagnosis can be made the disease is usually too extensive for operation. The preoperative diagnosis of hydatid of the spine can never, in their view, be more than a probability. Bériel and Leriche (1923) record a hydatid cyst of the sacrum which compressed the sacral nerves and caused sciatica ; in this case the preoperative diagnosis of hydatid disease was made tentatively. Recently in this country Lambert Rogers (1938) has removed from the spinal canal an extradural hydatid which had produced a compression paraplegia.

No attempt has been made to give a complete review of the literature on this subject and attention has been confined to illustrative cases, mainly from this country. In the literature of countries where, as in parts of South America and Australia, there is abundant clinical material for the study of echinococcosis, numerous examples of spinal hydatid, some with successful operative treatment, are to be found. But most of these records are in journals not available in this country, and it has not been possible to verify the references.

The present case emphasizes certain points which emerge from the records. It brings out the great difficulty which the diagnosis of hydatid disease of the

spinal canal always presents ; the intradermal reaction of Casoni and the complement-fixation test would doubtless have been of the greatest value, but in the present case, in a locality where hydatid disease even in its commoner sites is seldom met with, there was nothing in the history or in the preoperative clinical findings to suggest that echinococcosis should be considered in the differential diagnosis. On the other hand the case serves as a reminder that the site of election of hydatid cysts in the spinal canal, extradurally and on the posterior surface of the cord, facilitates operative removal, and that the results of operation are frequently most favourable.

Summary

A case is reported of hydatid cyst in the spinal canal of a child ; pressure on the cord had produced paresis and parasthesia of the lower limbs ; operative treatment by laminectomy resulted in a complete cure.

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