Case report

Neurofibrosarcoma following radiotherapy for ankylosing spondylitis

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Bentley, S. J., Davis, P., and Jayson, M. I. V. (1975). *Annals of the Rheumatic Diseases*, 34, 536-538. Neurofibrosarcoma following radiotherapy for ankylosing spondylitis. Twenty-six years after radiotherapy to the spine for ankylosing spondylitis a patient developed a neurofibrosarcoma initially around the left sacroiliac joint. It is likely that this was a long-term complication of the radiotherapy.

The long-term hazards of radiotherapy are well known so that it is now little used for ankylosing spondylitis. We here report a new complication—a neurofibrosarcoma developing in a patient treated some 26 years previously.

History

In 1946 at the age of 19 the patient developed low back pain and polyarthritis, and was diagnosed as having ankylosing spondylitis. In October 1948 he received 20 radiotherapy treatments with a total of 2000 rads to each of lumbar spine, dorsal spine, cervical spine, and a 15 × 10 cm area over both sacroiliac joints. Further treatments of 1600 rads and 1400 rads were given to the left shoulder and lumbar spine, respectively, in April 1949 and October 1950. Between 1952 and 1973 he suffered occasional flares of back pain which were treated with physiotherapy and phenylbutazone by his general practitioner. In August 1974 he presented with increasing low back pain of 9 months' duration. He appeared ill with obvious weight loss, but the general examination and the peripheral joints were normal. The dorsal and lumbar spines were rigid and there was marked tenderness of the left sacroiliac joint extending across the midline. Both ankle jerks were absent.

X-ray of the pelvis showed an extensive osteolytic lesion around the left sacroiliac joint eroding the sacrum and the ilium. There was also a similar but much less marked change in the right side of the sacrum (Fig. 1). Open biopsy showed an extensive pale brown gelatinous avascular tumour. Microscopy showed this to be a sarcoma with round, spindle, and giant cells, a dense reticulin pattern, marked degeneration, and no mitotic

figures (Fig. 2). The appearances were those of a neurofibrosarcoma.

The patient was treated with palliative radiotherapy and analgesics. He failed to respond and died with ureteric obstruction and renal failure in October 1974.

Discussion

In the past decade radiotherapy has become less acceptable as treatment for ankylosing spondylitis. It provides only temporary relief to locally inflamed areas of the spine and does not arrest the progress of the disease. In any event radiotherapy appears inferior in analgesic action to phenylbutazone (Mason, 1964).

More important, however, are the reports of the long-term complications. Pulmonary fibrosis and transverse myelitis may occur, but more serious is the increased incidence of local and systemic malignancies (Court-Brown and Doll, 1965) and particularly leukaemia (Court-Brown and Doll, 1957). Tumours at sites within the beam of radiation are usually carcinomas of the skin or viscera. Sarcomas of bone and soft tissues are rare. Doll (personal communication, 1975) has knowledge of only 5 bone sarcomas, and 7 fibrosarcomas; and Edgar and Robinson (1973) have reported five patients with fibrosarcomas. Our case fulfils the four criteria of Cahan, Woodward, Higinbotham, Stewart, and Coley (1948) for a post-irradiation tumour, and the coincidence of sites of radiotherapy and tumour make it likely that the neurofibrosarcoma is a direct complication of the treatment.

FIG. 1 *X-ray showing extensive destructive changes markedly involving the left sacroiliac joint, but also the right side of the sacrum*

FIG. 2 *Biopsy of lesion*

References

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