Case report

Diffuse intramuscular lipoma responsible for monomelic hypertrophy

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Abstract

Background: Lipomas are common benign soft tissue, however intramuscular lipoma, is an often misdiagnosed clinical entity. The aim of this case is to present an atypical infiltrating lipoma and discuss its diagnostic and therapeutic features.

Clinical features: A 24 year-old patient presented to our consultation with a swelling of the right lower limb from childhood and gradually increasing in size. Clinical examination revealed a firm and extended swelling along the leg, thigh and foot giving an aspect of monomelic hypertrophy. There were no signs of vascular or nerve compression.

Intervention and outcome: Pathological study showed a lipoma. Magnetic resonance imaging (MRI) of the right lower limb objectified a large infiltrating lipoma affecting muscles of the posterior compartment of the thigh and the right leg with no signs of malignancy. The diagnosis of intramuscular lipoma was done. The surgery was proposed but the patient refused.

Conclusion: This case report described an adult female patient with an unusually intramuscular lipoma where magnetic resonance imaging was playing an important role in the diagnostic. Indeed the intramuscular is a benign fatty tumor with characteristic appearance on MRI. Its management should be considered in multidisciplinary collaboration.

Keywords: Infiltrating lipoma; Intramuscular; Monomelic hypertrophy; Magnetic resonance imaging

1. Introduction

Lipoma is the most common form of soft-tissue tumor (Kransdorf, 1995). It is composed of mature adipocytes (Terzioglu et al., 2004) and is usually localized in the subcutaneous tissue, but can also occur in deeper locations (Munk et al., 1997).

Intramuscular lipoma is a deep lipoma; it's relatively rare and has been described in many locations, but has been exceptionally reported as monomelic (Fedell et al., 1978).

The etiology of this lipoma is unknown. Copcu argued that in rare cases they may arise following trauma (Copcu, 2003). This hypothesis was explained by the fact that blunt trauma is always a cause of inflammation in soft tissues. Mediators secondary to inflammation could play a role in the differentiation of precursors of mature adipocytes (Mori et al., 2004) and can probably explain focal lipoma. Another hypothesis was proposed by Mori et al.; they suggested the association of neurogenic or myogenic disorder in the lesions and that fat cell overgrowth into areas of muscle involution could modulate the infiltrative

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growth of these tumors into the surrounding muscle (Mori et al., 2004). In addition, sex hormones may influence the proliferation and differentiation of adipocytes and thus the formation of adipose tissue; the local estradiol concentration in adipose tissue may be a stimulus to the proliferation of preadipocytes (Gregoire, 2001).

The aim of this article is to describe an exceptional case of intramuscular lipoma responsible for monomelic hypertrophy, and describe its clinical features, differential diagnosis, imaging characteristics and treatment options.

2. Case report

A 24-year-old woman was admitted in dermatological department for a 14 year history of swelling of the right lower limb. The patient reported that the lesion has begun in the right foot at the age of 10 years, was removed by surgery but had recurred 1 year later. The swelling had increased gradually in size to involve the entire lower limb (LL). She denies a history of trauma or cancer.

Physical examination noticed a firm and painless swelling of the sole of the right foot predominantly on the proximal part which extended to the posterior leg and thigh with gluteal fold deviation giving an aspect of monomelic hypertrophy (Fig 1). We also found two café au lait spots on the trunk. There were no signs of vascular or nerve compression. Laboratory investigation revealed a normal level of plasma lipid.

Pathological examination of the foot objectived typical and matures adipocytes interspersed with skeletal muscle bundles (Fig. 2). Magnetic resonance imaging (MRI) of the right LL showed two large intramuscular lipomas, involving the muscles of the posterior compartment of the thigh and the right leg with no signs of malignancy (Fig. 3a and b). The diagnosis of intramuscular lipoma was made. A surgical therapy was proposed, but the patient refused.

3. Discussion

Our case described an unusual case of intramuscular lipoma from childhood with monomelic hypertrophy.

Intramuscular lipoma was first described by Paget in the trapezius muscle in 1856 (Kindblom et al., 1974). This subtype of lipoma is rare (1%) and is characterized by infiltration of the muscle tissues or growth between the muscle fibers (Rosai, 1996; Batsakis, 1979). They occur without gender predilection, generally during middle or late adulthood (majority between 40 to 60 years) and began rarely in childhood as in our case (Bjerregaard et al., 1989; Dionne and Seemayer, 1974).

Intramuscular lipoma is often presented without clinical symptoms and therefore grows to a large size before detection unless they compress neurovascular structures (Terzioglu et al., 2004; Nigri et al., 2008). The remarkable characteristics of this subtype of lipoma are large dimensions at diagnosis (over than 5 cm), a deep subfascial location and a predilection for the large muscle groups of the shoulder, upper arm, hip and thigh (Kindblom et al., 1974). They are not always as well circumscribed as the superficial type (Fletcher and Martin-Bates, 1988).

Monomelic distribution is exceptional, to our knowledge just one case was described in English literature; it was a report of a girl of 12 years but hypertrophy of her lower right limb was less developed as in our case (Fedell et al., 1978).

Several differential diagnoses must be excluded (Ohguri et al., 2003). In our case, because of monomelic hypertro-
phy of the LL and the aspect of the foot, we first eliminated Proteus syndrome and neurofibromatosis by the absence of other diagnostic criteria.

Radiologically, standard radiography reveal osseous and cartilaginous changes within the masses, but MRI is the gold standard for assessing soft tissue masses and eliminates liposarcoma (Matsumoto et al., 1999). On MRI intramuscular lipoma may appear homogeneous or inhomogeneous depending upon the amount of intermingled muscle fibers within the fatty mass. In most cases the margin of the lesion is infiltrative but 60% of cases may still appear to be rather well defined (Matsumoto et al., 1999). As in our case, on T1 and T2-weighted images the lesion demonstrates high signal intensity, similar to normal fat. Fat-suppressed sequences demonstrate signal suppression, again similar to normal fat. Linear structures with low signal intensity on both the T1 and T2-weighted sequences are seen in most lesions, and represent muscle fibers within the tumor.

Detailed histological examination is essential to characterize intramuscular lipomas and to eliminate liposarcoma (Matsumoto et al., 2000). It shows mature uni-vacuolated adipocytes of uniform size interspersed with skeletal muscle bundles and varying amounts of fibrous stroma which forms incomplete capsules in some cases (Kindblom et al., 1974). In places the fatty tumor completely replaces the muscle bundles. The fatty cells grow into the endomyosial space, and remaining muscle fibers show extensive damage without atypical nucleus, mucous degeneration, polymorphism, and mitosis (Fletcher and Martin-Bates, 1988).

Surgical excision is the best way to treat intramuscular lipoma. However, the recurrence rate for infiltrating lipomas has been reported to range between 3% and 62.5% (Su et al., 2011). This difference may be explained by inadequate resection of the tumor or misdiagnosing it as an intramuscular lipoma or by the fact that this type of lipoma is not always well circumscribed.

Indications for excision include a mass greater than 5 cm in size, a subfascial location, a tumor that is growing, clinical features such as pain, firmness, or irregularity, or a questionable preoperative needle biopsy result. Another reason for excision includes a patient’s cosmetic concern (Serpell and Chen, 2007). In our case, despite the large size of the tumors and its appearance the patient refused surgery because of risk of recurrence.

We think that lipoma in our patient can be explained by the hormonal theory because of the occurrence of lesions during the prepubertal period; but the presence of unilateral involvement could be explained by the presence a local factor may be exacerbated by surgery on the right foot during childhood.

4. Conclusion

Our case described unusual unilateral and large intramuscular lipoma located on the muscles of the posterior compartment of the thigh and the right leg where imaging was playing an important role in the diagnosis.

Conflict of interest

None declared.

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None.

References

