CASE REPORTS

PRIMARY HYPERPARATHYROIDISM PRESENTING AS SPINAL CORD COMPRESSION: REPORT OF A CASE

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A report of a case of primary hyperparathyroidism presenting as spinal cord compression due to a brown tumour of the vertebra is presented. Coexisting chondrocalcinosis illustrates one of the several radiological features seen in hyperparathyroidism.

THE clinical presentation of primary hyperparathyroidism is protean. It may be discovered serendipitously when the serum calcium level is measured during the investigation of a patient. It may manifest itself because of symptomatic hypercalcæmia, i.e., by nausea, vomiting, anorexia, and depression. Involvement of the skeleton may result in bone pain, fracture, or symptoms due to brown tumours themselves. These commonly occur in the long bones, ribs, metacarpals, and mandible. True neoplastic osteoclastoma, however, can occur at these sites.

We report here a case of paraplegia due to spinal cord compression by a solitary brown tumour in a patient with unrecognized primary hyperparathyroidism. The radiological and pathological aspects of the condition are emphasized.

CLINICAL RECORD

A 64-year-old woman was referred to the Neurosurgical Unit of the Manchester Royal Infirmary with a twelve-hour history of back pain and inability to move her legs. She had suffered from back pain following a fall twelve months previously. Since that time she had had intermittent attacks of sharp pain, especially on the left, exacerbated by coughing and sneezing. Over this period she had lost one stone in weight.

On the morning of the day of admission she awoke to find that she could not move her legs and was unable to pass urine. On questioning she admitted having had episodes of depression and anorexia and volunteered a history of recent thirst, polyuria, and nocturia.

Physical examination revealed an apathetic and dehydrated patient. Her blood pressure was 170/90 mm Hg. There was a moderate dorsal kyphosis, and the spinous processes in the lower dorsal spine were tender on palpation. There was flaccid paraplegia of both lower limbs, and all sensory modalities were absent below the umbilicus. The bladder was palpable and insensitive. Plain X-ray films of the dorsal spine demonstrated collapse of T10, with a paravertebral soft tissue mass (Figure 1).

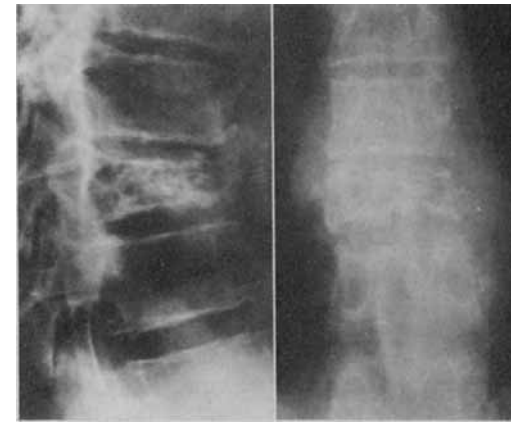

FIGURE 1: Skiagram of dorsal spine, showing collapse of vertebra with a paravertebral soft tissue mass.

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A cisternal myelogram confirmed the presence of a complete block at T10 (Figure 2). The spinal cord and dura were displaced to the right by an extradural space occupying lesion. A provisional diagnosis of carcinomatous metastasis with compression paraplegia was made, and decompression laminectomy was performed.

To the naked eye the tumour had all the typical features of a metastatic deposit. It was granular and friable. The spinal cord was displaced to the right. The extradural tumour was removed until free pulsation was observed and a catheter could be passed both upwards and downwards. The biopsy report suggested an osteoclastoma. Histologically, there were numerous osteoclastic giant cells separated by spheroidal and spindle shaped mononuclear cells. Mitoses were infrequent, and there was little fibrous reaction or osteoid formation (Figure 3).

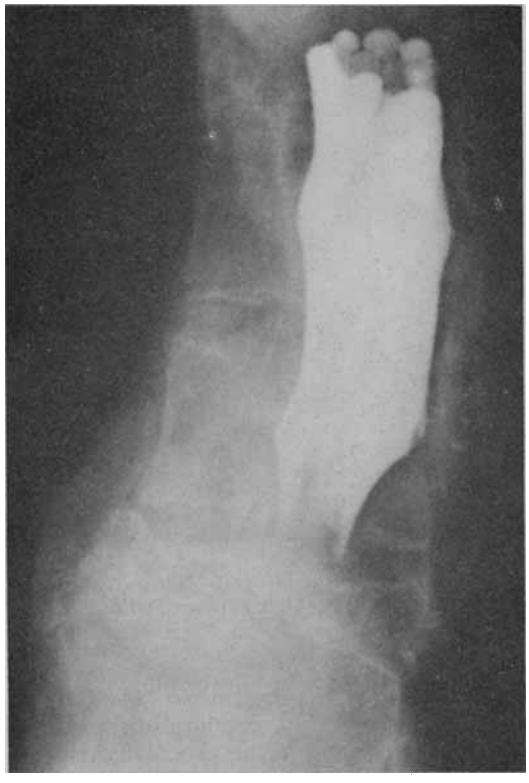

FIGURE 2: Cisternal myelogram showing complete block to flow of Myodil at the level of the collapsed vertebra.

Following receipt of this report, the presence of hypercalcæmia was confirmed by the finding of a serum calcium level of 14 mg/100 ml. The serum level of phosphorus was 4 mg/100 ml and of magnesium 1 mg/100 ml. Surprisingly, the serum alkaline phosphatase level was only 13 KA units/100 ml. The creatinine clearance rate was 16 ml per minute. The parathyroid hormone level was estimated by immunological assay to be greater than 15 ng/ml (normal up to 0·7 ng/ml).

Subsequent radiographs of the hand, shoulder, pelvis and knees demonstrated calcification in the triangular ligaments of the wrist joints, around the shoulders, the fibrocartilage of the symphysis pubis, and the menisci of both knees (Figures 4, 5). These findings of chondrocalcinosis strongly supported the suspicion of primary hyperparathyroidism.

In the first postoperative week the patient became dehydrated and this condition was associated with a serum calcium level of 19 mg/100 ml, and the blood urea was 140 mg/100 ml. Rehydration with normal saline was associated with a fall in the serum calcium level to 14 mg/100 ml. The addition of oral phosphate supplement (Sando P) further reduced the hypercalcæmia.

Neurological recovery was minimal, and a repeat myelogram again demonstrated an extradural lesion in the left anterolateral aspect of the spinal canal. A further operation was performed and the tumour mass was removed.

The third and final operation was performed three weeks later, when a parathyroid adenoma weighing 15 grammes was removed from behind the left lobe of the thyroid gland. Histologically it consisted entirely of clear cells. Normocalcæmia followed the operation, with the parathyroid hormone level falling to normal (0·5 ng/ml). The blood urea fell to around 50 mg/100 ml, and the patient's general wellbeing improved dramatically, with increase in appetite and weight gain. Regrettably, she remained paraplegic.

DISCUSSION

The initial biopsy finding was compatible with the presence of a giant cell tumour or a brown tumour of hyperparathyroidism. The similarities between these two lesions have been well described (Jaffe, 1933; Lichtenstein, 1965; Dahlin, 1967; Willis, 1967). Giant cell tumours have been described in the jaw in association with hyperparathyroidism and these regress after removal of the parathyroid adenoma (Clark and Taylor, 1972). Histologically, such tumours cannot be distinguished from osteoclastomata or tumours of parathyroid origin. The radiological changes of hyperparathyroidism are much more likely to occur in the jaw and vertebræ than in the hands and feet in cases of moderate severity when there is a negative calcium balance (Jaffe, 1972). Brown tumours may occur in the vertebral bodies and weaken them. Vertebral collapse has been described as a presenting symptom in hyperparathyroidism (Pyrah, 1966), but the opportunity for histological examination to confirm the diagnosis is infrequent.

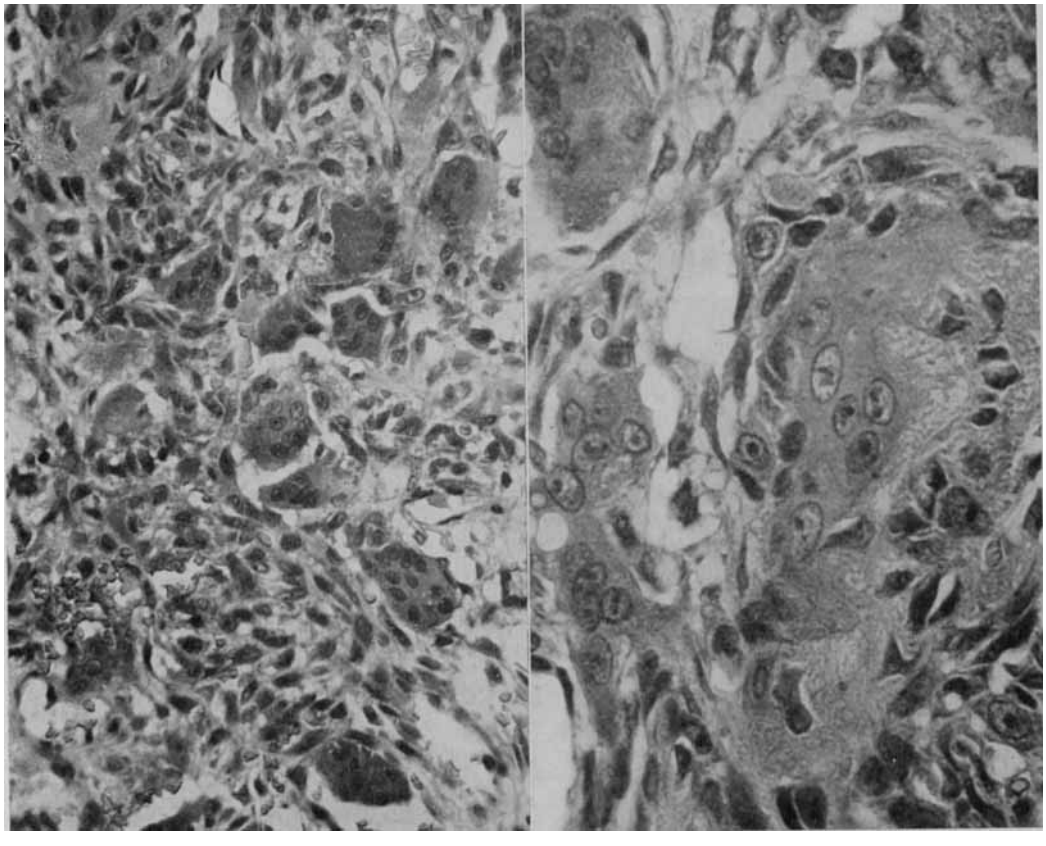

FIGURE 3: (left) photomicrograph showing numerous osteoclastic giant cells separated by spheroidal and spindle shaped mononuclear cells (H. & E., × 100); (right) osteoclastic giant cells (H. & E., × 200).

Normal osteoclasts cannot be distinguished from giant cell osteoclasts by histochemical methods (Schajowicz, 1961), and according to Lichtenstein (1965), the differentiation of giant cell tumours from similar lesions will not be resolved by histochemical means. Ultrastructural examination has been similarly unrewarding (Spjut *et alii*, 1971). Whether newer techniques of examination of isolated osteoclasts will be more helpful remains to be seen (Walker, 1972).

In our patient, the radiological appearance of the collapsed dorsal vertebræ with myelographic evidence of an extradural mass certainly suggested the diagnosis of a metastatic deposit or a myeloma. Evidence suggesting an osteoclastoma makes further radiological investigations in search of other manifestations of hyperparathyroidism mandatory. On this occasion, chondralcalcinosis was clearly demonstrated in the articular cartilages. The more commonly recognized bone changes in hyperparathyroidism such as subperiosteal resorption in the phalanges were absent. Since Ekolf's case report in 1952 radiologists have become increasingly aware of articular cartilage calcification in primary hyperparathyroidism.

Joint cartilage calcification is said to occur in approximately 18% of patients with primary hyperparathyroidism (Dodds and Steinbeck, 1968). The average age of patients showing chondralcalcinosis in primary hyperparathyroidism has been reported to be between 50 and 60 years. It has been suggested that joint cartilage calcification in primary hyperpara-

thyroidism is probably related to hypercalcæmia. Further, it has also been postulated that these patients are in an age group where the cartilage concomitant is altered by age and hence creates a favourable situation for cartilage calcification in the presence of prolonged hypercalcæmia. This theory would account for the much higher incidence of joint cartilage calcification in primary hyperparathyroidism than would be expected on a

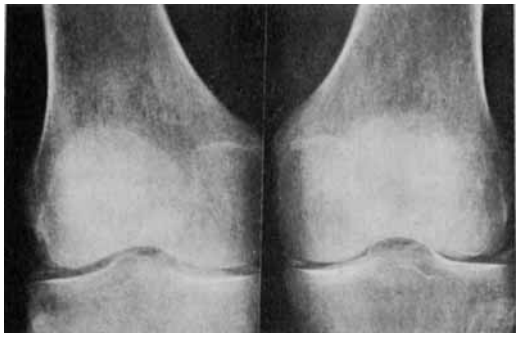

FIGURE 4: Radiograph of knees showing chondrocalcinosis in both menisci.

degenerative basis alone. In some cases, a brown tumour of hyperparathyroidism may be the only skeletal lesion. The great majority of osteoclastomata unrelated to hyperparathyroidism are situated in the long bones, although a brown tumour of hyperparathyroidism may also occur in these sites. It is rare for either condition to present in the thoracic vertebræ.

The combination of chondralcalcinosis and giant cell tumour, albeit in a single cystic lesion, should strongly suggest the possibility of primary hyperparathyroidism. A number of comprehensive surveys (Pyrah, 1966; Hellstrom, 1962) and textbooks provide an authoritative account of hyperparathyroidism, its various manifestations, and their management. As far as a survey of the literature can show, this case report demonstrates an extremely rare presentation. Only one other similar case has been reported (Shaw and Davies, 1968). It is interesting to note that in the present case, the serum alkaline phosphatase level was not raised. It is recognized that the plasma alkaline phosphatase level is raised only in the presence of demonstrable radiological bone disease. The other patient with vertebral brown tumour, described by Shaw, had a high alkaline phosphatase level, but there was obvious subperiosteal resorption, with other manifestations of hyperparathyroid osteodystrophy.

As compression paraplegia is usually dealt with by neurosurgical units, it is pertinent to emphasize the great variety of clinical conditions that can be encountered in patients with spinal cord compression. Metastatic carcinomatous compression must be first on the list of causes. These tumours are usually osteolytic, and the diagnosis is rarely in doubt. When vertebræ are affected, the intervertebral disc is at first spared, and in nearly half of the cases the nature of the tumour may be

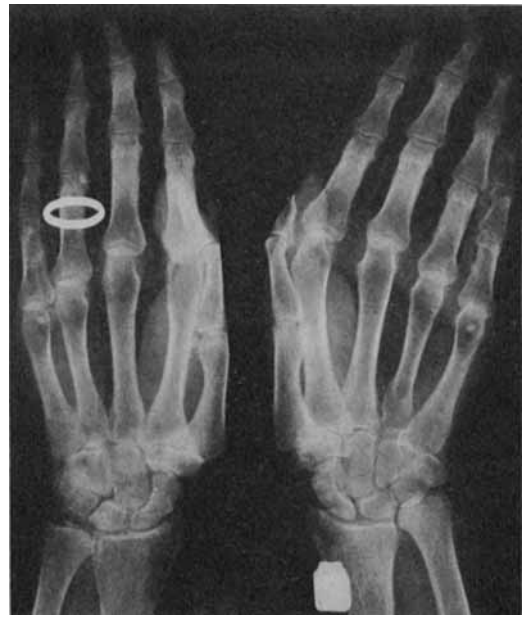

FIGURE 5: Radiograph of wrist showing calcification in the triangular ligament of the left wrist joint.

unknown (Smith, 1965). Operation is undertaken for decompression and to establish or confirm the diagnosis.

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SPINAL EPIDURAL ABSCESS IN PREGNANCY

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A case of spontaneous epidural abscess occuring in a pregnant woman is reported. Some delay in diagnosis occurred. Decompression resulted in prompt recovery and improvement of a mild neurological deficit.

SPINAL epidural abscess is a rare condition which may have disastrous neurological sequelæ (Hulme and Dott, 1954), and a fatal outcome in untreated cases is well recognized (Baker *et alii*, 1975). Early recognition and treatment are required, since if the neurological deficit is profound or longstanding, the prospects of functional recovery are poor. The purpose of this paper is to report a case of spinal epidural abscess secondary to posterior vertebral element osteomyelitis which occurred in a pregnant woman. To our knowledge, this association has not been documented previously, and the report serves to emphasize the diagnostic difficulties in an unusual set of circumstances.

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CLINICAL RECORD

A 27-year-old woman presented to the prenatal clinic at the Royal North Shore Hospital in the 30th week of her second pregnancy. Her first pregnancy nine years previously had been uncomplicated and had resulted in the vaginal delivery of a normal, full-term infant.

The patient gave a three-week history of continuous, severe, lumbosacral back pain, with radiation to the right sacroiliac region, the right groin, and the anteromedial aspect of the right thigh, and later radiating also to the area of the left greater trochanter. She also complained of difficulty in walking, and large doses of proprietary analgesics had provided little relief. Her pain was increased by movement and made worse by sitting and standing. The patient had also experienced rigors.

On direct questioning she admitted to slight urinary incontinence, hesitant micturition, and inability to control flatus. She denied the presence of any pustular skin lesions prior to the onset of her back pain.

Her past history included an admission to hospital following a drug overdose six years previously,