Original Articles

Primary Bone Tumour of the Spine: Surgical Treatment and Histopathological Analysis

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Abstract

Background: Primary bone tumour of the spine are rare but not uncommon. These include both benign and malignant lesions. Surgical resection and stabilization in some cases are the treatment of choice. Adjuvant radio and/or chemotherapy are needed in malignant cases.

Methods: We retrospectively reviewed all the primary bone tumour of the spine that were surgically treated in the department of Neurosurgery of Dhaka Medical College Hospital and National Institute of Neurosciences and Hospital Dhaka, Bangladesh between 2010 and 2015.

Results: Eighteen cases were included that presented at a median age of 36 years (range 14 to 49 years). Pain was the most common presenting symptom. Three patients had significant neurological deficits at time of presentation and in most cases there was an improvement after surgery. A variety of surgical strategies was employed with the use of adjuvant radio- or chemotherapy in six cases. Six benign and twelve malignant tumours were treated.

Conclusions: Primary bone tumours of the spine are often associated with a significant delay in diagnosis. Surgical strategy should be individualized for each case. Acceptable results can be achieved with early surgery and adequate chemo-radiotherapy.

Key words: primary bone tumour, spine, surgical treatment, histopathology.

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Introduction:

Primary bone tumour of the spine is a heterogynous group of conditions that includes both benign and malignant tumours, with only 6% of these tumours considered malignant. The spine is a frequent site of metastasis, but primary bone tumours of the spine are rare and make up only 4.2% of spinal tumours. Eleven per cent (11%) of primary musculoskeletal tumours occur in the spine with certain tumours having an affinity for the spine. Although rare, these tumours offer a considerable diagnostic dilemma and therapeutic challenge. Timely diagnosis is crucial, as immediate attention could affect overall prognosis.³

The most common primary tumour of the spine is metastatic deposits. A number of both benign and

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malignant tumours may arise primarily from the spine. Among benign tomour osteoid osteoma, osteoblastoma, osteochondroma, giant cell tumour (GCT), aneurysmal bone cyst (ABC), eosinophilic granuloma (EG), haemangioma are common. Among malignant lesion malignant lymphoma, sarcoma, ewing sarcoma, osteosarcoma, chondrosarcoma, malignant fibrous histiocytoma (MFH), fibrosarcoma, chordoma, plasmacytoma/multiple myeloma and peripheral primitive neuroectodermal tumour (pPNET) are common.

Primary bone tumours are more common in the thoracic and lumbosacral regions than in the cervical spine.⁴ These tumours occur according to a typical anatomical distribution within the vertebra. In general, malignant tumours occur more frequently in the anterior elements and benign tumours in the posterior elements.^{3–5}

The aims of treatment of these axial neoplasms include complete resection if feasible, preservation of neurological function and stabilisation of the spine if needed.⁴ The aim of our study was to review the primary bone tumours of the spine treated at our unit regarding the presentation, the surgical strategy employed and the outcome.

Materials and Methods:

This is a retrospective study. All the primary bone tumours of the spine that received surgical treatment

at the department of Neurosurgery of Dhaka Medical College Hospital and National Institute of Neurosciences and Hospital Dhaka, Bangladesh from 2010 to 2015 were included in this study. We excluded all secondary deposition in the spine. We treated 18 cases during this period that were included according to these criteria. A case note and imaging review was done where patient demographics, details of presentation, clinical information and the management strategy were analyzed. All complications and mortalities were recorded.

Results:

Between 2010 and 2015, a total 18 patients were diagnosed as primary bone tumour of the spine. The median age at presentation was 36 years (range 14 to 49 years) with eleven(61.12%) male and seven(38.88%) female patients affected. The median follow-up was 18 months (range 3 to 60 months).

Table-IDistribution of patient by age.

Age group	Frequency	Percentage
0 to 10 years	0	0
11 to 20 years	3	16.67
21 to 30 years	7	38.89
31 to 40 years	4	22.22
41 to 50 years	4	22.22
Total	18	100

Table-IIDistribution of patient by sex.

Sex	Frequency	Percentage
Male	11	61.12
Female	7	38.88
Total	18	100

Table-IIIDistribution of patient by site of involvement.

Site of involvement	Frequency	Percentage
Cervical	1	5.55
Dorsal	7	38.89
Lumber	6	33.34
Sacrum	4	22.22
Total	18	100

The most utilized diagnostic tool was MRI and CT scan of the spine. It helped to diagnose the case as well as planning of surgery. X ray is no longer a standard diagnostic tool of spine tumour, it was used in all cases as c complimentary diagnostic tool. It also helped to find out the extra spinal involvement like rib cases in two cases. Pre-operative CT guided FNAC was done in seven cases. Post-operative histopathological study was done in all cases.

Primary bone tumour of the spine most commonly involved the dorsal (38.89%) and the Lumber (33.34%). It affect the sacral (22.22%) and the cervical spine (5.55%) less frequently. (Table-3)

Table-IVDistribution of patient by histopathological findings.

Туре	Histopathology	Frequency	percentage
Benign	-Osteoid osteoma	1	5.55
	-Haemangioma	1	5.55
	-Giant cell tumour	1	5.55
	-Aneurysmal bone cyst	2	11.11
	-Esonophilic granuloma	1	5.55
Malignant	-Myxoid Chondrosarcoma	a 1	5.55
	-ewing's sarcoma	2	11.11
	-Chondrosarcoma	2	22.22
	-Chordoma	3	33.33
	-Multiple myeloma	2	22.22
	-pNET	2	22.22
Total		18	100

Pain was the most common presenting symptom (15 from 18 cases) and four cases presented with pain alone. Pain was mostly axial pain in the affected region (11 of 18 cases), although three patients presented with chest pain and one with flank pain. Other presenting symptoms included neurological symptoms and odynophagia. One patient presented with a recurrence of a malignant aggressive tumour (Ewing sarcoma of sacral spine) following incomplete resection at another unit. Ten patients had no neurological deficit at time of presentation. Two patients presented with radicular symptoms and another three had significant neurological deficits at time of presentation. One case presented with significant neurological deficit with a diagnosis of benign aggressive lesions (Aneurysmal bone cyst). Two of these cases had significant improvement after surgery. We had no cases of neurological deterioration following surgery. We treated six benign and twelve malignant tumours. The histopatghological diagnoses are summarised in Table 4. In all cases the surgery was performed by the senior author. A

variety of surgical strategies was utilised. An intralesional resection or debulking of the tumour was performed in six cases and a marginal resection was performed in nine cases to correlate with diagnosis. In six patients a fusion was performed by transpedicular screws and rods. Adjuvant therapy was used where indicated. Two of the recurrent cases

were in malignant tumours and one in benign aggressive tumour. In the one recurrent malignant tumour a repeat attempted marginal resection was performed. The other two recurrent cases were not re-operated as resection was not possible with acceptable morbidity. We had three deaths in the series on further follow up.

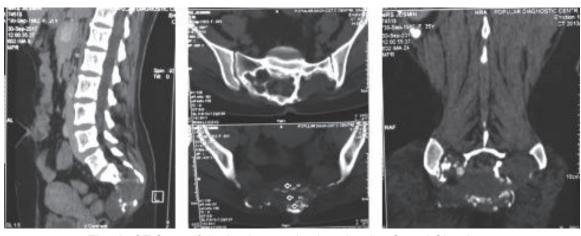


Fig.-1: CT Scan of the lumbosacral spine showing Sacral Chordoma.

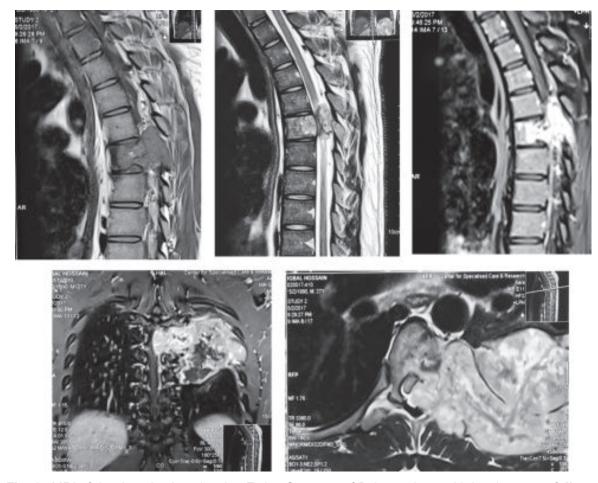


Fig.-2: MRI of the dorsal spine showing Ewing Sarcoma of D-9 vertebrae with involvement of ribs.

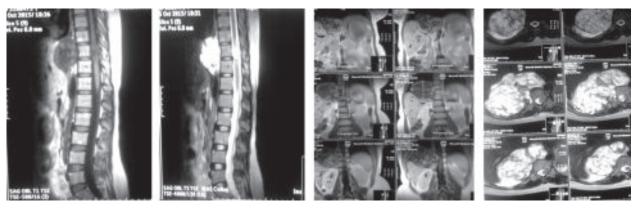


Fig.-3: MRI showing extensive Chondrosarcoma of the dorsal spine.

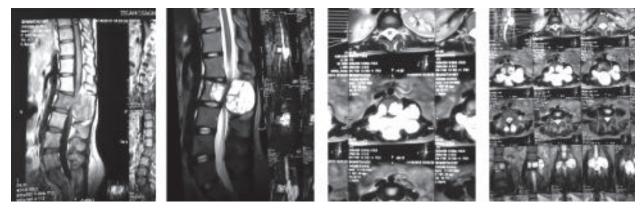


Fig.-4: MRI of the dorsolumber spine showing Aneurysmal Bone Cyst of the L-2 vertebrae.

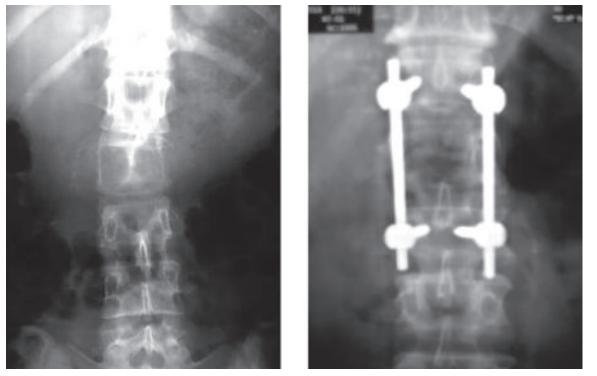


Fig.-5: Pre and post-Operative X-ray of L-2 Aneurysmal Bone Cyst.

Discussion:

The most common presenting complaint of patients with primary bone tumour of the spine is pain, with roughly 60% complaining of axial pain and 25% of radicular symptoms.3 The presence of a neurological deficit carries a worse prognosis. 11 Pain at rest or night pain is red flag symptoms that warrant further investigation. Spinal deformity is obvious when present, but occurs in less than 10% of patients. The most common cause of a painful scoliosis in adolescents is an osteoid osteoma.5 A delay in diagnosis is common and our finding is confirmed in other series. ⁴ This is due to the presenting symptoms often being vague, pain often being the only symptom, and the initial radiographs often only showing subtle or no abnormalities leading to the symptoms not being properly investigated. Most patients report a slow, gradual onset of pain in the involved area. In benign tumours of the spine an average of 26 weeks of symptoms before presentation are reported.4 This was confirmed in our series where the average delay in presentation was 7 months.

A variety of classifications systems is used for primary bone tumours of the spine. The Enneking staging system¹² was initially described for primary bone tumours of the appendicular skeleton, but is applied to tumours of the spine as well. It prognosticates as well as guides surgical resection margins and has been validated in the literature. The Weinstein-Boriani-Biagini surgical staging system¹³ is an anatomical classification based on the Enneking system that describes the number of vertebra as well as the number of sectors within a vertebra involved. Recommendations are then made for the most appropriate approach to resection of the lesion. We used the Enneking staging system. According to this system we treated six benign and twelvw malignant tumours. Of the benign group five could be classified as active and seven as aggressive. Appropriate treatment may be observational (Enneking grade 0 latent lesions) or surgical (most other lesions), depending on the level of pain, instability, neurologic compromise and the natural history of the lesion.⁵ The aims of surgery are complete resection of the lesion where feasible with preservation of neurological function. We used a variety of surgical strategies customised to the patient and the tumour. In peripheral malignant sarcomas the most important factor affecting survival is complete resection of the tumour with a wide

margin. 14,15 There are however no reports prescribing surgical margins in spinal tumours. Traditional intralesional resection in a piecemeal fashion is thought to increase the likelihood of local recurrence. True wide resection would cause unacceptable morbidity as it would involve resecting the segment of spinal cord. Total en bloc spondylectomy in a single stage from an all posterior approach was described by Tomita. 14 Here wide margins are achieved except at the pedicles and occasionally the spinal canal. This approach decreases local recurrence rates. 16-18 In benign aggressive lesions surgical eradication also provides the best long-term cure.6,18 We had three cases of local recurrence in our series. Two were in malignant tumours where a marginal resection was performed. The other recurrent case was a thoracic giant cell tumour encapsulating the aorta and forcing the surgeon to perform an intralesional resection.

Recent advances in chemotherapy have led to improved survival in malignant primary bone tumours, including in the spine. ^{7,19} Adjuvant radiotherapy is indicated in some malignant and benign aggressive lesions. ^{6,9} The use of local radiotherapy does increase the risk of major local complications, including sepsis. ^{8,10} We used radio- and chemotherapy in a number of cases as guided by our local oncology unit. The main limitations of this study are the retrospective character and the relatively small numbers.

Conclusion:

Most primary bone tumour of the spine in our population occurred in adults and was malignant. Surgical treatment resulted in pain reduction and neurological improvement, allowing a rapid adjuvant therapy in good clinical condition. An experienced multidisciplinary team of tertiary care center should perform diagnosis and treatment of these extremely rare tumors. In order to optimize management and elaborate guidelines, we encourage national or international registries, so that correct epidemiology, better classification and understanding, improved diagnostics, superior treatment and an overall better outcome may be achieved. We conclude that primary bone tumours of the spine are often associated with a significant delay in diagnosis. Surgical strategy and adjuvant therapy should be individualized for each case. Acceptable results with minimal complications can be achieved with this approach.

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