HYDATID DISEASE OF THE SPINE

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Symptomless hydatid disease may be discovered incidentally by routine radiological examination. Symptoms, if they occur, are usually caused by pressure on adjacent structures, but occasionally a dramatic onset is associated with the infection or spontaneous rupture of a cyst, particularly in the liver or lung. In the relatively rare instances when the disease affects bone, pressure symptoms and pathological fractures predominate. Radiological examination is usually of value and sometimes diagnostic. With modern freedom of travel the disease may be carried to areas where it is not suspected, and the interpretation of radiographs thus acquires particular importance.

The disease is caused by the ingestion of the ova of the Taenia Echinococcus. The definitive host is usually the dog, which becomes infested by eating offal from an intermediary host harbouring hydatid cysts. In most parts of the world, the intermediary host is commonly the sheep. The scolecis within the cysts develop into the mature worms in the intestine of the definitive host. Their ova are then excreted in the faeces. Faecal contamination of food or water may then lead to human infestation. When the ovum enters the intestinal tract of an intermediary host, its envelope is dissolved. The released embryo may migrate through the intestinal wall to enter the portal venous system. The embryos develop into larvae in about three weeks. The larva forms a cyst within which scolecis grow and within which daughter cysts often develop. The host reacts by forming an ectocyst from connective tissue. About 75 per cent of these embryos are trapped in the first filter, provided by the liver, and 15 per cent are caught in the second filter, provided by the lungs (Howorth 1945). The remainder pass these barriers and may settle elsewhere. Cysts developing in soft tissue are spherical and may reach a diameter of several centimetres. Death of the parasite is often followed by calcification of the ectocyst (Jidejian 1953).

BONE INVOLVEMENT

When embryos are deposited in bone, destruction occurs by mechanical pressure without inflammatory reaction. Because of the rigidity of bone tissue, the cyst enlarges unusually slowly, and its shape is often irregular and not spherical as in other tissues. Later, the cortex may be perforated, and relatively exuberant growth then protrudes into the adjacent soft tissues where the usual reaction of ectocyst formation may be observed. Such ectocysts may also, in time, calcify.

Incidence of bone lesion.—Howorth (loc. cit.), in an extensive review of the literature, concluded that about 1,000 cases with bone involvement had been recorded and that these represented an incidence of about 1 per cent. Out of 1,874 cases of hydatid disease in the Hydatid Registry of the Royal Australasian College of Surgeons, 24 per cent involved bone (Woodland 1949).

Site of bone lesions.—The commonest site of bone lesions in hydatid disease is usually considered to be the pelvis. Howorth found this to be involved in about 36 per cent of cases. The spine was involved in 18 per cent and the sacrum in 11 per cent. Bellini (1946), Pugh (1951) and Woodland (loc. cit.), however, believed that the spine is most often affected—in 50 per cent of all cases according to the first author.
Case 1—Three large hydatid cysts are present in the right lung. The right sixth rib is expanded throughout most of its length, with cortical thinning and oval areas of translucency.

Case 1. Figure 2—The cysts in the lung and the rib lesion are again visible, but the spine appears normal. Figure 3—Seven months later, T.4 vertebra shows partial collapse, and non-specific destructive changes have developed in T.6 and 7 vertebrae.
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RADIOLOGICAL CHANGES

The radiological changes in skeletal hydatid disease may be considered in relation to the stage at which they are observed. 

*Early bone lesions* are characterised by ill defined areas of translucency, without sclerotic or periosteal reaction, resembling a bunch of grapes. If observed at this stage, the condition can barely be differentiated from other cystic lesions, such as simple or aneurysmal bone cysts, osteoclastoma, fibrous dysplasia or osseous tuberculous foci. 

*Developed bone lesions* show well defined areas of translucency, which tend to be circular or oval in the long axis of the affected bone. The pattern, however, may be less regular, especially when adjacent daughter cysts are present. The margins are always smooth and show surrounding sclerosis. This sclerosis indicates bone reaction and, although usually attributed to secondary infection, is more likely a simple response to pressure. At this stage the bone may expand, with thinning of the cortex. The cysts having once begun to develop in a bone, with a predilection for a highly vascularised area of spongiosa, tend eventually to spread throughout its length. The stage is now set for a further development, namely rupture of the hydatid tissue through the cortex and the periosteum. 

*Advanced bone lesions* are thus accompanied by a break in the cortex and the development of associated soft-tissue shadows with smooth and rounded margins. Such lesions may attain considerable size and have been described by Bellini (*loc. cit.*) as “ossifluent abscesses.”

**Lesions in the spine**—These deserve special consideration. In the early stages a cystic area may arouse suspicion, in addition to the conditions mentioned above, of a primary or secondary neoplasm, including vertebral angioma. In the late stages, which develop slowly, the areas of translucency in the affected vertebrae are partly masked by reactive sclerosis. “Ossifluent abscesses” may be visible as soft-tissue shadows, and the heads of the adjacent ribs may be involved. In oblique views the laminae may be seen to be affected. This has been stated to tend to exclude tuberculosis. Much more important in this differentiation is that the intervertebral spaces in hydatid disease remain intact and are not narrowed unless secondary infection has developed. That would hardly be expected if such extensive bone involvement were tuberculous. 

A more difficult differentiation is to be made from a plasmacytoma, in which soft-tissue masses, formed by growth of the tumour through the cortex, may present a very similar appearance. Moreover, this tumour does not provoke a marginal sclerotic reaction. Rounded shadows caused by hydatid cysts in the lungs may dispel doubt, practically excluding myeloma (Hodges, Phemister and Brunschwig 1941). Another differentiating factor is probably the rib involvement, although even in this case the transition to multiple myelomatosis may be suspected.

**CLINICAL MATERIAL**

This paper reports three cases of spinal involvement, which are the only examples of skeletal hydatid disease encountered at the American University Hospital and the Orient Hospital, Beirut, during two years from October 1954. In each of these cases cord pressure symptoms were prominent. Attention has recently been called to this feature by Mills (1956). The total number of cases of hydatid disease of all types presenting during this period could not be determined accurately because operation in some cases was refused, but is estimated at forty-five. These figures should not be taken to indicate a high incidence of bone lesions, because Jidejian (*loc. cit.*) reported a series of 190 cases from the American University Hospital without skeletal involvement.

**CASE REPORTS**

**Case I**—A man aged thirty-six was admitted in March 1946 with back pain of three years’ duration, radiating from just below the angle of the right scapula to the axilla and nipple. Tenderness was found over the right sixth rib, and radiological changes affecting the rib were ascribed to plasmacytoma. Five months later, the patient was readmitted with partial
Case 2—Bilateral paravertebral shadows with smooth, rounded margins at T.9 level. Expansion and destruction of head of right ninth rib. Thinning of right pedicle of T.9.

Fig. 4
Case 2. Figure 5—Antero-posterior tomograph (64 centimetres) showing the paravertebral shadows. Figure 6—Antero-posterior tomograph (4 centimetres) showing expansion and destruction of the right ninth rib.
paraplegia. Myelography revealed an incomplete block at the level of T.12 vertebra, becoming complete at T.9. On decompression, hydatid cysts were found in large numbers. Improvement followed, but recurrences necessitated further operations. In November 1955 six hydatid cysts were removed from the fourth thoracic vertebra, with some relief. In June 1957 further cysts were removed from the first thoracic vertebra and the posterior mediastinum. After this operation little function returned to the lower extremities, and later the paralysis became complete below this level.

Review of the radiological findings—Radiological examination of the chest in October 1947 (Figs. 1 and 2) revealed no spinal abnormality, but three large round opacities in the right lung represented hydatid cysts. In addition, the right sixth rib was expanded, with thinning of the cortex by oval and circular areas of translucency. This process involved the whole length of the rib except its anterior end. A further lateral radiograph of the chest, taken after laminectomy and removal of the hydatid cysts in the lung, showed destructive changes in the bodies of T.6 and T.7 vertebrae, and partial collapse of T.4. The disc spaces were well preserved and it seemed likely that these destructive changes represented early manifestations of hydatid disease. The appearance, as indicated above, was not diagnostic at this stage.

Operation was followed by frank osteomyelitis with the development of much deformity and sclerosis, which masked the changes of hydatid disease in subsequent radiographs of the spine. Comment—This case illustrates early involvement of the thoracic spine. Incidentally, advanced changes were present in the right sixth rib.

Case 2—A man aged forty-five, admitted in December 1955, had complained of back pain with progressive numbness and weakness of both lower extremities for one and a half years, and inability to walk for three months. At the same time he noticed kyphosis developing in the thoracic region. For one month before admission he had suffered difficulty in urination

**FIG. 7**

Case 2—Partial collapse of T.9 vertebra, with some irregular sclerosis within the body.
The disc spaces are well preserved.
and had become constipated. Examination revealed kyphosis with a bulge of the thoracic vertebrae between T.6 and T.10, without tenderness. Chest examination was negative. The liver was not enlarged and no masses were palpable in the abdomen. Spastic paraplegia, with increased reflexes, was demonstrated. Pain and touch sensations were everywhere intact. Vibration sense as well as the sense of position were equivocal in the lower extremities. The clinical diagnosis was that of a compressive lesion of the spinal cord of undetermined nature. The Casoni test was negative and eosinophilia was not found.

**Radiological examination**—In the antero-posterior view of the thoracic spine (Fig. 4) para-vertebral shadows with smooth rounded margins were evident at the level of T.9 vertebra, that on the right being larger than that on the left. They were more clearly shown in the 64-centimetre tomograph (Fig. 5), and the 4-centimetre tomograph (Fig. 6) demonstrated destructive changes in the head of the right ninth rib. These had expanded the rib and eroded the cortex. The appearance was very like that in Case 1. The right pedicle of T.9 vertebra had also been eroded. The lateral view (Fig. 7) confirmed that the body of T.9 vertebra was partly collapsed, and some irregular density was visible within it. The disc spaces were well preserved. Myelography revealed an incomplete block at the level of T.12.

After laminectomy the patient made an uneventful recovery, with complete relief of the paresis and disappearance of sensory changes. The reflexes in the lower extremities were still slightly exaggerated. He remained well until last seen in April 1958. Pathological examination of material from the ninth thoracic vertebra and the right ninth rib confirmed the diagnosis of hydatid disease.

**Case 3**—A woman aged thirty was admitted in June 1956 complaining of recurrent mid-thoracic pain since January 1948, which radiated into her shoulders. Six months before admission she suffered a severe bout of pain in the mid-thoracic region which radiated into both legs. Four months later she noticed difficulty in walking and rapidly became bedridden. On admission she was unable to pass urine and was severely constipated. Paraplegia in flexion, with a complete sensory level at T.5 vertebra, was demonstrated. Clinical examination of the chest and abdomen revealed no abnormality.

**Radiological examination** of the thoracic spine revealed widely destructive changes in the fourth, fifth and sixth thoracic vertebrae. In the antero-posterior view (Fig. 8) marked erosion of the left sides of these vertebral bodies was evident. The corresponding left pedicles had been destroyed. The posterior part of the left fifth and sixth ribs showed gross destruction and adjacent oval cavities; these indicated intrinsic bone disease in addition to any changes of pressure erosion that might be present. A large circular soft-tissue opacity lay in the left chest, posteriorly and in contact with the bone lesions. The intervertebral disc spaces were intact. In the lateral radiograph (Fig. 9) central areas of destruction were seen within the affected vertebrae. The destruction of the pedicles and the presence of the soft-tissue mass were again demonstrated. This case illustrates the very advanced stage of the disease.

**At operation** in June 1956 the fifth and sixth left ribs were found to be eroded along a length of about ten centimetres from their necks. Just under the intercostal muscles and in the left costo-vertebral angle a large degenerate hydatid cyst filled with a great number of daughter cysts, was accidentally opened. The area was thoroughly cleaned. During this procedure small daughter cysts were seen to extrude through small holes in the lateral aspect of the body of T.5 vertebra. The dura was seen from its lateral aspect because the pedicle had been completely eroded. No significant improvement followed the operation, and, after discharge from hospital, the patient failed to return for follow-up examination.

**DISCUSSION**

The radiological findings in this disease can be mistaken for a number of other conditions, most important among them malignant disease of bone. In Case 1 the diagnosis was known
Case 3—The pedicles and the left sides of the bodies of T.4, T.5 and T.6 vertebrae have been eroded, together with the posterior parts of the left fifth and sixth ribs. In association with these changes, a large circular soft-tissue mass is present in the left hemithorax. The disc spaces are intact.

Case 3—Central areas of destruction are visible in affected bodies. The erosion of the pedicles and the soft-tissue mass is again shown.
when we first saw the patient. In Case 2 one of us favoured hydatid disease; the other a plasmacytoma progressing to multiple myelomatos is. He was influenced by the presence of myelographic blockage well below the site of the bone lesion, suggesting the presence of tumour tissue within the neural canal. This was, in fact, caused by the presence of numerous hydatid cysts. In Case 3 hydatid disease and neurofibroma were considered possible, but we both favoured the former. In the advanced stage it is less difficult to make a correct diagnosis, but, in two of these cases, malignant disease was seriously considered. The greatest difficulty arises in early disease. Nevertheless it is important that a correct pre-operative diagnosis be made because treatment depends upon it, and ill advised biopsy may rupture the cysts, with consequent recurrences. The differential diagnosis will rest on positive Casoni or positive Weinberg tests, but these are often falsely negative. Eosinophilia may give an indication of the disease, but is a common finding in intestinal infestations. A chest radiograph showing rounded globular shadows in the lung, together with involvement of the vertebrae, may suggest multiple metastases, and the patient might be left to become paralysed on the assumption that the condition was inoperable. In metastatic carcinoma adjacent vertebral bodies are not usually affected, and costal lesions affect only part of a rib, which is not often expanded. The presence of soft-tissue shadows besides a destructive vertebral lesion, with intact disc spaces, should bring this condition into the differential diagnosis.

**SUMMARY**

1. The radiological features of skeletal hydatid disease are discussed. Osseous lesions occur in about 1 to 2 per cent of cases, bone being involved only after the embryos have passed the filters provided firstly by the liver and secondly by the lungs. At first, ill defined areas of translucency appear which are not diagnostic. In developed lesions, clear-cut destructive areas, with a surrounding sclerotic reaction, become visible. The cysts thin and expand the cortex and tend to spread throughout an affected bone. In advanced stages the cortex is ruptured, and exuberant hydatid cyst growth takes place in the adjacent soft tissue. Around this an ectocyst forms, which may later calcify, indicating death of the parasite. The progress of the disease is very slow.

2. Three cases of affection of the thoracic spine are described, and the differential diagnosis is considered, particularly from plasmacytoma and neurofibroma. Each case presented with cord pressure symptoms. Operative decompression relieved these totally in one case, incompletely in another, and not at all in the third and most advanced case.

3. With rapid and easy travel in the modern world hydatid disease is liable to be seen in areas where it is not endemic.

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**REFERENCES**


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