CASE REPORT

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Intraosseous angiolipoma of head of humerus - An extremely rare entity, with review of literature

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ABSTRACT

Lipoma is a common soft tissue tumor. There are only very few reported cases of intraosseous lipoma in the literature. Intraosseous angiolipomas are even rarer. Angiolipomas are benign tumors consisting of mature adipose tissue and abnormal vessels that occur in patients during their teens or early twenties. Most are found as multiple lesions, often located on the arm or trunk. Predominantly, they are subcutaneous lesions; intraosseous angiolipomas are primarily found in the mandible, ribs, and calvarium. Exact nature of these lesions is debatable. To the best of our knowledge, this is the first reported case of intraosseous angiolipoma of the head of humerus and 9th in the series of reported cases of intraosseous angiolipoma including four of mandible, two of ribs, and two of skull.

Key words: Angiolipoma, head of humerus, intraosseous

INTRODUCTION

Adipose neoplasms affecting bone include soft tissue lipoma and liposarcoma which secondarily involve bone, parosteal lipoma arising in the subperiosteal tissue and intraosseous lipoma and liposarcoma.[1] Intraosseous lipoma is the rarest of benign bone tumor. According to Dahlin, the incidence of intraosseous lipoma is 1 in 1000 cases of all bone neoplasms, Mirra observed 2 intraosseous lipomas in 3000 bone neoplasms, a frequency of 0.1%.[1] A variant of intraosseous lipoma, the intraosseous angiolipoma is an even less common entity.

Angiolipoma, a histological variant of lipoma is one of the rare tumors, histologically consisting of mature adipose tissue and interspersed proliferated vascular component. It accounts for 5–17% of lipomas.[2] Lipoma is a benign soft tissue tumor of mature adipose tissue with no cellular atypia, occurs anywhere in human body where adipose tissue is present. They can be encapsulated or diffuse.[3] Angiolipomas are benign tumors that usually occur in patients during their late teens or early twenties.[4] Most are found as multiple lesions and are often located on the arm and trunk, with the forearm being the most common site. Although predominantly subcutaneous lesions, intraosseous angiolipomas have a predilection for the mandible and ribs with two cases involving the skull reported in the literature.[5-8] The most recent classification of benign lipomatous tumors includes the following categories: Classic lipoma, lipoma variants such as angiolipoma, chondroid lipoma, myolipoma, and spindle cell/pleomorphic lipoma, all hamartomatous lesions, diffuse lipomatous proliferation, and hibernoma.[9]

Although adipocytes are distributed throughout the bone marrow of the human skeleton, lipomas have been considered infrequent primary intraosseous tumors. The first intraosseous lipoma was described by Brault in 1968, involving the diaphysis of the femur. Since then, intraosseous lipomas have been reported in the fibula, the tibia, the ulna, and frontal bone, calcaneus, humerus, and the rib.[10-12]

We report the first case of intraosseous angiolipoma of the head of humerus and 9th case of intraosseous angiolipoma reported till date in literature.

CASE REPORT

A 17-year-old male presented in orthopedic outdoor with complaint of pain in the right shoulder. X-ray was done which showed lytic lesion at the upper end of humerus. Radiologically, the lesion was intraosseous with intact cortical bone, showing septations and trabeculae [Figures 1-3]. He had the previous history of fractures of the head of humerus twice in past 2 years. Differential diagnosis of cystic lesion was kept. Diagnostic biopsy was done. There was intraoperative bleeding from the site. The histopathological section on microscopic examination showed mature trabeculae bone intermixed with mature adipocytes and variably sized dilated vascular channels. No cytological atypia, hyperchromasia, mitosis, or lipoblasts were present. There was no increased cellularity or epithelioid cells. There was no evidence of hematopoietic marrow component [Figs 1 and 2].

DISCUSSION

The term angiolipoma was first described by Bowen in 1912 and differentiated from lipoma histologically in 1960 by Howard *et al.*[13] The first case of intraosseous angiolipoma was reported by Polte *et al.,* in 1976, in the left body of the mandible. Till date, only eight cases of intraosseous angiolipoma have been described, four in mandible, two in ribs, and two in skull. The present case is the

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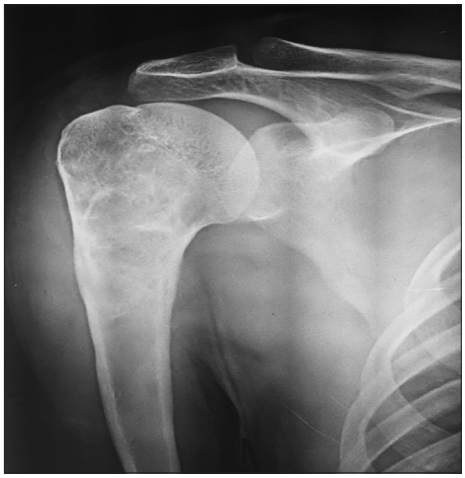

Figure 1: X-ray, lytic lesion, septations, and trabeculae

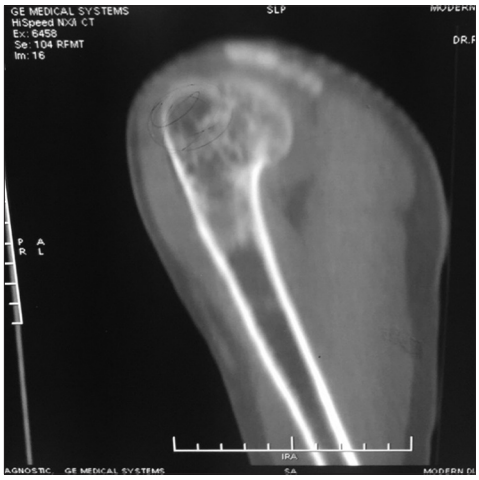

Figure 2: Computed tomography scan showing lytic lesion, septations, and trabeculae

first to be reported in the head of humerus and 9th in the series. Table 1 shows summary of the reported cases of intraosseous angiolipoma till date.

Angiolipomas are morphologically different from lipomas. Radiographically, intraosseous angiolipomas are seen to arise within the marrow space and expand the bone. On computed tomography imaging, lesion exhibit characteristics consistent with its fatty component, in addition to having bony trabeculae and septations. The appear hyperintense on T1 magnetic resonance imaging, with enhancement and T2 imaging demonstrates flow voids, indicating increased vascularity.

Characteristics of angiolipoma are the presence of 50% mature adipocytes, interspersed angiomatous proliferation in the tumor, fibrin thrombi, and presence of numerous mast cells, absence of nuclear pleomorphism, mitoses or necrosis, and absence of other mesenchymal elements (smooth muscle and neural tissue) in contradistinction to other lipomatous lesions such as angiomyolipoma, angiofibrolipoma, angiomyxolipoma, liposarcoma, and hemangioma.[14,15]

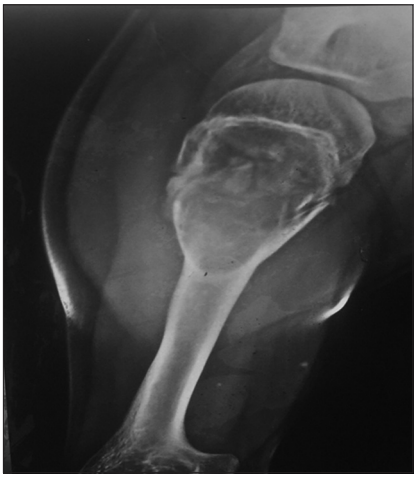

Figure 3: X-ray, fracture of head of humerus 1 year back

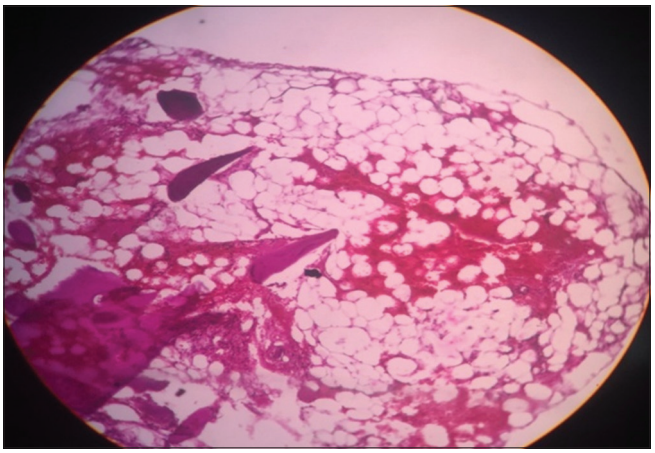

Figure 5: Histopathology, H and E stain, ×40

Scattered fibrin thrombi and presence of mast cells differentiates it from lipoma.[16] Other differential diagnosis is hemangioma. It is believed that angiolipoma is preceded by a lipoma which undergoes capillary and fibrous proliferation from the periphery.[17] The pathogenesis of angiolipomas is unclear. It may derive from embryonic sequestration of multipotential mesenchymal cells and this process becomes activated at puberty by hormones.[13] Some suggest that trauma may lead to angiolipoma.[18] However, most of the angiolipomas do not have a history of trauma. It is supposed that mast cells may play a role in increased vascularity of angiolipoma. Shea and Prieto reported 10 times increase in number of mast cells in angiolipoma than lipomas.[19] Mast cells promote angiogenesis by producing vascular endothelial growth factor, transforming growth factor-beta, and tumor necrosis factor-alpha which promotes inflammation, tryptase stimulates proliferation of vascular endothelial cells.[20] Intraosseous angiolipoma may thus represent either hyperplasia of fat with increased vascular channels or a true neoplasm. There are insufficient data available for a conclusive pathogenesis. Complete surgical excision is the main treatment of angiolipomas and believed to be curative; no further treatment is needed in most lesions.

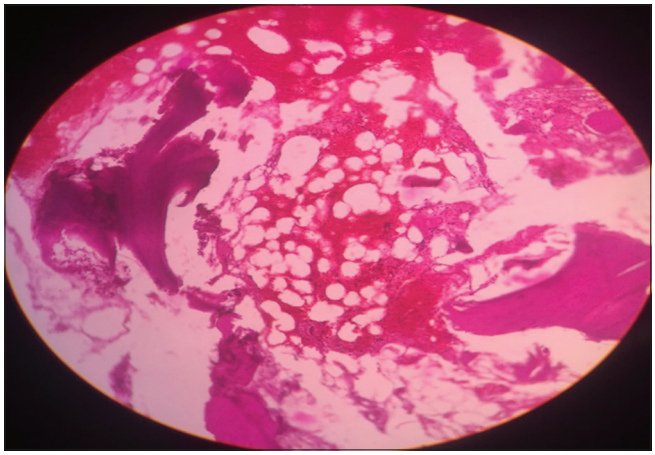

Figure 4: Histopathology, H and E stain, ×40, section shows bony trabeculae mature fat and blood vessels

Table 1: Summary of reported cases of intraosseous angiolipoma

S.No.	Author	Age/Sex	Site
1.	Polte *et al.*, 1976	39/Male	Left body of mandible
2.	Lewis *et al.*, 1980	50/Female	Left body of mandible
3.	Hall *et al.*, 1986	27/Male	Right 3rd rib
4.	Manganaro *et al.*, 1994	51/Female	Left body of mandible
5.	Mardi and Sharma, 2007	--	Rib
6.	Yu *et al.*, 2009	50/Male	Parietal bone of skull
7.	Hemavathy *et al.*, 2012	21/Female	Left body of mandible
8.	Atilgan AO *et al*, 2014	16/female	Frontal bone of skull
9.	Present case	17/Male	Head of humerus

CONCLUSION

Angiolipoma is a variant of lipoma exhibiting proliferating capillaries admixed with mature adipocytes and intraosseous angiolipoma is rare entity. We present an extremely rare case of intraosseous angiolipoma of the head of humerus in 17 years male, the first case to be reported and 9th case of all intraosseous angiolipoma reported till date. The diagnosis of angiolipoma must be considered for these types of lytic bone lesions.

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REFERENCES

1. Manganaro AM, Hammond HL, Williams TP. Intraosseous angiolipoma of the mandible: A case report and review of the literature. J Oral Maxillofac Surg 1994;52:767-9.
2. Kacar S, Kuran S, Temucin T, Odemis B, Karadeniz N, Sasmaz N, *et al.* Rectal angiolipoma: A case report and review of literature. World J Gastroenterol 2007;13:1460-5.
3. Burić N, Krasić D, Visnjić M, Katić V. Intraosseous mandibular lipoma: A case report and review of the literature. J Oral Maxillofac Surg 2001;59:1367-71.
4. Chahlavi A, Staugaitis SM, Yahya R, Vogelbaum MA. Intracranial collision tumor mimicking an octreotide-SPECT positive and FDG-PET negative meningioma. J Clin Neurosci 2005;12:720-3.
5. Hall FM, Cohen RB, Grumbach K. Case report 377: Intraosseous lipoma (angiolipoma) of right third rib. Skeletal Radiol 1986;15:401-3.
6. Lewis DM, Brannon RB, Isaksson B, Larsson A. Intraosseous angiolipoma of the mandible. Oral Surg Oral Med Oral Pathol 1980;50:156-9.
7. Mardi K, Sharma J. Intraosseous angiolipoma of the rib. Indian J Pathol Microbiol 1997;50:606-7.
8. Yu K, Van Dellen J, Idaewor P, Roncaroli F. Intraosseous angiolipoma of the cranium: Case report. Neurosurgery 2009;64:E189-90.
9. Furlong MA, Fanburg-Smith JC, Childers EL. Lipoma of the oral and maxillofacial region: Site and subclassification of 125 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2004;98:441-50.
10. Miller WB, Ausich JE, McDaniel RK, Longo JJ. Mandibular intraosseous lipoma. J Oral Maxillofac Surg 1982;40:594-6.
11. Polte HW, Kolodny SC, Hooker SP. Intraosseous angiolipoma of the mandible. Oral Surg Oral Med Oral Pathol 1976;41:637-43.
12. Barker GR, Sloan P. Intraosseous lipomas: Clinical features of a mandibular case with possible aetiology. Br J Oral Maxillofac Surg 1986;24:459-63.
13. Howard WR, Helwig EB. Angiolipoma. Arch Dermatol 1960;82:924-31.
14. Gonzalez-Crussi F, Enneking WF, Arean VM. Infiltrating angiolipoma. J Bone Joint Surg Am 1966;48:1111-24.
15. Silva-Junior GO, Picciani BL, Costa RC, Barbosa SM, Silvares MG, Souza RB, *et al.* Oral soft-tissue angiolipoma: Report of two cases of rare oral lipomatous lesion with emphasis on morphological and immunohistochemical features. J Oral Sci 2013;55:85-8.
16. Nguyen L, Zwagerman NT, Grandhi R, McFadden K, Richardson RM. Intraosseous angiolipoma of the cranium: Case report and review of the literature. Surg Neurol Int 2014;5:79.
17. Greer RO, Richardson JF. The nature of lipomas and their significance in the oral cavity. A review and report of cases. Oral Surg Oral Med Oral Pathol 1973;36:551-7.
18. Hemavathy S, Roy S, Kiresur A. Intraosseous angiolipoma of the mandible. J Oral Maxillofac Pathol 2012;16:283-7.
19. Shea CR, Prieto VG. Mast cells in angiolipomas and hemangiomas of human skin: Are they important for angiogenesis? J Cutan Pathol 1994;21:247-51.
20. Ribatti D, Belloni AS, Nico B, Salà G, Longo V, Mangieri D, *et al.* Tryptase- and leptin-positive mast cells correlate with vascular density in uterine leiomyomas. Am J Obstet Gynecol 2007;196:470, e1-7.

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