



International Journal of Current Research Vol. 9, Issue, 02, pp.46136-46138, February, 2017

RESEARCH ARTICLE

A RARE PRESENTATION OF INFILTRATING ANGIOLIPOMA

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ARTICLE INFO

Article History:

Received 15th November, 2016 Received in revised form 25th December, 2016 Accepted 19th January, 2017 Published online 28th February, 2017

Key words:

Angiolipoma, Infiltrating, Histopathology.

ABSTRACT

Angiolipomas are benign neoplasms of adipose tissue with a rich vascular component and are classified as either infiltrating or noninfiltrating. Noninfiltrating angiolipoma is seen in young individuals, present as painful, soft, subcutaneous nodules and is treated by enucleation. Infiltrating angiolipoma is a rare neoplasm with only 23 previously reported cases. These lesions are usually unencapsulated or rarely partially encapsulated and tend to infiltrate bony, neural, muscular, and fibrocollagenous tissue. Treatment of infiltrating angiolipomas is aimed at wide excision with radiotherapy indicated for cases of recurrence. A rare case of infiltrating angiolipoma of the nasopharynx causing the eustachian tube dysfunction in a 63 year old male is reported.

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Citation: Kafil Akhtar, Mohd Rafey, Noora Sayeed and Afzal Anees, 2017. "A rare presentation of infiltrating angiolipoma", *International Journal of Current Research*, 09, (02), 46136-46138.

INTRODUCTION

Angiolipomas are extremely rare benign tumors composed of mature lipomatous and angiomatous elements. These are rare benign mesenchymal tumours that are distinguished from common lipomas by a marked degree of vascularisation (Gonzalez-Crussi, 2006). They are differentiated into non-infiltrating and the even less frequent infiltrating angiolipomas. Infiltrating angiolipomas are tumors consisting of adipose tissue marked by vascular proliferation and are nonencapsulated or partially encapsulated. These tumors are very rare in the head and neck. This is a rare case report of infiltrating angiolipoma arising from the nasopharynx causing the eustachian tube dysfunction.

Case Summary

A 63-year-old male presented to the surgical clinic with a history of pain in the left ear and nasal obstruction for the last 3 months. The pain was intense and throbbing. There was no history of associated skin changes, fever, hoarseness, or upper extremity weakness or numbness. He had no history of infection or trauma. USG showed a sessile mass, 5x5 cm in size, non-pulsatile and ill-defined at the left superior-lateral wall of the nasopharynx. Magnetic resonance imaging revealed a heterogenous tumor, enhancing on IV contrast.

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The mass was excised by a transnasal endoscopic approach, which on cut section showed a soft, ill-defined yellowish mass with specks of haemorrhage (Figure 1). Microscopic examination showed univacuolated adipose cells mixed with coalescing single or clusters of blood-filled vessels lined by flattened endothelial cells with narrow, elongated, basophilic nuclei, invading the muscle (Figures 2 and 3). Fibrin thrombi occupied a few of the vessel lumens. A histological diagnosis of infiltrative angiolipoma was made. The patient is currently free of growth after endoscopic surgery at 12 months of follow up period.

DISCUSSION

Lipomas are the most common soft-tissue tumors (Gonzalez-Crussi, 2006). Angiolipoma is a variant of lipoma, with a prominent vascular component, constituting only 6%- 17% of all lipomas (Guzey, 2011). Infiltrating angiolipoma is a very rare subgroup of angiolipomas that shows infiltration to the skeletal muscle and fibrocollagenous tissue (Gonzalez-Crussi, 2006). Gonzalez-Crussi et al first described six cases of unencapsulated lipomatous lesions containing blood vessels, skeletal tissue and deep peripheral soft tissue, called infiltrating angiolipoma (Guzey, 2011). It is usually nonencapsulated or partially capsulated and much more deeply located than other non-infiltrating forms. Angiolipomas are reported in the head and neck region, spine and the thigh. Angiolipomas of the neck tend to be infiltrative in nature. They usually present as painful or tender subcutaneous masses in

young adults. Infiltrating angiolipomas also can lead to muscular pain and neural deficits (Gonzalez-Crussi, 2006 and Konya, 2012).



Figure 1. The mass on cut section showed a soft, ill-defined yellowish mass with specks of haemorrhage

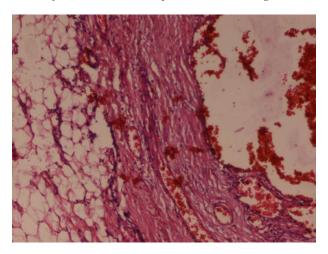


Figure 2. Microscopic examination showed univacuolated adipose cells mixed with coalescing single or clusters of blood-filled vessels lined by flattened endothelial cells with narrow, elongated, basophilic nuclei, invading the skeletal muscle. Haematoxylin and Eosin x 10 X

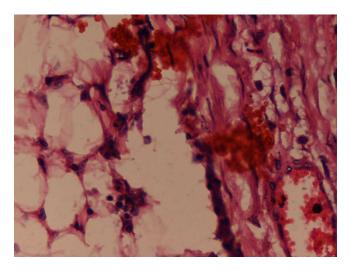


Figure 3. High power of Figure 1. Haematoxylin and Eosin x 40 X

Spinal extradural angiolipomas are rare benign tumors, and most of them are noninfiltrating tumors. Extradural infiltrating spinal angiolipomas extending into the vertebral bodies or posterior vertebral arches are also described in literature (Enver, 2013 and Akyuz, 2013). These tumors are mostly located at the thoracic region and anterior or in the anterolateral extradural space, and they generally infiltrate

only one vertebra (Enver, 2013 and Akyuz, 2013). On ultrasound, angiolipomas are subcutaneous nodules, partially compressible, predominantly hyperechoic with interspersed low echoes (Dalambiras, 2010). On MR imaging, this lesion is capsulated with signal characterisitics nearly following fat on all sequences (Hattori, 2015). Characteristic serpiginous signal voids and linear fibrous bands are present. Infiltrating angiolipomas, however, are much larger tumours, deeply sited, with infiltration into adjacent muscles and compartments. While MR signal characteristics are similar to that of angiolipoma, the lesion is poorly circumscribed, with large vascular channels. On computed tomography, the mass has a slightly hypoattenuating mottled to lobulated appearance with a few hyperattenuating mineralised specks throughout (Jee, 2010). The components of nonfatty regions in angiolipomas are small vessels and capillaries to a variable degree with hyposignal intensity and are well enhanced after infusion of contrast media (Alobid, 2014). The septa of well-differentiated liposarcoma are composed of muscle fiber, lipoblasts, vessels, and inflammatory cells; thus, the signal intensities of nonfatty regions of well-differentiated liposarcoma have been reported with similar findings to those of angiolipoma (Hattori, 2015 and Withrow, 2011). Angiolipomas may be more strongly enhanced than well-differentiated liposarcomas, however, because of the vascular structure of angiolipoma. Treatment of angiolipomas is complete surgical excision. The infiltrating type can create difficulty in excision, with high recurrence rate. In cases where adequate resection cannot be obtained, radiation therapy may be used (McEntee, 2010). A case of successful treatment with α-interferon of a giant infiltrating angiolipoma also has been reported (Akyüz, 2013).

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