Intramuscular Lipoma of Thigh: A Review of Literature

M R Shanker¹, S Prajwal²

¹Associate Professor, Department of General Surgery, Adichunchanagiri Institute of Medical Sciences, B.G Nagara, Nagamangala Taluk, Mandya District, Karnataka, India, ²Junior Resident, Department of Surgery, Shyamnur Shivashankararappa Institute of Medical Sciences and Research Centre, Davangere, Karnataka, India

Abstract

Lipomatous tumors are the most common among the mesenchymal tumors and constitute a large and complex group arising from adipose tissue. A majority of these are located in the subcutaneous tissue. They include the classic lipoma and its variants (fibrolipoma, angiolipoma, myolipoma, ossifying lipoma, chondrolipoma, spindle cell lipoma, and pleomorphic lipoma), lipoblastoma, hibernoma, and infiltrating lipoma including lipomatosis. Intramuscular lipomas (IMLs) constitute <2% of lipomas. They form an important group of deep-seated lipomas occurring deep to deep fascia. In the recent studies by various authors, better understanding of IMLs has led to identify different subtypes of these lipomas as well circumscribed, infiltrative, and mixed consisting of both infiltrative and well circumscribed. Various studies have described distinctive histopathological and imaging features to differentiate these lipomas from others such as atypical lipomatous tumors and hibernomas. This has helped to a large extent in the formulating treatment plan. IMLs can occur in any muscle but occur more commonly in thigh region of a middle-aged male. They frequently pose a challenge to the treating surgeon as they closely resemble atypical lipomatous tumor/well-differentiated liposarcoma. Even with all the available modalities of investigations and histopathological examination, it may be difficult to accurately diagnose these preoperatively.

Keywords: Intramuscular, Lipoma, Thigh

INTRODUCTION

Lipomas are the most common type of benign mesenchymal tumors. They are grouped as either superficial or deep depending on their location in relation to the deep fascia. Intramuscular lipomas (IMLs) are included among deep-seated lipomas.¹ Although they can occur in any small or large muscle of the body, they are more often found in proximal thigh muscles. Fletcher and Martin Bates in their study of 51 cases of both IML and intermuscular lipomas found IML to be 1.8% of all adipose tumors, occurring mostly in middle-aged and trunk region. Whereas intermuscular lipomas formed 0.3% of all adipose tumors, mostly in anterior abdominal wall.²

Paget, in 1853, described a lipoma infiltrating the Trapezius muscle, and in 1946, Regan et al. were the first to describe IML as a rare variant of lipoma. They first introduced the term infiltrating lipoma.³ Later Greenberg et al. recognized that infiltrating lipomas may be either intramuscular or intermuscular based on the classification of Moricini which differentiated between lipomas located either within or in between the muscle.⁴ McTighe and Chernev did a thorough study of IMLs, and they used the terms intramuscular and intermuscular, reserving the term infiltrating lipoma only when there is clear radiological, surgical, or microscopic evidence of infiltration of the muscle or any adjoining structure.⁵

They described the three subtypes of IML, well circumscribed, infiltrating, and mixed consisting of both components. Fletcher Martin-Bates found that 80% were of infiltrative and 17% were of the well-defined type. Most IMLs are located within a single muscle with only few reported involving two or more muscles.⁶

For example a case of well-circumscribed IML, treated in Hospital is presented.

A 43-year-old male reported with painless swelling left thigh, with no prior trauma, slowly increasing in size over 1-year period to attain the size of 16 cm × 10 cm
at the time of presentation. Except for discomfort in the thigh while walking, the patient had no other problem. Joint movements were free. The swelling was non-tender, soft in consistency with diffuse margins occupying the midportion of the anterior compartment of thigh. Horizontal mobility was possible with the relaxed muscle. The swelling became prominent, less mobile and firm on putting muscle into contraction. The skin over the swelling was normal. There were no features of compartment compression or distal neurovascular compromise. The swelling was non-compressible and non-pulsatile. He had no other swelling (Figure 1).

Plain X-Ray thigh revealed a radiolucent shadow in the anterior compartment. A 16 cm × 10 cm ovoid echogenic mass was seen in the rectus femoris muscle USG. Computed tomography (CT) scan was not done. Magnetic resonance imaging (MRI) scan showed well-defined T1 hypointense and T2 hyperintense lesion in rectus femoris muscle, measuring 16 cm × 10 cm in size. The lesion appeared to be confined to the muscle with no obvious infiltration of the lesion into adjacent muscle. The neurovascular bundle was seen separately with no obvious invasion. The visualized marrow appeared normal in signal intensity. The picture was suggestive of benign lesion of rectus femoris muscle likely to be lipoma. (Figures 2 and 3). FNAC was suggestive of simple lipoma.

On exploration of thigh, a well-circumscribed oval-shaped swelling of 16 cm × 09 cm size yellowish lipomatous mass was found after longitudinally splitting the fibers of rectus femoris muscle (Figure 4). Deeper part of the swelling was adherent to the muscle fibers (Figure 5). The mass was soft and lobulated (Figure 6). Neurovascular bundle was free. Cut surface was homogeneous with no areas of hemorrhage or necrosis (Figure 7). Post-operative recovery was complete with normal range of limb movement and normal gait.

Histopathological examination revealed uniform and mature adipocytes with their nuclei pushed to the periphery of the cell. The lesion was well delineated from the muscle fibers. No areas of necrosis, hemorrhage, cellular atypia, or any lipoblasts were noted. There were no entrapped muscle fibers noted within the tumor mass.

At 14 months of follow-up, the patient was asymptomatic with no evidence of recurrence on clinical examination or USG scan.

**DISCUSSION**

Soft tissue tumors, most of them, if not all, arise from the primitive multipotent mesenchymal cell, as per morphological, immunohistochemical data from experimental animals. Murphy et al. believe that malignant transformation of lipomas is non-existent, and the rare reported cases likely represented sampling errors or misdiagnosis at initial investigation. Most lipomatous tumors arise from white fat cells. Whereas brown fat cells give rise to hibernomas. IMLs need to be differentiated from closely resembling intramuscular hibernomas and atypical lipomatous tumors. The term atypical lipoma was coined by Evans et al., in 1979, as a designation for well-differentiated liposarcomas that occur in a subcutaneous or intramuscular location. Since then, the World Health Organization has further refined the terminology and suggests that the term atypical lipoma should be applied only to subcutaneous extremity lesions and that all other such masses should be designated well-differentiated liposarcomas.

**Age and Sex Incidence**

IML is known to occur in any small or large muscles of extremities or trunk, in the age group of 40-70 years, but more commonly in the thigh muscles. They are slow growing causing few or no symptoms. Bjerregaard et al. studied 12 cases of IML and their cases were in the age group between 26 and 72 years. Most of the other studies reported similar age group.

Gender predilection is not clearly established. Different series have reported varying incidence in male and female. Most of the studies reported increased incidence in males. Ramos-Pascua reported male: female ratio of 3:1.

**Anatomical Site**

Although common site is thigh, IML can occur at any site in the body including cardiac muscle. Rare sites such as muscles of hands and feet and other sites are also reported.

**Etiology**

The pathogenesis of IML remains obscure. A wide variety of theories such as metaplasia, trauma, surgery,
Figure 2: Magnetic resonance imaging of thigh-T2-weighted image showing hypointense signal intensity lesion within muscle

Figure 3: Magnetic resonance imaging thigh-T1-weighted image showing hyperintense signal intensity lesion within muscle
chronic irritation, and congenital development have been suggested. Signorini and Campiglio reviewed nine cases of lipoma which occurred following trauma. However, the pathogenetic mechanisms were not understood. Instead suggested the hypothesis of a true adipose tissue neoformation following the trauma. Nigri et al. reported a case of Giant IML 14 years after blunt trauma. Bjerregaard et al. tried to establish some association with hormonal imbalance, but they found no evidence of estrogen receptors in any of them. Some have suggested chromosomal abnormalities.

Clinical Features
IMLs are generally slow growing and painless. However, IML at some sites can cause pain and neuralgia. Various manifestations occur depending on their location. A pectoral muscle lipoma mimicks breast lump, orbital muscle lipoma causes proptosis, temporal muscle lipoma causes headache, a chest muscle lipoma may cause shadow on X-Ray mimicking a pulmonary nodule, and a supraspinatus muscle lipoma may cause impingement restricting shoulder movements.

Imaging Studies
Plain X-Ray may be unremarkable or may show soft tissue shadow. Rarely, they may show ossification in those rare cases of ossification or pure IML. A number of studies have tried to define the characteristics of simple IML as against the atypical lipomatous tumor/well-differentiated liposarcoma on USG, CT, and MRI. Ultrasonographic picture of IML is of a hyperechoic as compared to muscle and heterogeneous mass within the muscle with irregular margins and interdigitations that create the typical striated appearance. When CT is employed, a radiodensity of <50 Hounsfield units is indicative of a soft tissue tumor composed of fat. However, no reliable distinction can be made between a benign lipoma and a malignant liposarcoma on CT. MRI is a better diagnostic modality.
McTighe, Chernov, and Nadege Petit-Clair described IML in detail along with their imaging and histological characteristics. The main characteristics of IML on MRI are round, oval, or fusiform homogeneous with occasional dumbbell-shaped mass. The T1-weighted image is hyperintense and T2-weighted image is hypointense. The muscle fibers intermingling within the lesion gives striated appearance with muscle fibers isointense to adjoining muscle. Well-differentiated liposarcoma on MRI is oblong or oval with dumbbell shape relatively more common and larger size. Heterogeneity due to intermingled fat and muscle is also a feature of infiltrating IML and do not suggest malignancy, though well-differentiated lipoma is also heterogeneous. Nodular and patchy intensities different from fat (non-adipose components), which may form almost 25% of the lesion is a feature of well-differentiated liposarcoma. Completely irregular margins is a feature of IML rather than liposarcoma. Liposarcomas are more often multilobular than simple lipoma with thicker (more than 2 mm) linear structures between the nodules. The septae and globular areas of the tumor tend to be enhanced more prominently in liposarcoma than simple IML.

Malignant tumors are generally larger than 10 cm size although size is strictly not the criteria to differentiate. Tumors of <5 cm have also been found to be malignant. The most statistically significant predictor of liposarcoma was thick/nodular septum. However, like CT, MRI does not allow an absolute, reliable distinction between a lipoma and liposarcoma. A study by Thornhill suggested that the addition of computer-assisted diagnosis may improve the ability of MRI to make this distinction.

Nuclear medicine imaging has not been used extensively in the diagnosis of IMLs. The role of nuclear medicine imaging for the diagnosis of IMLs is limited and not very well established.

Histopathology

Needle biopsy may be inconclusive in detecting atypical lipomatous tumor due to its non-homogeneity. The majority of IML is homogeneous, well circumscribed, and delineated from the muscle and rarely shows areas of hemorrhage or necrosis. Fibrous tissue condensation at the periphery to form a capsule is seen in the well-circumscribed type. Striations due to bundles of muscle fibers passing through the tumor may be seen in the infiltrating type. Microscopically, IML consists of mature univacuolated adipocytes. In the infiltrative type, these adipocytes are seen to be found in between the muscle bundles giving checkerboard appearance on the transverse section and striped appearance on longitudinal section. There is paucity of vascularity. Well-differentiated lipoma shows multivacuolated lipoblasts, cellular pleomorphism, marked vascularization, and mitotic activity. Areas of necrosis infiltrated with lipid-laden macrophages and chronic lymphocytes may be seen. Intramuscular hibernomas, on the other hand, may present as huge deep intramuscular soft tissue mass in adults, closely mimicking well-differentiated liposarcoma or simple IML. Hibernomas are composed mainly of brown fat cells of varying size with granular, eosinophilic, multivacuolated cytoplasm in addition to varying amounts of white fat cells, spindled cells, and myxoid stroma distributed among them. In contrast to IML, hibernomas show increased vascularity. Simple IML lacks presence of lipoblasts, cellular pleomorphism, or nuclear atypia, as is seen in liposarcoma. Chondroid and osseous metaplasia have been reported in long-standing cases. Most of the IML may show features of muscle atrophy as was also found in our case.

Cytogenetic Study

Lipomatous tumors exhibit chromosomal aberrations. Cytogenetic study is of great help in differentiating these soft tissue tumors closely resembling one another. In equivocal cases; immunohistochemistry can be supplemented by fluorescence in-situ hybridization analysis. IMLs show translocation of 12q14-15, 6p21-22, 13q12-14, and 13q22. Lipoma is reported to be MDM2, CDK4, and p16 negative. Atypical lipomatous tumors are generally MDM2, CDK4, and p16 positive. Hibernomas have a translocation involving 11q13 and 10q22 and are negative for CDK4 and MDM2. Immunohistochemistry shows expression of protein S-100.

The cytogenetic study was not done in our case due to lack of facility.

Treatment

The treatment of IMLs depends on the size, location, and the symptoms. Small sized and asymptomatic tumors can be observed. The mainstay of the treatment is surgical removal. The extent of excision recommended varies from simple R0 excision to wide excision and even compartmental excision, in an attempt to reduce local recurrence, which varies from 3-60%. Hyun Ho Han et al. in their review of 27 cases routinely did frozen section biopsy in all their cases. 1 cm margin resection including surrounding muscle fibers was done for cases reported as liposarcoma on frozen section and R0 resection was done for the remaining of their cases. They did not have any recurrences at 3-year follow-up.

Enzinger and Weis (1983, soft tissue tumors) recommended compartmental excision to prevent recurrence. Bjerringgaard et al. treated 12 of their cases with local excision and had recurrence of about 50% among them. Regan et al., 1946 and Dionne and Seemayer, 1974 had similar results following local excision. Kindbiom et al., 1974 did not have any recurrences following wide
excision or myectomy. The follow-up period in these varied from few months to several years. Fletcher and Martin-Bates follow-up period was from 14 months to 19 years. None of the well-circumscribed tumors recurred locally in the study by Fletcher and Martin-Bates. Su Chi, Hong JK, and Chang IL did not have recurrence at 2-year follow-up following wide excision in all their cases of infiltrating IML. Recurrent tumors need to be subjected to surgery once again and malignancy needs to be excluded. Atypical lipomatous tumors may recur sometimes to form dedifferentiated liposarcoma requiring radiotherapy. Otherwise, there is no role of radiotherapy or chemotherapy.

**CONCLUSION**

IMLs are benign slow growing tumors. A lot of studies have focused on accurate pre-operative diagnosis and differentiating them from atypical lipomatous tumors. Misdiagnosed cases of atypical lipomatous tumors result in local recurrence often dedifferentiated. Some studies recommend compartmental excision to prevent local recurrence. Accurate pre-operative diagnosis is important using various criteria. The clinical features, imaging pictures, tissue studies, and cytogenetics report should be analyzed to come to a reasonable diagnosis before planning the treatment.

**REFERENCES**


Source of Support: Nil, Conflict of Interest: None declared.