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CASE REPORT

Ossifying parosteal lipoma of the thoracic spine: a case report and review of literature

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Introduction: Lipomas are derived from the mesodermal germ layer and are frequently encountered in adults, and account for almost 50% of all soft tissue tumors. Lipomas are classified based on their component tissues and location. A rare subtype, ossifying parosteal lipoma, accounts for 0.3% of all lipomas and occurs with intimate association with the underlying periosteum of the adjacent bone. Though lipomas are considered to be benign tumors, ossifying parosteal lipomas can manifest symptoms due to their location and relationship to nearby skeletal tissues. We herewith report the first known case of ossifying parosteal lipoma presenting in the region of the thoracic spine.

Case presentation: An otherwise healthy adolescent boy presented with a 3-year history of a slowly enlarging painless thoracic mass. A general physical examination was normal, aside from a painless 10 cm mobile, hard mass along the posterior spine in the region of T4 through T6. Musculoskeletal and neurovascular examinations were normal. An ultrasound suggested a solid, cylindrically shaped mass with diffuse ossification. The mass was resected, and the pathology revealed ossifying parosteal lipoma without evidence of malignancy.

Conclusion: Ossifying parosteal lipomas are rare, benign soft tissue tumors that should be added to the differential diagnosis of thoracic masses.

Keywords: *ossifying parosteal lipomas*; *rare benign soft tissue tumors*; *pathology diagnosis*

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Lipomas are derived from the mesodermal germ layer and are frequently encountered in adults, accounting for almost 50% of all soft tissue tumors (1). They can occur anywhere in the body, but are localized in the subcutaneous tissue. There are many subtypes of lipomas. The classification is based on their constituent tissues and locations. A rare type of tissue involvement is ossification. Less than 1% of lipomas were ossified in one study of 635 cases (2). Ossifying lipomas are usually located in the intraosseous region or adjacent to bone tissues. In such cases, they are classified as parosteal ossifying lipomas (3). Parosteal lipomas are rare, accounting for 0.3% of all lipomas (4). To the best of our knowledge, only three cases have been reported involving the cervical spine (5–7) (Table 1), and there have been none reported involving the thoracic spine.

Case report

An otherwise healthy adolescent boy presented with a 3-year history of a slowly enlarging painless mass alongside his thoracic spine. There was no history of trauma or other prior similar tumors. His physical examination was normal, with the exception of a painless, hard, 10 cm mobile mass in the thoracic region. It was cylindrically shaped, smooth, and located longitudinally along the spine. There were no lymph nodes palpable in the axillary or groin regions. Musculoskeletal and neurovascular examinations were normal. No laboratory abnormalities were seen. An ultrasound of the thoracic back showed a homogeneous, discrete solid mass with diffuse calcifications. The mass was excised in whole. It was strongly adherent to the T4, T5, T6 spinous processes, but there was no invasion to the spinal cord.

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Table 1. Summary of age, sex, location, treatment, and follow up of ossifying lipomas that are being reported

Age	Sex	Location	Treatment	Follow up	Author
20 years	M	Frontal region	Excision		Cil et al.
49 years	F	Gleno-humeral region	Excision		Davis et al.
47 years	M	Thigh	Excision	No recurrence at 18 months postoperatively	Demiralp et al.
55 years	M	Thigh	Excision		Heffernan et al.
15 years	F	Cervical spine C1–C2	Excision	No recurrence at 6 months postoperatively	Bohm et al.
21 months	F	Cervical spine	Excision	Neurological intact 2 years after surgery	Brones et al.
46 years	M	Wrist	Excision	No recurrence at 6 months postoperatively	Demirkan et al.
78 years	F	Vertebral region	Excision	No recurrence at 5 months postoperatively	Nakamizo et al.
51 years	F	Femur	Excision		Bridge et al.
28 years	M	Fibula	Excision		Liem et al.
46 years	M	Neck	Excision		Kameyama et al.
83 years	F	Hand and wrist	Excision	Nerve palsy recovery after excision	Salama et al.
51 years	F	C2–C6 spinous process	Excision	No recurrence at 6 months postoperatively	Yang et al.

On gross pathology, the mass consisted of a fibroadipose tissue mass (7.8 cm in greatest dimension) embedded with irregularly shaped bone and cartilage tissue. No necrosis or cystic change was identified (Fig. 1). Microscopically, the lesion was composed of mature adipose tissue, interspersed with disorganized trabecular bone, cartilage, and fibrotic tissue. The bony tissues were either woven or lamellar in nature, with linings of osteoblasts and focal multinucleated osteoclasts. The osteocytes were small in size and evenly distributed. The inter-trabecular spaces were filled with variable stages of osteoid material with numerous dilated blood vessel sinuses. Focal patchy cartilage tissue was present among with normal bone tissue. The chondrocytes were at different stages of development, and rare double-nucleated chondrocytes were noted. There was no evidence of prominent atypia or sarcoma. The post-operative course was uneventful, and the patient returned to his full, complete functional status.

Discussion

Due to their origin in the mesodermal germ cell layer, lipomas in the deep tissues are less common. Though lipomas are typically composed of only mature adipose tissue, other mesenchymal elements, such as smooth muscle

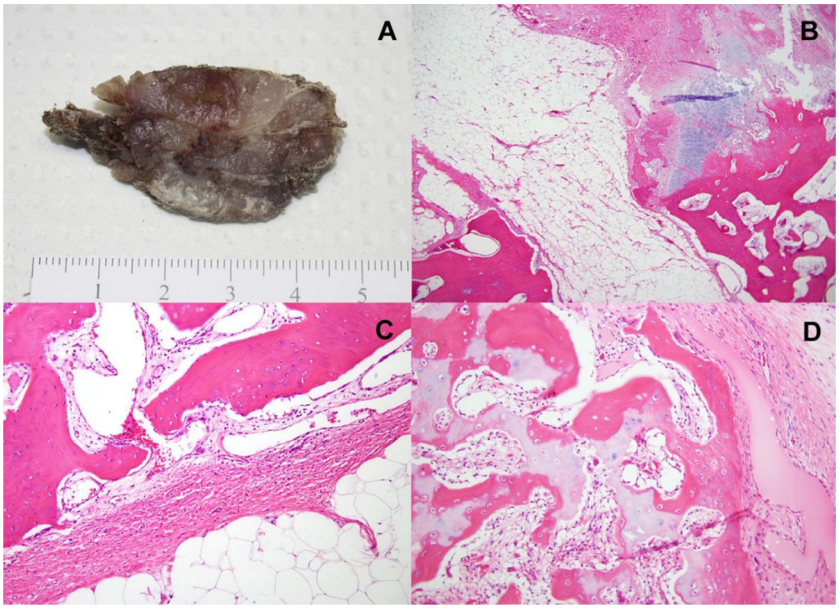

Fig. 1. (A) A cross section of the mass showed multiple fragments of soft tissue and bone; (B) low power histological evaluation showed mature adipose tissue with interspersed trabecular bone, cartilage, and fibrotic tissue (H&E, 4 ×); (C, D) higher power showed the intertrabecular spaces are filled with variable stage of osteoid with numerous dilated blood vessel sinuses. Multinucleated osteoclasts are also present (1D) (H&E, 20 ×).

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or fibrous, cartilage or bone tissue may occasionally be found. If osseous or chondral components are present, the lipoma usually has an intimate connection to adjacent bony structures.

In contrast to subcutaneous lipomas, which are more commonly found in the neck and back, parosteal lipomas are more common in the extremities, occurring adjacent to the diaphysis (8) or diametaphysis (9) of nearby long bones. The most frequently affected sites are the femur, radius, tibia, and humerus. In some series, the skull is also involved (10–11). They have also been reported in the tibia, humerus, scapula, clavicle, ribs, pelvis, metacarpals, metatarsals, and mandible (4). To our best knowledge, there have been no previous reports of thoracic parosteal lipoma in the literature (Table 1).

The age of reported patients with parosteal lipoma range from 3 months to the eighth decade. In about 50% of cases, the patients were aged between 40 and 70. Ossifying lipomas tend to present as large painless lesions that have been present for a long period of time (12). Histological examination typically shows disorganized bone and cartilage tissue. Necrosis may or may not be seen. Critical to the diagnosis is the presence of mature fatty tissue as the predominant tissue composing the mass (13). The differential diagnosis of parosteal lipoma includes teratoma, dermoid tumor, osteochondroma, liposarcoma, myositis ossificans, osteosarcoma of soft tissue, and secondary ossification due to trauma or metabolic changes. Only histopathology examination can differentiate among these entities.

The clinical presentation of ossifying lipoma depends upon the site of origin. When in intimate contact with the periosteum of adjacent long bones, parosteal ossifying lipomas may potentially erode into bone and joint spaces producing symptoms similar to that of degenerative joint disease. In other cases, they can compress adjacent nerves and present as painful lesions. For example, carpal tunnel syndrome can result if the tumor arises in the wrist (14). The clinical course is usually indolent and gradually progressive, consistent with the slow growth of these tumors.

Presence of parosteal ossifying tumors adjacent to spinal vertebrae can be troubling due to the presence of nearby important neurological structures. Farley FA et al., Wilkinson CC et al., and Choi HJ et al. (5–7) reported separate cases of cervical ossifying lipomas. Fortunately, all of these cases presented asymptomatically and underwent excision without complications or recurrence. Had these patients, as well as our patient, not undergone excision but allowed to progress, it is possible that all would have experienced undesired neurologic sequelae. It is fortunate that the proximity of the spine to the skin facilitates early detection and treatment. In general, although parosteal lipomas are benign processes, their size and pressure effects may potentially stretch and impinge upon nerves, resulting in neurologic damage. Surgical excision of such tumors should therefore be expeditious, utilizing care and expertise in freeing important nerve branches from the surfaces of the tumor (15).

The pathogenesis of ossifying lipoma is under debate. One theory holds that these tumors arise after repetitive trauma, metabolic changes, or ischemia, leading to metaplasia of pre-existing fibrous elements within the lipoma and development into osteoblasts (3, 16). One possible link with ossification is transforming growth factor (TGF)-β, as monocytes stimulated by TGF-β secrete chemotactic and mitogenic cytokines that recruit endothelial and mesenchymal cells to promote the synthesis of collagen and associated bone matrix constituents (17). One cytogenetic study of soft tissue parosteal lipoma demonstrated the presence of chromosomal translocation t(3; 12) (q28; q14), suggesting that bone and soft tissue lipomas are cytogenetically similar and likely share a common histopathogenesis (18).

Another theory proposes that mesenchymal stem cells in adipose tissue cause osteoid bone formation. Lin et al. (19) found that mesenchymal stem cells from human lipoma tissue can differentiate into adipocytes, osteoblasts, and chondrocytes after induction in cell culture. Another electron-autoradiographic study of normal and tumor-transformed adipose tissue showed the capacity of adipose tissue cells in lipomas to proliferate and differentiate into osteoid and bony tissue (20).

The treatment for parosteal lipoma is complete surgical resection, as was performed in our case. Incomplete removal can result in local tumor recurrence. Dissection of a soft-tissue lipoma or parosteal lipoma lying adjacent to the bone often necessitates subperiosteal dissection, osteotomy, or segmental resection of the bone.

Conclusions

Ossifying parosteal lipomas are rare benign soft tissue tumors. This entity should be added to the differential diagnosis of a thoracic mass.

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