Heterotopic new bone formation in the cervical spine following head injury

Case report

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Heterotopic new bone formation, associated with head injuries, usually affects the shoulder, elbow, and hip joints. The authors present a case with an unusual involvement of the cervical spine, confirmed by bone scanning and computerized tomography. Although the etiology of heterotopic new bone formation is not well understood, factors such as minor local trauma and increased release of growth hormone are considered as possible contributors. The role of drugs and surgery is discussed.

KEY WORDS • new bone formation • cervical spine • head injury • heterotopia

HETEROTOPIC new bone formation (HNF), first described by Dejerine and Ceiler, has attracted the attention of many clinicians over the last 60 years, and numerous reports have been published describing HNF in a variety of clinical conditions. Usually, large joints are affected, especially the shoulder, hip, elbow, and (rarely) the knee joints. Small joints are not involved. We are describing a case with an unusual HNF of the cervical spine, a phenomenon not previously described.

Case Report

This 24-year-old man was admitted on December 10, 1981, after a fall from a considerable height. He was unconscious and decerebrate, requiring ventilation because of respiratory disturbances.

Examination. X-ray films of the skeleton, including the cervical spine, showed no abnormalities, but a right pneumothorax was noted on chest films. Computerized tomography (CT) of the brain showed a small hemorrhage next to the fourth ventricle, edema of the left hemisphere with a slight shift of the midline to the right, and a small intracerebral hematoma in the right frontoparietal region. Other radiological findings included right intrarenal and retroperitoneal hematomas. The patient had pulmonary complications, with recurrent pneumonia on the right side, and a lung abscess that had to be drained surgically.

The patient was transferred to the Loewenstein Hospital on March 1, 1982. At that stage, his eyes were open but he was unable to follow objects and was unresponsive. About 4 months after injury he started swallowing and became gradually more alert. His recovery thereafter was slow. Two and a half years after his fall, he was fully conscious and well oriented in time and space, but showed brain-stem dysfunction, including nystagmus, bilateral ataxia, and right hemiparesis. Surprisingly, after being in prolonged coma, his cognitive functions were quite well preserved. He had high levels of serum alkaline-phosphatase during the follow-up period, which lasted over 3 years. No other abnormal biochemical or hormonal findings were noted.

Radiological Findings of HNF

Skeletal survey, performed 3 months after the injury, revealed HNF at the right shoulder and around the hip joints. A lateral x-ray film of the cervical spine showed an ill-defined mass running obliquely from just below the spinous process of C-2. X-ray films were repeated every 3 months. On later radiographs, the HNF's became more organized and consolidated, as shown in Figs. 1 and 2. A bridge of bone had become established between the greater trochanter of the right femur and the pelvis, leaving the hip joint at a fixed flexion contracture of -30°. A technetium bone scan, obtained 2 years after injury, revealed increased activity...
around the right hip joint and in the cervical spine. A CT scan of the cervical region taken about the same time showed HNBF at various levels of the cervical spine (Fig. 3). Density measurements of the mass were similar to those of the adjacent vertebrae.

A surgical attempt to release the contracture around the right hip joint, performed in March, 1984, was only partially successful.

Discussion

Heterotopic new bone formation (HNBF) accompanies a variety of insults to the nervous system, of which trauma, either to the brain or to the spinal cord, is the most common. The etiology is not understood, although factors like minor trauma, such as induced by external manipulation of joints during physiotherapy, have been considered. Mendelson, et al., examined 500 hemiplegic patients and found HNBF in only six. Analysis revealed that all six either underwent intracranial operation, had evidence of intracerebral hemorrhage, or were in coma for various periods of time. None of the hemiplegic patients who did not lose consciousness developed HNBF, even though some were receiving intensive physiotherapy for subluxation of the shoulder joint. Mendelson, et al., concluded that minor trauma most probably does not play a role in the etiology. A close association with prolonged coma and brain-stem lesions was also described in patients with

Fig. 1. Plain films 2 years posttrauma showing consolidated heterotopic new bone formation of the right axilla (above) and at the right hip joint, forming a fixed bridge of bone (left).

Fig. 2. Plain films, anteroposterior (left) and lateral (right) views, 20 months posttrauma showing consolidated heterotopic new bone formation running from just below the spinous process of C-2 toward the lateral process of C-5.

Fig. 3. Computerized tomography scans of the cervical spine at C-3 (left), C-4 (center), and C-5 (right) 20 months after injury. The soft tissues, vertebrae, and heterotopic new bone formation (HNBF) are shown (upper). There is no continuity of bone between the HNBF and the adjacent vertebrae (lower).
Cervical heterotopic new bone formation

traumatic brain damage: the longer the coma the higher the number of joints involved.\(^4\)\(^5\)

The incidence of HNBF varies greatly among series. Sazbon, \textit{et al.},\(^8\) described an incidence higher than 75\% in a series of patients with prolonged coma; but other authors have mentioned incidences between 10\% and 20\%. These differences relate to the fact that the populations studied in these series are different and the real incidence is probably less than 20\%.

Abnormal bone formation has been described in genetic disorders like myositis ossificans progressiva, but in this condition, like in posthemorrhagic myositis ossificans, primarily the muscles are involved. The association between HLA B27 and ankylosing spondylitis led Weiss, \textit{et al.},\(^12\) to study the possibility of similar genetic predisposition, but none of the 20 patients examined for histocompatibility antigens was B27-positive. Sazbon, \textit{et al.},\(^9\) have shown that the levels of growth hormone found after administration of L-dopa in patients with prolonged traumatic coma were significantly higher in the patients who developed HNBF. Histological studies of specimens obtained during surgical resections of ankylosing HNBF were of no help as the tissue resembled normal bone. The only significant changes found in the serum of these patients are prolongation of increased levels of alkaline phosphatase. However, Kewalramani\(^3\) summarized the literature concerned with the biochemical changes associated with HNBF by stating that no laboratory tests are diagnostic and even alkaline phosphatase levels can be within normal limits in these patients.

The present patient had HNBF involvement of the shoulder and hip joints and an unusual involvement of the cervical spine. The appearance of the cervical lesion paralleled the findings in the other joints. Technetium bone scans showed increased activity in the cervical spine, similar to that found around the other involved joints. A CT scan of the cervical spine clearly delineated the bone mass, the density of which equaled the density of the adjacent vertebrae. Histological study of tissue obtained during surgery showed organized bone.

No definite medical or surgical therapy is available, and recurrence has been reported even after successful surgery.\(^7\) Studies of the relationships of collagen, alkaline phosphatase, and pyrophosphate\(^1\) have led to the use in this disorder of ethane-1-hydroxy-1,1-diphosphonic acid (EHDP) by Stover, \textit{et al.}\(^10\) Although early reports showed favorable results, the more recent ones have been less optimistic.\(^3\)\(^11\) Further research into the nature of HNBF is needed in order to identify factors causing this disabling condition.

References


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