

## MEDICAL REVIEW

# Primary Bony Tumors of the Pediatric Spine

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### REPORT OF A CASE

A nine year old boy presented with an eight month history of neck and back pain. The child had been experiencing persistent progressive neck and upper back pain for at least one year. The pain was typically worse at night and was immediately relieved by the use of nonsteroidal anti-inflammatory drugs (NSAIDs). The child also complained of progressive right shoulder and arm pain with some loss of strength and numbness. There was no history of trauma, but he did report being relatively active in sports prior to the development of these symptoms. Physical examination revealed tenderness over the lower cervical spine as well as pain on both flexion and extension of the neck. Rotation of the neck was also limited, to a greater extent on the right than on the left. Sensory and motor examinations were normal. Further examination revealed that the pain associated with cervical movement radiated along the distribution of the C5 nerve root to the right shoulder and arm. Radiographic evaluation of the cervical spine revealed sclerosis of the right lateral arch of C5. This was compared to a

normal plain radiograph of the same area six months prior on initial presentation. Bone scan demonstrated a corresponding area of increased uptake and CT scan showed sclerosis of the right lateral arch of C5 with a central nidus calcification. An MRI showed low signal on T1-weighted images and a high signal on T2-weighted images. The clinical and radiologic presentation was thought to be consistent with osteoid osteoma of the cervical spine.

### INTRODUCTION

Persistent backache without a history of trauma is an uncommon complaint in children and adolescents, and when it does occur it should be fully investigated [1]. When obtaining the history, questions should be asked about the mechanism of onset, exacerbating factors, and frequency, duration, and severity of the pain. In children who seek medical attention, the incidence of organic causes for symptoms is high. Imaging studies are very helpful, establishing a diagnosis approximately 50 percent of the time [2]. It has been reported that 85 percent of children with pain

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lasting more than two months will have a specific diagnosable lesion [3]. Primary neoplasms of the spine in children are relatively infrequent. Nevertheless, neoplasms represent an important group of entities for diagnostic consideration in the child with a solitary lesion of the spine. The vast majority of spinal neoplasms are disorders without the predilection to metastasize, while a very small group represent primary malignant tumors. In children, the most common benign neoplasms of the spine, include osteoid osteoma, osteblastomas, and aneurysmal bone cysts. All of these tend to cause lysis of part of the vertebra without involving the affected vertebral body or intervertebral disc [4]. Primary nonlymphoproliferative malignant tumors affecting the vertebral column include Ewing sarcoma or primitive neuroectodermal tumor, and osteogenic sarcoma.

Site of origin is a vital clue in differentiating pediatric spinal tumors, with the posterior elements mostly being affected by osteoid osteomas, osteblastomas, and aneurysmal bone cysts, and the anterior elements (vertebral body) being more prone to be affected by hemangiomas and eosinophilic granulomas [5]. These lesions can also be characterized by their imaging features. A small radiolucent nidus with central calcification in the posterior elements of the vertebral body, as in this case, usually indicates the diagnosis of osteoid osteoma. Large expansile lesion with multiple fluid-fluid levels are usually aneurysmal bone cysts, and aggressive osseous and soft-tissue involvement is typical of osteosarcoma [6].

In one of the largest series of pediatric spinal tumors, local (79 percent) and radicular pain (39 percent) were the primary complaints, followed closely by neurologic deficits (74 percent) [7]. The duration of symptoms was significantly shorter with malignant tumors (11 weeks) compared with benign tumors (26 weeks).

Radiographic abnormalities were seen in the vast majority (98 percent) of cases. Recurrence was seen in six of forty-five children and the overall mortality rate was 6.7 percent occurring only with malignant tumors.

We now review primary tumors of the vertebral column, highlighting some of the key features of the more common lesions.

## OSTEOID OSTEOMA

Osteoid osteomas account for approximately 11 percent of all benign bone tumors [8]. Eighty percent of patients are between the ages of five and twenty-four at presentation, with a 3:1 ratio of affected males to females [9-11]. This is a benign lesion, approximately 1 to 2 cm in size, with clear margins from the encompassing reactive bone. Microscopically, it is well circumscribed and composed of randomly interconnecting trabeculae of woven bone that is prominently rimmed by osteoblasts. The stroma surrounding the tumor bone consist of loose connective tissue that contains many dilated and congested capillaries [58]. Ten to 20 percent of all osteoid osteomas are found in the spine. The lumbar area is most commonly affected, but other areas of the spine such as the cervical and thoracic regions may also be involved. There have even been some reports of osteoid osteoma affecting the sacral region [4, 12, 13]. Osteoid osteomas usually presents with a persistent backache. The pain is described as an aching pain worse at night and typically relieved by small doses of aspirin or other NSAIDs. Patient may also have localized vertebral or paravertebral tenderness. There have been reports of patients with soft tissue swelling over the spinous process of the involved vertebra in addition to tenderness over the spine [14].

Osteoid osteomas arising in the spine are usually located in the posterior ele-

ments. The perpendicular region of the arch is most often affected and reactive sclerosis of the pedicle, lamina, or transverse process is usually present. Less than 10 percent of spinal osteoid osteomas affect the vertebral body. These lesions are usually associated with painful curvature, with the concavity ipsilateral to the tumor site, without significant spinal rotation [15]. Osteoid osteoma is the most common cause of painful scoliosis in adolescents. The painful curvature is secondary to paravertebral muscle spasm, with the muscles located closest to the nidus exhibiting the greatest spasm and creating the concave curvature [15, 16]. A lateral location of the lesion within the vertebral body or bony arch is more likely to lead to the development of scoliosis [17]. Although initially attributable to paravertebral spasm, scoliosis may consequently result in growth disturbances, due to longstanding hyperemia causing localized overgrowth of bone and vertebral deformity [18, 19]. Osteoid osteomas in the cervical spine may present with painful torticollis with increased pain on motion [20].

Occasionally osteoid osteomas of the spine may not elicit a significant bony reaction and so may be difficult to assess on plain radiographs. The use of a radioisotope such as technetium-99m bone scan is very useful in demonstrating areas of increased uptake indicative of the neoplasm. The nidus of the tumor may not be visible on routine studies, but a CT scan is usually able to detect central nidus calcification [21].

Therapy should include symptomatic relief, but surgery is usually indicated. There are three main approaches to the removal of the nidus (1) wide *en bloc* resection, (2) unroofing of the nidus by gradual removal of overlying reactive bone with curettes and burrs, and (3) percutaneous CT-guided core drill excision and destruction of the nidus by laser,

radiofrequency, or absolute ethanol [23]. Surgical removal of the lesion usually relieves the pain and other associated symptomatology, but may present special technical problems due to the location of the lesion [24]. Symptomatic relief with aspirin should be initiated alone if surgery is too risky as the lesion sometimes resolves spontaneously.

## OSTEOBLASTOMA

Osteoblastoma is a benign tumor of bone that is histologically identical to an osteoid osteoma but is several centimeters larger. It may measure up to 10 cm at its greatest diameter [24]. The primary difference between the two is the tendency for the osteoblastoma to form a less sclerotic and more expansile mass. Osteoblastoma occurs with only one tenth the frequency of osteoid osteoma, accounting for approximately 1 percent of primary bone tumors [25]. The age range for this tumor is similar to that of osteoid osteomas, with most patients being younger than twenty years old [26, 27]. Males are also more affected than females with a 2 to 1 male to female ratio [28]. Osteoblastomas affect all areas of the vertebral column including the sacrum. Like osteoid osteoma, this lesion commonly affects the posterior elements; it is an expanding osteolytic lesion that may affect not only the lamina and transverse processes but even the adjacent rib.

The presenting symptoms of osteoblastoma are primarily pain, scoliosis, and muscle spasms, but the pain is much less proven than that of osteoid osteoma, and it is not as easily relieved by aspirin [29]. Due to the size of these lesions they are more likely to be first noted as a palpable mass [31]. Some patients may also present with neurologic symptoms. In particular, lesions in the neural arch may extend into the intraspinal canal and compress the spinal column,

resulting in numbness, tingling, radicular pain, and even paraplegia [32, 33].

Spinal osteoblastomas are typically expansile with a scalloped or lobulated contour, well-defined margins, and possibly a sclerotic rim. Only 50 percent of these tumors elicit a reactive sclerotic reaction, and over 30 percent of them will extend to involve the adjacent vertebral body. This can serve to distinguish them from osteoid osteomas where involvement of the vertebral body is uncommon and sclerosis the rule [4]. The stroma of both osteoblastomas and osteoid osteomas is highly vascular and may exhibit some degree of secondary aneurysmal bone cyst degeneration [27]. Differentiation between an aneurysmal cyst and an osteoblastoma may be difficult. Proper differentiation requires review of all tumor areas to make a definite histologic distinction. Osteoblastomas may also appear very similar to osteogenic sarcomas. This confusion may especially arise when only histological sections without roentgenograms are present. However, close histologic analysis should reveal an absence of the sarcomatous connective tissue, tumor giant cells, and frequent mitoses seen in osteosarcomas. Schajowicz and Lemo have reported a subset of osteoblastomas termed malignant osteoblastomas, a rare osteoblastoma that metastasizes but still has the histologic characteristics of a benign lesion and may even be classified as a low grade osteosarcoma [34]. The MRI appearance of spinal osteoblastomas is varied and shows no characteristic features. MRI may also overestimate the extent of the lesion due to extensive reactive changes in the surrounding soft tissue. CT scanning is the investigation of choice for the characterization and local staging of suspected spinal osteoblastomas [35]. Medullo-spinal angiography can confirm the vascular involvement of the tumor nidus and is required to identify arteries supplying the spinal canal that would modify operative strategy [36].

## ANEURYSMAL BONE CYST

Aneurysmal bone cysts are benign, vasocystic tumors characterized by the presence of channels and spaces of various size that usually contain blood but may also contain clear fluid [24, 37]. They may exist in either of two forms: a primary form that arises *de novo* or a secondary lesion associated with another benign tumor such as giant cell tumor, chondroblastoma, or osteoblastoma. Bonakdarpour, Levy, and Aegerter in their study reported that approximately 65 percent of cases are primary and 35 percent secondary [38]. Aneurysmal bone cysts are seen during the first and second decades of life. Unlike osteoid osteomas and osteoblastomas, they are found more commonly in females with a 2:1 male:female ratio [39]. In the spine, the tumor may be located in either the posterior elements — pedicles, laminae, spinous processes — or in the vertebral body [40].

The patient may initially complain of local pain of weeks to months duration at the site of the lesion. With continued enlargement of the lesion there may be compression of the nerve root leading to motor weakness and sensory disturbances. Lesions in the lumbar region may result in loss of bladder and bowel control [24]. There have been reports of progression of symptoms in pregnancy, possibly due to the hyperemic-hemorrhagic characteristics of the lesion [39]. CT scan are often helpful in confirming the diagnosis and showing fluid levels in the cavity, and MRI may give further information on the fluid content. Angiography may show characteristic intense, diffuse accumulation of contrast media.

Radiotherapy has been used in the care of patients with aneurysmal bone cysts of the spine, and at a dose of 30 to 40 GY has proved effective [41]. Most surgical cases utilize simple curettage, where the posterior elements are resected and the involved structures curetted. Recently selective embolization of nutrient vessels

has proven effective in producing ossification of the lesion; percutaneous injection of a fibrosing agent (Ethibloc) has been used with a successful outcome. This may be used in conjunction with surgery or by itself [42]. A post-operative CT or MRI is recommended after embolization to obtain a baseline with which to compare future imaging.

### HEMANGIOMA

Cutaneous hemangiomas are the most common tumors of infancy, affecting approximately 10 percent to 12 percent of children by age one [43]. Hemangioma of the bone is much less common. They may occur as solitary or diffuse lesions. Solitary lesions are often in the vertebral bodies where they are almost a normal variant. These lesions are typically asymptomatic and are usually found incidentally. They have a typical radiographic appearance with either coarsened trabeculae lying adjacent to the vascular channels or multifocal lytic areas creating a honeycomb pattern [44].

### EOSINOPHILIC GRANULOMA

Eosinophilic granulomas encompass a group of conditions that are characterized by the presence of granulomatous lesions with proliferation of Langerhan cells, a particular type of antigen presenting cell or histiocyte [45]. The disease has a peak incidence between the ages of one and three, with a 2:1 ratio of affected males to females [46]. The vertebrae are the second most common site affected (the skull is the first) within the vertebral column. The cervical vertebrae are most commonly affected followed by the thoracic and lumbar vertebrae. The lesion usually arises in the vertebral body with varying degrees of involvement and collapse [9]. Spinal cord compression secondary to extension into the epidural space producing paralysis is a rare complication

[47]. The prospect of spontaneous remission and variations in clinical presentation have made establishing a treatment regimen problematic.

### MALIGNANT TUMORS

The majority of primary and metastatic malignant tumors involving the spine occur in adults. Rarely, tumors such as Ewing's sarcoma and osteogenic sarcoma may affect the pediatric spine. Ewing's sarcoma is the second most common primary malignant bone tumor of children and the fourth most common malignant tumor of bone overall [48]. Approximately 65 percent of Ewing's sarcoma occur in the second decade of life; there is a 1.6:1 male predominance [48, 49].

The most common site of origin is the femur (20 percent); tumors in the vertebral column represent only a small fraction of Ewing's sarcoma. Local pain is the primary complaint (90 percent of patients). The pain may be intermittent which may serve to obscure the diagnosis. Ewing's sarcoma of the spine frequently results in neurological deficits. In the Mayo clinic series, 58 percent of patients with disease of the spine presented with a neurologic manifestations, including tingling sensation, sciatic pain or motor weakness [50]. Forty-seven percent of patients in this series had an open biopsy and underwent a decompressive laminectomy. All patients received radiation therapy in various dosages, intensive combination chemotherapy was administered to 32 of the patients. Nine patients were free of disease at the final follow-up examination (follow-up ranged from 6 to 184 months). With the advent of effective chemotherapy the 5-year survival rate has increased from 5 percent to 15 percent to 75 percent; at least 50 percent are long-term cures [58]. No significant correlation was found between the location of the tumor in the spine and the length of disease-free survival, overall survival, or incidence of metastatic disease [50].

Primary osteosarcoma of the spine is an exceedingly rare disease in the pediatric population. When it does occur, distinction between osteblastoma and osteosarcoma is the primary concern. Osteoblastoma usually originates in the posterior elements and extends into the vertebral body, whereas osteosarcoma tends to arise in the vertebral body and extend into the posterior elements [51]. Usually located on lumbar vertebrae it can also be found elsewhere in the vertebral column [52]. Microscopically, the tumor cells vary in size and shape and frequently have hyperchromatic nuclei. Bizarre tumor giant cells are common, as are mitoses. The formation of bone by the tumor cells is most characteristic of osteosarcoma and the neoplastic bone has a coarse, lacelike architecture. It can also be deposited in broad sheets or as primitive trabeculae [58]. Its radiographic aspect is one of lysis in 49 percent of cases, but sclerosis can also be seen in 27 percent of cases. Osteosarcoma is usually classified as either primary or secondary. Primary osteosarcoma occurs without evidence of pre-existing lesion or prior exposure to radiation. Secondary osteosarcoma arises from a pre-existing lesion or bone that has been irradiated. There have been reports of children treated with radiation for retinoblastoma and neuroblastoma who were then diagnosed with osteosarcoma of the cervical vertebrae up to ten years later [53]. Patients usually present with local pain or swelling at the affected site occasionally brought to attention by trauma. The prognosis of primary vertebral osteosarcoma is poor. The average survival time is only 15.3 months, and the relative risk of recurrence compared to a femoral lesion is 3.9 [52].

One of the most important factors affecting survival prognosis and the tendency for recurrence was the ability to obtain complete resection at the time of resection [7]. The incidence of spinal deformity or instability after multilevel

lumbar or thoracolumbar laminectomy in children and young adults is quite significant (28 percent) [54]. Children should be followed carefully for many years after the initial surgery for early detection of any deformities or instabilities of the spinal column.

Kawahara and Tomita in their evaluation of total *en bloc* spondylectomy for complete resection of primary vertebral tumors reported seven patients operated on between 1989 and 1993. They found that all of the patients had partial or complete pain relief after this procedure, none of the patients worsened neurologically, and only one recurred locally. This new surgical technique offers one of the most radical therapies for primary malignant tumors [55].

## CONCLUSION

Evaluation of back pain in the pediatric population may sometimes be a perplexing task. Many patients present with intermittent symptoms that may be non specific, but back pain with no history of trauma may be the first clue to the occurrence of a possible pathologic process. Careful radiographic evaluation is required. Stella and DeSanctis, in a review of 50 children presenting with vertebral tumors found that there was often a delay before proper diagnostic studies were obtained [56]. When vertebral column lesions are suspected, a bone scan followed by CT imaging has proven to be useful even if plain films are normal. If the diagnosis is unclear after bone scan and CT, MR imaging may be required. In fact, some authors advocate MRI scan of the spine as the investigation of choice [2, 57]. The vast majority of tumors are benign such as osteoid osteoma and osteoblastoma. These tumors have a characteristic clinical and radiologic presentation. Very rarely, malignant tumors such as Ewing's sarcoma and osteogenic sarcoma appear in the pediatric spine. In patients in

whom no diagnosis can be made at first, continued monitoring is appropriate because a diagnosis may become apparent with time. Benign tumors are usually well managed with surgery. The malignant tumors have a poor prognosis even when aggressive chemo and radiation therapy is employed. An initial high index of suspicion is key to making an early diagnosis and possibly improving the outcome.

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