Accurate diagnostic algorithm for patients with rare pathology of thenar intramuscular lipomas

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Abstract
Intramuscular lipomas which are classified as the subgroup of infiltrative lipomas histologically, are seen rare in the hand. They often localized at the thenar eminence when they do occur in the hand. In such locations, lipomas must differentiate from ganglion cysts which is the most common hand mass. They may cause certain complications due to median nerve entrapment as ganglion cysts. Magnetic resonance imaging and histopathologic investigation are the most effective and reliable methods of confirming the diagnosis. Five patients with the symptoms of mild thumb pain, numbness in the radially innervated digits of hand, and difficulty while gripping objects were included in this study. Radiological assessment included plain radiography, ultrasonography (USG), magnetic resonance imaging (MRI) and electromyography (EMG). Excisional biopsy for all masses within marginal borders were performed. Pathological results revealed intramuscular lipomas. All symptoms were disappeared after excision of the masses. No recurrence and complication was observed at final follow-ups. Intramuscular lipomas are benign lesions with an excellent prognosis after adequate excisions and after clinical suspicious, algorithmic imaging approach in diagnosis can be used by any physician easily as we mentioned in the study.

Keywords: Intramuscular, hand, lipoma

Introduction
Lipomas are one of the most common benign soft tissue tumors. They are formed by mature white fat cells (i.e., lipocytes) [1-3]. They are histologically classified into three groups: angiolipoma, intramuscular (infiltrative) lipoma, and spindle cell lipoma. They rarely occur in the hand, where they may be superficial and deep. Deep soft tissue lipomas located under the deep fascia (sub muscular, intramuscular, or intermuscular) are less common than superficial lipomas [1-4]. Among those pathologies, intramuscular lipoma is a rare variant.

In this report, we present the diagnostic and therapeutic algorithm of the intramuscular lipomas of the hand, especially seen under thenar site.

Material and Methods
5 patients between April 2016 to May 2017 were admitted to outpatient clinic because of progressive intumescence of the hand that had first appeared recently. All patients were female with an average age of 56.6 years (range, 47 to 68 years). The masses were observed on right hand in three of the patients, whereas the remaining had on thenar site of left hands. In addition to this symptom, they reported mild thumb pain, slight numbness in the radially innervated digits of hand, and difficulty while gripping objects. Physical examination revealed large, deeply located masses in the thenar area of hands (Figure 1). The mass was stable and free from the underlying bone, but it became mobilized with muscle movements. The range of motion (ROM) of the thumb in affected hand was assessed fully, and there was no hypoesthesia. At first, ganglion cysts were thought at clinical assessment as the most common benign soft tissue tumors of hand prior to other soft-tissue masses, such as lipomas of the hand. Moreover it is difficult to differentiate lipomas from ganglion cysts by palpation only. In distinction of such benign masses, ganglion cysts allow passage of light on trans-illumination while lipomas do not. However, positive Tinnel’s sign (compression neuropathy sign) may be helpful in confirming lipoma, which cause higher mass-related compression effect than ganglion cyst.

As an initial radiological assessment, x-rays showed radio dense masses over the thenar area without any bony pathology.

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Ultrasoundographies of the thenar area showed various size hyperechoic intramuscular solid masses. Subsequently, magnetic resonance imaging (MRI) revealed a well-defined intramuscular lipomas with regular borders and same signal intensity as mature adipose tissues (Figure 2). Masses also had a low-intensity signal on fat-suppressed MRI scans, suggestive of intramuscular lipoma. Finally, median nerve electromyography (EMG) was performed to assess the hypoesthesias of the patients, however no significant entrapment neuropathy was observed. Then we did not need to perform incisional biopsies.

Surgical procedures
An excision within marginal borders was planned for all masses. All patients had regional anesthesia with tourniquet applied of affected extremity. Surgeries were performed with thenar site incisions while the patients were under regional anesthesia. The fascias were opened, the abductor pollicis brevis muscles were separated by blunt dissections, and the masses were excised within marginal resection fields (Figure 3). Hemostasis was secured before closure, and the excision materials were sent to pathology for histopathological examination in diagnosis.

Results
The average operation time was 60 minutes (range, 50 to 65 minutes) and complications such as neurologic deficit, infection or recurrence were not observed postoperatively. Pathology revealed intramuscular lipomas for all samples (Figure 4). The patients were followed up for about one year postoperatively without any comorbidities. They had regained full range of movement of thumb, hand and wrist. 3 questions were asked to patients to consider any sign of recurrence clinically. One is about the numbness of the hand, the other is about the degree of the pain of the thumb, and the last question is related with cosmetic appearance of the thenar site of the hand to check if there is any residual tumor. All questions were answered in every clinical assessment, then no further imaging study or treatment modalities were performed.
Discussion

Lipomas are the most common soft tissue tumors and may be observed anywhere in the body. The exact etiology is unknown, however in a report, most of the intramuscular lipomas occur in large muscles exposed to trauma where the joint’s range of motion is big and much power is used [5]. They are usually localized superficially. Myhre-Jensen reported only 1–2% deep-seated lipomas in their large series [6]. Al-Qattan reported only 3 intramuscular lipomas in their series of 67 upper extremity benign fatty tumors [7]. Weiss and Goldblum classified deep-seated lipomas according to their locations as follows: intramuscular or intermuscular [8].

Moreover intramuscular lipomas are uncommon and occasionally encapsulated, which causes abnormal invasion of soft tissues. They are also known as infiltrating lipomas due to their abnormal characteristic growth pattern. Though both terms (intramuscular and infiltrating) have been used interchangeably, the truth is that not all of them are infiltrating [9]. Lipomas are rarely observed in the hand and very few reports of hand lipomas have been published till now. Lee et al. reported six intramuscular lipomas of the hand, four of which were located in the thenar region and two of which were in the hypothenar area [10]. Zamora et al. reported a case of an intramuscular lipoma that was located in the thenar eminence and was associated with the median nerve distribution [9]. Cribb et al. reported 10 upper extremity lipomas, 5 of which were located in the hand. The number of lipomas in our study is compatible with the literature. The lipomas were in thenar, hypothenar, and wrist locations, and two of them caused paresthesia due to median nerve compression [11]. Schoffl reported a thenar-seated lipoma that caused bony destruction [12]. Zakaria reported five hand lipomas with thenar, midpalmar, and hypothenar localizations [13].

All of these previously described tumors were in intramuscular locations, and they rarely caused median nerve compression. Clinically, they can reach large sizes and cause hypoesthesia with compromised functionality of the hand, mainly with regard to gripping ability. Pain is a late and uncommon and usually observed in deep and large lipomas due to compression or expansion of the adjacent soft tissues [14]. If lipomas do not reveal any symptoms, they can only be followed by periodic follow-ups.

However, intramuscular lipomas are mostly symptomatic masses where the surgical treatment is inevitable. The recommended surgery is marginal or wide excision to prevent recurrences. Debulking is also an acceptable option if the mass is in an unsuitable area or if complete resection will lead to a functional impairment [15]. Patients in current study had disability of gripping function of the thumb and hypoesthesia of the radial innervated digits due to tumor mass effect. So we performed marginal excisions on all masses where the symptoms were disappeared after removal of tumors.

In radiological assessment, MRI is the most effective and reliable imaging method for diagnosing soft tissue tumors. Infiltration of the neighboring tissues visualized on MRI scans may raise suspicion of a liposarcoma. X-rays and computed tomography can be useful for the detection of tumor calcifications, which cannot be detected by MRI and ultrasonography [16]. Nuclear medicine imaging has not been used extensively in the diagnosis of intramuscular lipomas [17].

Ultrasonography shows the hyperechoic mass with irregular margins and skeletal muscle interdigitations, which create a striate appearance with the distinction of liposarcomas. These images allow confident diagnosis of intramuscular lipomas [9]. EMG should be included as a diagnostic modality if a patient has any nerve compression symptoms, as ours did. In addition, many authors recommend biopsy of the tumor to identify the lesion histologically in cases where no diagnosis has been made on the basis of imaging studies. Moreover, in case of giant lipomas, a biopsy should be performed to differentiate from liposarcoma (Table 1) [18].

Table 1. A flowchart of the diagnostic and therapeutic algorithm of the palpable masses of the hand.
As in abovementioned studies, it is considered that symptomatic lipomas should be excised surgically within marginal borders. Recurrence is extremely rare, but when it occurs, it is due to inadequate excision of the tumor [18]. Zamora et al. reported a recurrence 2.5 months postoperatively [9]. Postoperative infection, neural damage, and formation of a painful neuroma are encountered as surgical risks [19]. Our patients had no recurrence or postoperative complications at one-year follow-up.

**Conclusion**

Intramuscular lipomas are benign lesions with an excellent prognosis after adequate excision. Physicians should be mindful of the possibility that the mass may appear as a liposarcoma; therefore, they should manage treatment by using an appropriate algorithm that incorporates MRI and biopsy for accurate diagnosis.

**Clinical Message**

Palpable hand mass should not ignore as a simple case, especially when they are atypical localized. Sometimes it is actually difficult to clarify the diagnosis in such cases. The algorithm mentioned above may be a guide to get satisfactory results.

**Competing interests**

The authors declare that they have no competing interest.

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**Ethical approval**

Ethical approval was received from our institutional review board.

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