Case Report (I)

SPINAL OSTEOSARCOMA: REPORT OF 2 CASES

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ABSTRACT

Primary osteosarcoma arising from the vertebral column is very rare. We report here two such patients. It tends to occur in slightly older age group with median age of 38 years, than Osteosarcoma of the extremity. Patient usually presents with pain and neurological deficit. It could be secondary to Paget's disease or to irradiation received for a prior cancer. The differential diagnosis from benign tumour like osteoblastoma must be made and is discussed. The prognosis is generally poor.

INTRODUCTION

Primary Osteogenic Sarcoma (OS) arising from vertebral column is rare and its reported incidence ranges from 0.85 to 2.0%. [1,2,3] we report here two such cases. Pertinent literature is being reviewed.

Case 1: Twenty-seven years old female, in July 1996, presented with backache and tingling–numbness of both legs followed by acute paraplegia. Imaging suggested non-expansile blastic lesion and destruction involving D10 and D11 vertebra (fig.I), Emergency laminectomy and decompression was done, and histopathology suggested low-grade osteosarcoma.

She received postoperative radiotherapy 50Gy in 25 fractions till Aug 1996. She was well for about 6 years. In December 2002, She developed recurrent symptoms and imaging suggested, large, irregular infiltrating mass lesion involving D9, D10 vertebrae, spinal canal, posterior paraspinal and left paravertebral region, which eroded posterior portion of body, pedicle and spinus process and compressed spinal cord anterolaterally on the right side (Fig.2). MR myelogram suggested extra dural block at D9, D10 level. Re-exploration and total removal of the mass was done. Metastatic work-up was negative. She received 4 cycles of combination chemotherapy consisting of cisplatin and doxorubicin between January and March 2003. This was follwed by radiation 60Gys in 30 fractions completed in May 2003. Subsequently, she had recurrent symptoms with signs suggestive of posterior column involvement. Her MRI suggested space-occupying lesion involving D8 and D9 vertebral bodies, left paraspinal muscles and epidural space with cord compression and syrinx formation at D8 level (figure-3). Her work-up for metastatic disease was still negative. She was offered high dose ifosfamide. She received 1st cycle on 25th June 2003. She underwent re-laminectomy in August 2003. At present she is on supportive care.

Case 2: Thirty-Two years old male presented with symptoms of pain in left lower limb, gradually progressing to left lower limb paresis in September 1998. His myelogram suggested extradural cord compression at L_1, L_2, L_3 (fig. 4). Laminectomy and removal of tumour relieved him of monoparesis. Histopathology was pleomorphic osteosarcoma, high grade. He received 6 cycle of combination chemotherapy consisting of cisplatin and doxorubicin during period from September 1998 to June 1999. He is under surveillance since then and regular follow-up, free of disease (Fig. 5).

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DISCUSSION

Primary osteosarcoma of the spine accounts for 1.2% of all cases of osteosarcoma.[1,2,3,4] As reported in Co-operative Osteosarcoma study (COSS) Group, both patients had pain and neurological deficit as a presenting complaint. Both patients were young adult. One patient was male and another female. The spine is commonly affected by uncomplicated Paget's disease.[6,7] Neither of the patient had preceding history of irradiation or Paget's disease of bone. In the COSS study, one out of twenty two patients had history of mediastinal irradiation for Hodgkin's disease, 18 years prior to development of thoracic spinal osteosarcoma.[5] In the another study by Kenneth et al [3], three out of 10 patients had Paget's disease, all being women over 50 years of age. Both of our patients had non-metastatic disease. In COSS study showing six out of 22 patients presented with metastatic disease on initial presentation. One patient had thoracic and the other patient had involvement of lumbar spine. Parida et al[8] reported a case of spinal Osteosarcoma arising from the spinous process of the third cervical vertebra in an 18 year old girl.

On histopathological evaluation, one patient had low-grade osteosarcoma and the other had pleomorphic high-grade osteosarcoma. Case 1 has local recurrence, after 6 years of disease free period. There was a change of grading from low grade to high-grade histopathology and therefore, change of behavior of the disease, as suggested by progressive disease, not responding to chemotherapy or radiotherapy. However, this phenomenon is not documented in literature for osteosarcoma of spine.

One patient case-1 received posto-perative radiotherapy. Radiation therapy has not played a relevant role in the management of the osteosarcoma because of the natural history of the disease.[9] However; it has been used as either preoperative, intra operative or postoperative treatment and for palliation. Weichselbaum and Cassadys[10] recommended definitive irradiation only for those with surgically unresectable lesions with the dosage of 66-70Gy. No standard guidelines have been established for the RT for spinal OS. In a series by Kenneth, 9 of the 10 patients received radiotherapy. In the COSS study,[5] 8 of 22 patients received RT. One patient received it as a postoperative adjuvant treatment, five patients as first line local therapy and one patient received high activity Samarium-153-ethylene diamine tetramethy-lene phosphonate (EDTMP; 150 mega becquerels per kg of body weight) with peripheral blood stem cell rescue in addition to 60 Gy irradiation to pulmonary metastasis. There was a tendency for an improved overall survival rate in the patients who received irradiation. Shives[11] et al and Sunderasan[12] et al report similar phenomenon. As reported in the COSS study targeted radiotherapy with Sm-153-EDTMP may offer an additional treatment option for some patients with inoperable osteosarcomas.

Spinal Osteosarcoma must be differentiated from benign conditions like osteoblastoma, aneurysmal bone cyst and giant cell tumour of the bone.[13, 14,15] The commonest benign condition is osteoblastoma. The clinical distinction may be obscure. Radiologically, benign osteoblastoma causes expansion of posterior elements, i.e. a pedicle, lamina or spinous process, and usually has a well-defined margins. In this study, the absence of expansile properties and involvement of vertebral body were characteristic of Osteosarcoma. Histopathologically, osteoblastoma may contain area of high cellularity with foci of bizarre nuclei. These foci contain no mitotic activity. Osteosarcoma has bizarre nuclear configuration and peripheral permeation of normal bone, atypical mitotic figures and malignant cartilage. Giant cell tumour is uncommon at this site and

can be differentiated by even dispersal of giant cells in the vascular stroma composed of plump and apparently benign giant cell. Aneurysmal bone cyst is an expansile lesion, not uncommon in vertebrae. Its microscopic pattern is usually quite characteristic, consisting of large and small blood filled spaces typically lined by fibrous or granulation tissue. The stroma often contains hemosiderin, histiocytes and extravasated blood. Osteoid production is often present.

All above conditions were ruled out histologically.

CONCLUSION

Osteosarcoma of spine is a rare entity with overall poor outcome. The newer techniques and advances applicable to the extremity Osteosarcoma must be applied to this poor subset to improve the outcome.

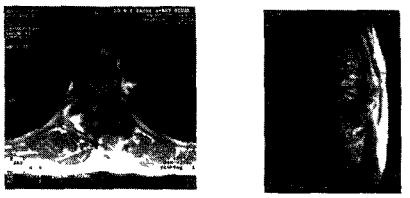

Fig.-I: MRI Dorsal Spine: (25/5/96): Hypointense mass in D10-11 level, posterior aspect of spinal canal pressing spinal cord anteriorly.

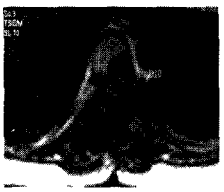

Fig.-II: MRI Dorsal Spine (9/12/02): Irregular, intensely enhancing infiltrating mass involving D9, 10 bodies, spinal canal, posterior paraspinal and left paravertebral region, compressing spinal cord anterolaterally on right side.

Fig.-III: MRI Dorsal Spine (17/6/03): S O L involving D8-9, vertebral bodies, posterior elements, left Para spinal mussels and epidural space with cord compression and localized syrinx formation within cord at D8 level.

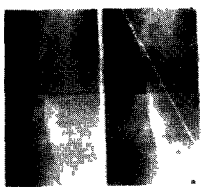

Fig.-IV: Myelogram showing extradural cord compression at L_1, L_2, L_3

Fig.-V: MRI of Lumbar Spine (3/2/04): Showing no evidence of disease

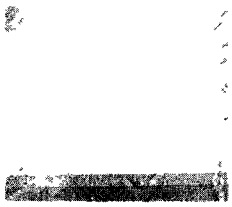

Fig.-VI : Section shows tumour cell osteiod formation. (H & E. 10X)

Fig. VII : Tumour cell with pleomorphic nuclei & mitosis. (H & E, 40X)

REFERENCES:

1. *Dahlin DC, Unni K K. Osteosarcoma of bone and its recognizable varieties. Am J Surg Pathol 1977; 1:61-72.*
2. *Goidanich IF, Battaglia L, Lenzi L, Silva E. Osteosarcoma: analysis of factors influencing prognosis in 100 cases. Clin Orthop 1966;48:209-222.*
3. *Kenneth W Bareviek, Andrew G Huvos, Julies Smith. Primary Osteogenic sarcoma of the vertebral column: A clinicopathological correlation of ten patients. Cancer 1980;46:595-604.*
4. *Unni K K. Dahlin's bone tumours general aspects and data on 11,087 cases, 5th ed. Philadelphia: Lippincott – Reven 1996.*
5. *Toshifumi Oraki, Silke Flege, Cilf Ciljenquist. Osteosarcoma of the spine: Experience of the co-operative study group. Cancer 2002;94(0):1069-1077.*
6. *Dorfman H D. Malignant transformation of benign bone lesion. Proc Nat Cancer Conf 1973;7:901-913.*
7. *Mc Kenna R J, Schwinn C P, Soong K V, Higinbotham N L. Osteogenic sarcoma arising in Paget's disease. Cancer 1964;17:42-66.*
8. *Parida DK, Biswal BM, Thulkar S, Rath GK. Osteogenic sarcoma of the third cervical vertebra: a case report with review of literature. Jn. Clin. Radiotherapy and Oncol 1997;12:121-3.*
9. *Carlos A Perez. Principles and practice of Radiation Oncology, 4th Ed. 2174-75. Publ:*
10. *Wichselbaum RR; Cassady JR. Radiation therapy in Osteosarcoma. In Jaffe N. ed. Solid tumours in childhood. Boca Raton, FL: CRC Press 1983:183.*
11. *Shives TC, Dahlin DC, Sim FH. Osteosarcoma of the spine. J Bone Joint Surg Am 1986;68:660-8.*
12. *Sunderasan N, Rosen G, Huvos AG. Combined treatment of Osteosarcoma of spine. Neurosurgery 1988;23:714-9.*
13. *Jaffe H L. Tumours and tumourous conditions of bones and joints Philadelphia: Lea and Febiger 1958.*
14. *Tumour and tumour like lesions of bone. Resnick diagnosis of Bone and Joint Diseases, 4th eds. Vol 4, 3790*
15. *Mc Leod R A, Dahlin D C, Beabout J W. The spectrum of osteoblastoma. Am J of Roentgenlo 1976;126:321-335.*