HYDATID DISEASE OF THE LUMBAR VERTEBRAE

Report of a Case

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The report of a single case of a rare localisation of hydatid disease needs to be justified. Vertebral hydatidosis is, fortunately, rare; Geroulanos in 1956 reported sixteen out of his series of 4,325 cases of hydatid disease at all sites (0.37 per cent). But the prognosis has been as gloomy as that of malignant disease of the spine, which is in sharp contrast to the good prognosis of hydatid disease at the more common sites. Further, not only does vertebral hydatidosis present great problems in its early diagnosis but also its successful management remains a persisting challenge.

For these reasons it is considered justifiable to report a study of a patient under treatment for twelve years, the last five of which have been spent under my care.

CASE REPORT

A student, now aged thirty-two years, spent his childhood in the Punjab, where he was in contact with hunting dogs. In 1951, after a fall at tennis, he developed pain in the left lumbar region, radiating to the left thigh, and a disc lesion was suspected. A radiograph taken in 1954 (Fig. 1) showed a defect in the body of the third lumbar vertebra and a diagnosis of crush fracture was made, which was later altered to Kummell's disease, and later still to tuberculous caries. Treatment for two years by immobilisation, streptomycin and isoniazid brought no relief of pain, and a tumour was suspected.

In 1956, at operation in the Irwin Hospital, New Delhi, the third lumbar vertebra was approached by the extraperitoneal lumbar sympathectomy route.

The bulging left psoas muscle was incised and numerous hydatid cysts were encountered and removed. The body of the vertebra was exposed and "scraped." The wound was drained. After operation the pain was relieved. Six months later a similar pain began on the right side and at the same level. A radiograph in 1957 (Fig. 2) showed that the spine was beginning to collapse and that there was formation of new bone anteriorly. In the same year a similar operation to that on the left side was done on the right side. Subsequently sinuses persisted on both sides from which discharged small cysts and pus. *Staphylococcus albus* was cultured from the pus. Two courses of radiotherapy were given to the posterior aspect of the affected vertebra, the second being early in 1958. No apparent benefit accrued.

He was referred to me by Dr R. Joshi, of the Irwin Hospital, New Delhi, in 1958 and was admitted to the Glenelg Base Hospital. His general condition was good. The lumbar curve was flattened and in each loin there was a sinus discharging pus and small daughter cysts. Many implanted cysts could be felt in the abdominal wall at the scars of the previous operations. The Casoni test produced a wheal eight centimetres by eight centimetres in thirty minutes. A blood examination showed that his haemoglobin content was 14.3 grammes (95 per cent) and that his white count was 13,300 per cubic millimetre. *Staphylococcus aureus* coagulase positive, resistant to sulphonamides and penicillin, was cultured from the pus. Malarial attacks occurred unless suppressants were exhibited.

Radiography of the lumbar spine showed that the body of the third lumbar vertebra had almost completely disappeared and that there was an apparently solid bridge of bone anteriorly from the body of second to the body of the fourth lumbar vertebra; this, in fact, was only a thin shell which had become detached from second lumbar vertebra. There was considerable instability of the lumbar spine and the angulation increased markedly in various positions. A sinograph showed an irregular cavity in the third lumbar vertebra with at least three

FIG. 1

FIG. 2

Figure 1—Lumbar spine in 1954 showing primary hydatid disease of the body of the third lumbar vertebra. Figure 2—Lumbar spine in 1957, eleven months after the first operation. Staphylococcal osteomyelitis has been added to hydatid disease of the third lumbar vertebra. There is a shell of new bone anteriorly and slight forward displacement of the spine below the level of the second lumbar vertebra is starting.

FIG. 3

FIG. 4

Figure 3—Lateral radiograph of lumbar spine in 1959 showing the bone graft and much new bone anterior to the second, third and fourth lumbar vertebrae. Most of the body of the third lumbar vertebra has disappeared and there is some parasitic infiltration of the antero-superior aspect of the body of the second lumbar vertebra and of the pedicles of the third lumbar vertebra. Figure 4—Antero-posterior radiograph of lumbar spine at the time of an intravenous pyelograph and left sinograph in 1962. The right transverse process of the second lumbar vertebra and both the transverse processes of the third lumbar vertebra have been removed. A tortuous irregular sinus extends from the left flank to the site of the body of the third lumbar vertebra and downwards into the right psoas muscle. There is bilateral hydronephrosis with the curve formed by the lower calyx and pelvis of the left kidney widened and the kidney is higher than usual. At operation there was a cyst five centimetres in diameter below the kidney with its adventitia including the left ureter.

cloacae communicating with the paraspinal muscles on the left side. The body of the second lumbar vertebra was invaded as, apparently, was the upper part of the body of the fourth lumbar vertebra. The lateral processes and pedicles of the third lumbar vertebra were also involved. Radiographs of the chest showed an unruptured cyst two to three centimetres in diameter in the lingula of the upper lobe of the left lung.

Over the next five years the patient had much treatment and many operations, starting in 1958 with removal of the implanted cysts from the abdominal wall, and, one month later, stabilisation of the lumbar spine by an Albee type of bone graft. Desensitisation to the hydatid antigen by a series of graded intradermal injections was done over the next nine months (Fig. 3). In 1959 the primary bony foci of the parasite, in the body of the third, and

FIG. 5

Diagram of the lateral aspect of the lumbar spine and transverse sections of the second and third lumbar vertebrae to show the position of the bone graft, the extent of bone removed and the site of new bone formation anteriorly.

in part of the body of the second, lumbar vertebrae were removed by curettage and rasping. Later in the same year the hydatid cyst in the lingula of the left lung was removed at thoracotomy.

In 1960 the laminae, transverse processes and pedicles of the third lumbar vertebra and the right transverse process and pedicle of the second lumbar vertebra were removed. At the end of that year some hydronephrosis of the right kidney was found and was attributed to involvement of the right ureter in the wall of the sinus in the anterior part of the right psoas muscle; the ureter was freed at operation.

In 1961 the Casoni test now gave a wheal 5 by 2.5 centimetres; also a further operation was done to remove cancellous bone from the body of the second lumbar vertebra. Later in the same year hydronephrosis of the left kidney was apparent.

Figure 4 shows a combined sinograph and intravenous pyelograph at the beginning of 1962. Five months later the left transverse process and pedicle of the second lumbar vertebra were removed, and after a further three months a hydatid cyst was removed, which was 5 centimetres in diameter and which was adherent to the ureter, pelvis and lower pole of the left kidney. At this site the ureter was included in the adventitia and had been torn by stretching within it which necessitated its repair. In the following year, 1963, both kidneys showed an increase in their hydronephrosis on the retrograde pyelographs, so dilation of the ureteric strictures at the level of the persisting sinus was done on left side. Figure 5 gives a diagrammatic summary of the operations on the spine.

The present condition of the patient is, in general, good; he has been employed as an industrial chemist continuously since early in 1959. He plays tennis, swims, has skin dived to seventeen feet and he drives his own car. There is one persisting sinus in the left flank, from which small cysts emerge from a still undiscovered focus. The loss of the lumbar curve is associated with rigidity of the lumbar spine but the general musculature is good.

DISCUSSION

Vertebral hydatidosis is extremely slow in development. This patient was probably infected in the first decade of life but the parasite did not extend through the periosteum until some ten years later. The onset of pain indicated the formation of cysts in the psoas muscles and the resultant rise of tension caused pressure on the lumbar spinal nerves at that level.

The radiographs taken in 1954 (Fig. 1) gave—typically—no suggestion of hydatid disease. There was no change in the general shape or size of the body of the vertebra, nor was there any change in the intervertebral spaces; there was no bony or periosteal reaction and no calcification in the surrounding area. Hydatid disease of bone gives no pathognomonic radiological signs but, when considered with its extremely slow development and the length of history of the pain, the more common possibilities of metastatic deposits and tuberculous disease may be eliminated. In this patient the lack of response to anti-tuberculous treatment led to the use of incision biopsy and the establishment of the diagnosis.

Drainage is almost inevitably followed by infection and staphylococcal osteomyelitis, which resulted in this patient. This alters both the clinical and the radiological picture. The radiographs of 1957 (Fig. 2) showed almost complete disappearance of the body and pedicles of the third lumbar vertebra, the formation of new bone under the anterior common ligament and an anterior shift of the lumbar spine below the level of the second lumbar vertebra.

It appears that no efficient scolicide was used at the first two operations and many scolices implanted in the operation fields grew into local secondary cysts. Rupture of these increased the sensitisation of the tissues to hydatid antigen.

TREATMENT

In the past the treatment of vertebral hydatid disease was limited to palliative procedures, removal of extra-osseous extensions, decompression of the spinal cord by laminectomy, and drainage. The pathological process within the bone inevitably progressed, often with persistent fistulae, to paraplegia and death.

The essential step in the cure of the condition is the removal of the primary parasitic foci in the bone, as well as the extra-osseous extensions. Only because of the great advances in anaesthesia, in orthopaedic surgery and in resuscitation has it become possible to consider such an attack on the bony foci in an attempt at complete removal. One of the first cases

so treated was recorded by Del Campo (1950) who excised in two stages the whole of the fifth lumbar vertebra.

The order in which the procedures were done on this patient was determined by the presence of implanted cysts, the severe instability of the lumbar spine and the high degree of sensitisation to hydatid antigen. In the past most case reports show that, after each subsequent operation, a greater degree of shock was encountered than at the previous operation. An important factor in this shock is the anaphylaxis as a result of tissue sensitisation to hydatid antigen. Once our patient had been satisfactorily desensitised no undue shock occurred at subsequent operations.

After the implanted cysts had been removed, and after the lumbar spine had been stabilised both by the bone graft and by the formation of a strong anterior bridge of bone, and after desensitisation had been achieved, the imperative attack on the bony foci was begun. Not only did this mean the removal of the cancellous tissue of the bodies of the second and third lumbar vertebrae, but also of those pedicles, laminae and transverse processes suspected of parasitic infiltration, which, for various reasons, was done in several stages by both the lateral and the posterior routes. That all foci have not yet been discovered and removed is shown by the fact that daughter cysts are still being extruded from the remaining sinus on the left.

The use of a saturated solution of sodium chloride as a scolicide—Perez Fontana in 1959 attributed the greater part of those bad effects after operation to the toxic effect of the formalin solution used as a scolicide and absorbed from the operation field. He, therefore, used glycerin carbolic acid (20 per cent) for ten minutes on the diseased tissue, thereafter washing freely with alcohol (90 per cent) and, beginning four days after operation, instilling a 20 to 30 per cent solution of sodium chloride daily through the drain tubes (Perez Fontana 1961).

In our patient the instillation of saturated solution of sodium chloride was used until the development of a communication between the sinus and the pelvis of the left kidney made it inadvisable to continue. It may be contended that it is unlikely that any scolicide in full strength can reach the extremity of the extremely narrow and tortuous sinuses; or that it is impossible for any scolicide to penetrate the microscopic intra-osseous diverticula typical of hydatid extension within bone. Thus the efficacy of action of scolicides can only be expected to occur when there has been free opening up of the primary bony foci.

The effect of an attenuated pyogenic infection—Dévé in 1948 considered that the surgical procedures for the treatment of vertebral hydatidosis were, at that time, hopeless; he therefore advised systematic drainage of the diseased area because it would result in an attenuated infection which induced a desirable osteogenesis. In our patient such an infection occurred after the first operation; this staphylococcal infection may have helped in the formation of the anterior bridge of bone, but it does not appear to have had any effect on the formation of daughter cysts and, by inference, on the survival of hydatid germinal elements.

It appears possible to us that drainage of hydatid foci may assist in averting the invasion of the spinal canal by providing a path of least resistance and easier exit for hydatid elements—acting, in fact, as a “safety valve.”

Involvement of both the ureters in the sclerosing areas on the anterior aspect of the psoas muscles at the level of the sinuses was a severe complication in our patient. So far these strictures have yielded to dilation with ureteral sounds. No attempt has been made to bypass the left ureteric stricture by interposing a segment of ileum, because of the risk of seeding hydatid disease into the peritoneal cavity.

The prognosis of vertebral hydatidosis is usually considered to be as hopeless as that of a malignant neoplasm. All that can be claimed in this patient is that the parasite has been controlled if not exterminated. With a stabilised lumbar spine he has earned his own living and enjoys an active life, but still has to “live with his disease.” Whether the site of the remaining active focus can be discovered and eradicated remains doubtful.

SUMMARY

1. A thirty-two-year-old man has been under treatment for twelve years for primary hydatid disease of the body of the third lumbar vertebra.
2. The second lumbar vertebra became involved, as were the soft tissues over a wide area, but the spinal canal was not affected.
3. A hydatid cyst was found in the left lung.
4. Management included stabilisation of the spine by bone graft, desensitisation to hydatid antigen, removal of the diseased vertebral bodies, pedicles and processes, the use of saturated solution of sodium chloride as a scolicide and the dilation of secondary ureteral strictures.
5. The patient, although not cured, has been enabled to "live with his disease" to earn his living and enjoy tennis and swimming.

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