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CT AND MRI FINDINGS OF A NECK INTERMUSCOLAR OSTEOLIPOMA: CASE REPORT AND REVIEW OF THE LITERATURE

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Abstract: *Osteolipoma is a rare lipoma-variant characterized by bone differentiation. It has the same benign prognosis of a classic lipoma, but intralesional non-fatty components require differential diagnosis from other benign and malign lipomatous entities.*

An intermuscolar osteolipoma occurring in the upper neck of a 68-year-old woman is described. MRI and CT images showed a circumscribed fat-containing lesion with intralesional areas resembling teeth and bones. The histological specimen revealed foci of osseous metaplasia within mature adipose tissue, suggesting the diagnosis of osteolipoma.

Keywords: *Lipoma, Soft tissue neoplasms, Neoplasms, Adipose tissue.*

INTRODUCTION

Osteolipoma represents a rare lipoma variant, consisting of mature adipose tissue and mature lamellar bone[1]. It has been reported in several locations, particularly in the soft tissue of the neck, in the oral cavity and in the extremities (adjacent to bone)[2-4]. It may be confused with other fat-containing lesions, mainly teratoma and well-differentiated liposarcoma[4,5].

We report a case of intermuscolar osteolipoma occurring in the upper neck of a 68-year-old woman, located in the left retromastoid region, adjacent to the occipital squamous bone. Paramastoid location appears to be particularly rare, no previous reports having been published in English literature[2,6] to our knowledge. Written informed consent was obtained from patient f or pubblication of this case report.

Case report

A 68-year-old woman was referred to our hospital complaining of vertiginous symptoms. Patient clinical history revealed a mass in posterior neck, discovered by chance about 2 years before. At the beginning she was asymptomatic for the lesion, and clinical features of a benign-like lesion – such as lipoma – moved her physician to perform a clinical follow-up. However, the patient gradually started to suffer from sub-continuous tensive pain in the neck. The remainder of her medical history showed a rectal cancer treated with radiotherapy and chemotherapy.

Our physical examination confirmed a non-tender mobile mass, which was palpated in the left retromastoid region, with cutaneous profile deformity. An ultrasonography exam confirmed the presence of a well-circumscribed lesion, located in

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the left side of the upper neck, with predominant fat content. The lesion showed inhomogeneous appearance, due to the presence of hyperechoic intralesional areas, with posterior acoustic shadows, interpreted as calcifications.

Subsequently, a head and neck MRI examination was performed, in order to exclude central causes of dizziness and, simultaneously, to obtain a more detailed evaluation of the left upper neck mass.

Unenhanced and enhanced MRI acquisitions – obtained using a 1.5 Tesla scanner – revealed the presence of a subfascial oval-shaped mass, deep-seated, postero-inferior to left retromastoid region. On Fast Spin-Echo (FSE) T1 and T2-weighted images (Figure 1), the lesion was located in the intermuscolar space between splenius capitis and longissimus capitis muscles. Neither vascular involvement nor muscular invasion was detected. It was predominantly hyperintense on T1- and T2-weighted images, with loss of signal intensity after spatial fat suppression (Figure 2). This imaging behavior clearly indicated the presence of lipomatous tissue. However, the diagnosis of a simple lipoma was excluded for the presence of hypointense linear and globular intralesional areas on FSE unsuppressed images. Diagnosis of liposarcoma was excluded, due to the lesion size (<10 cm) and tumor composition greater than 75% in fat. Moreover, stable measurements of lesions and patient clinical history suggested a benign diagnosis.

To better investigate the low-signal internal areas, an unenhanced CT scan was proposed. The exam confirmed the presence of a well-circumscribed predominantly fat-attenuating intermuscolar mass, showing a contact surface with occipital and temporal bones (Figure 3). Neither bone erosion nor focal cortical hyperostosis were reported. In addition, non-fatty intralesional elements were confirmed, appearing as high density scattered areas, with the same attenuation of bones (Figure 3). They were interpreted as a mixture of calcifications, bone fragments and cartilagineous elements. In view of their shape, the presence of teeth was not excluded. Radiological hypothesis of an atypical lipoma, dermoid tumor or teratoma was finally proposed.

The patient underwent surgical excision of the mass. The specimen consisted of a lobulated, finely encapsulated yellowish mass with a smooth surface. After sectioning, bony streaks with red bone marrow were visible. Histopathological examination disclosed that the tumor consisted of mature adipose tissue, with some foci of osseous metaplasia (Figure 4). The final diagnosis of osteolipoma was formulated.

Discussion

Lipoma represents almost 50% of all soft-tissue neoplasms[7]. It generally affects males and females equally, occurring predominantly from fifth to seventh decade of life[8]. It may arise from any site where fat is present, showing superficial or deep location[2,9].

Typical lipomas are composed entirely of homogeneous mature adipose cells, without cellular atypia[10], and may contain a few thin, discrete septa. Benign histological variants of lipoma, containing other mesenchymal elements, are less frequently encountered and have been named according to the type of tissue present. In a recent WHO classification, these variants of soft tissue tumor include: angiolipoma, myolipoma, chondroid lipoma, extrarenal angiomyolipoma, extra-adrenal myelolipoma, spindle cell/pleomorphic lipoma and hibernoma[11-14].

In addition to these WHO variants, "conventional lipomas" – as reported by Nelsen et Al – "can occasionally show areas of bone formation

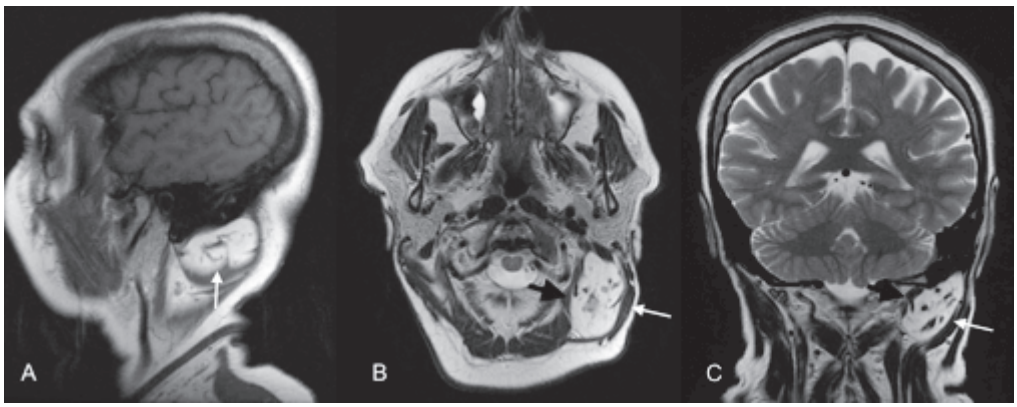

Figure 1. Unenhanced Fast Spin-Echo (FSE) MRI acquisitions – obtained using a 1.5 Tesla scanner. Figure 1A shows the presence of a sub-fascial oval-shaped mass, deep-seated, postero-inferior in left retromastoid region. On Fast Spin-Echo (FSE) T1 and T2-weighted images (Figures 1B and 1C), the lesion was located in the intermuscolar space between splenius capitis and longissimus capitis muscles (*white and black arrows*).

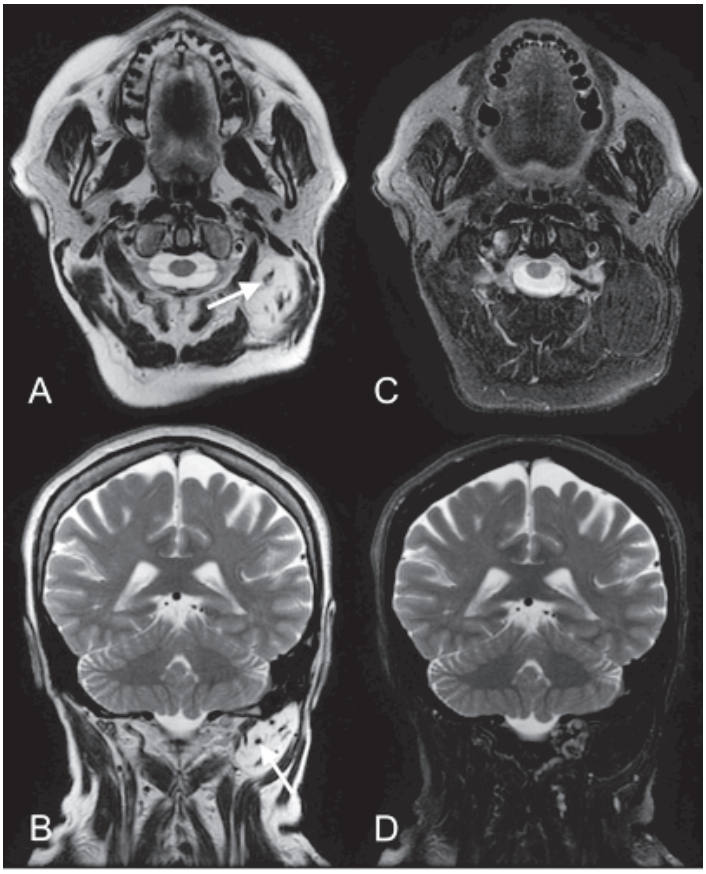

Figure 2. Axial and coronal FSE T2-weighted images. On Figures 2A and 2B, lesion has same signal intensity of the fat. After spatial fat suppression (Figures 2C and 2D), it shows loss of signal intensity. This imaging behavior clearly indicates the presence of lipomatous tissue. However, the diagnosis of a simple lipoma was excluded for the presence of hypointense linear and globular intralesional areas on FSE unsuppressed images (*white arrows* on figures 2A and 2B).

(osteolipoma), cartilage (chondrolipoma), abundant fibrous tissue (fibrolipoma), or extensive myxoid change (myxolipoma)"[15].

The "ossifying lipoma" was first reported in literature by Plaut in 1959[16], describing a lipoma exhibiting mature lamellar bone in a predominant adipose component. Subsequently, the terms "osteolipoma" and "lipoma with osseous metaplasia" have been interchangeably applied to indicate this entity[1,17]. Furthermore, the term "osteochondrolipoma" has been preferred by some authors in the presence of cartilaginous differentiation[6,18] and endochondral ossification. In contrast to the high incidence of lipomas, the occurrence of osseous differentiation within lipomas is considered extraordinarily rare[4-6,13]. In a large series of lipoma retrospectively evaluated by Allen et Al, less than 1% were ossified[19].

Nevertheless, many of the reported cases are located adjacent to the periosteum, with a broad attachment to the underlying bone surface. This variant has been reported as "ossifying parosteal lipomas"[4], and is more common in the extremities, frequently adjacent to the diaphysis or diametaphysis of the femur and rarely elsewhere (scapula, ribs, pelvis, metacarpals, metatarsals, mandible and skull)[17-18,20-21].

On the other hand, the subtype of osteolipoma independent from bone has been mainly described in the head and neck regions[2], both in intracranial (tuber cinereum, hypothalamus, suprasellar cistern, interemispheric area) and extracranial anatomical sites (oral cavity, parapharyngeal spaces, submandibular region)[2,6,22-27]. Apparently, only one case of osteolipoma involving deep neck musculature has been reported; Yang et Al, infact, described an intramuscolar osteolipoma involving the posterior neck muscles in the midline[5].

Osteolipoma formation and origin have been previously debated, and several theories have been formulated. Some Authors[18,24,27] have suggested that occurrence of osseous and cartilaginous differentiation could be a reflection of the presence of mesenchymal stem cells, which differentiate into lipoblasts, chondroblasts or osteoblasts, and fibroblasts, characterizing a "mesenchymoma". Alternatively, it has been suggested that osseous components arise from chondro-osseous metaplasia of pre-existing fibrous elements into osteoblasts. Explanations proposed for this metaplasia are repetitive trauma or ischemia secondary to increased tumor's blood supply[1,4,10,28].

Clinically, osteolipoma is usually a painless, slow-growing mass with an evolution measurable in

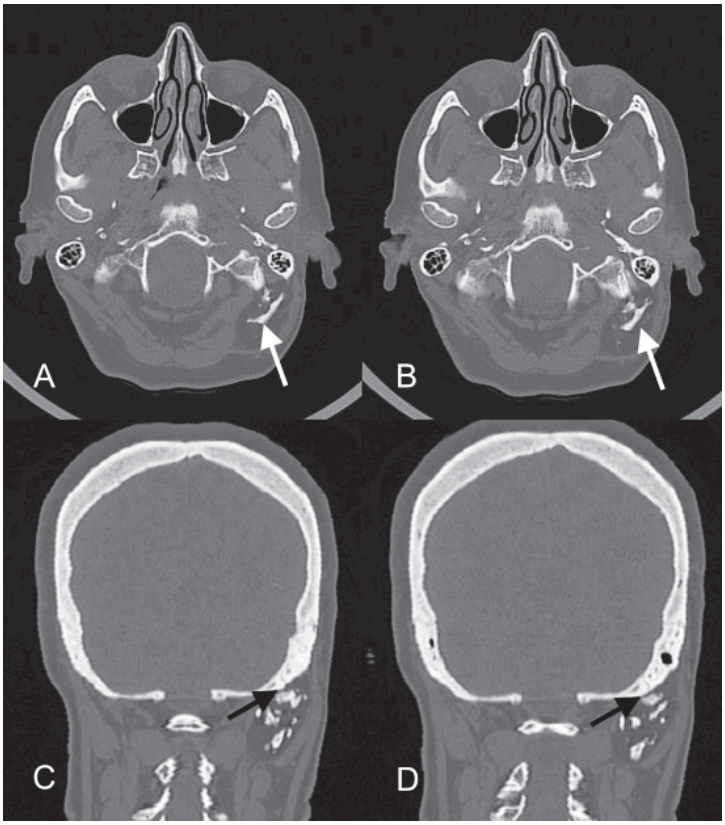

Figure 3. Axial CT scans and coronal multiplanar reconstruction images. The images confirmed the presence of a well-circumscribed predominantly fat-attenuating intermuscolar mass. Non-fatty intralesional elements were confirmed (*white arrows* on Figures 3A and 3B), appearing as high density scattered areas, with the same attenuation of bones (Figure 3). They were interpreted as a mixture of calcifications, bone fragments and cartilagineous elements. Lesion shows a contact surface with occipital and temporal bones (Figure 3C and 3D). Neither bone erosion nor focal cortical hyperostosis were reported.

years. Lesions are generally diagnosed in patients over 40[2]. It has a constantly benign natural course.

Imaging features deviate from typical features of a classic lipoma, consisting of a uniform lipomatous mass with or without thin unenhanced or slightly enhanced septa[29].

MRI usually shows a well-circumscribed lobular mass, predominantly isointense to subcutaneous fat in all pulse sequences. The lesion is generally inhomogeneous for the presence of intratumoral thickened septa and globular areas of non-adipose tissue, appearing as hypointense areas on both T1- and T2-weighted images. Larger osseous excrescences may have an heterogeneous appearance, demonstrate internal signal equal to those of bone marrow, while smaller excrescences present only a component with persistent low SI, which represents cortical bone [30]. On CT scans, osteolipoma has been described as well-circumscribed encapsulated soft tissue mass with fatty matrix and irregular trabeculae of bony density[4,26] or wide densely calcified portions[2,6]. Sometimes additional focal areas of intermediate attenuation have been observed between fat tissue and calcifications, corresponding to chondroid tissue. In our case calcific densities were scattered and curvilinear. CT is a valid diagnostic tool in the evaluation of ossifying parosteal lipomas, which may show an underlying bone reaction (undulating cortex, hyperostosis, osseous excrescences)[6].

The differential diagnosis includes lesions which may contain simultaneously adipose tissue and bone and/or calcifications. Osteolipomas with scattered trabecular bone-densities in a predominantly fatty matrix – as observed in our case report – need to be differentiated from other pathological entities, such as teratomas or dermoid cysts, other lipoma variants and well-differentiated liposarcomas with bone metaplastic changes. Particularly,

Figure 4. Surgical excision of the mass. The specimen consisted of a lobulated, finely encapsulated yellowish mass with a smooth surface. After sectioning, bony streaks with red bone marrow were visible (*black arrows*). Subsequently, histopathological examination confirmed mature adipose tissue, with some foci of osseous metaplasia.

when non-fatty components within lipomatous tumors are detected, radiologists should always keep in mind the possibility of well-differentiated liposarcoma as a possible diagnosis. A differential diagnosis from Teratomas and dermoid cysts could be very difficult. These embryonal neoplasms may occur anywhere along the path of primordial germ cell migration. Most teratomas and dermoid cysts contain fat, which is considered an imaging hallmark for these lesions[31], and calcifications, which could be focal, rim-like, or rarely represented as teeth or bone. Some calcifications could be differentiated from bone elements with difficulty.

The prognosis for osteolipoma is favourable, as it is for conventional lipomas[3]. Surgery is indicated in the presence of large masses showing compression of adjacent structures, or in cases of masses with uncertain characterization, or even for cosmetic reasons.

Conflicts of interest statement and funding: Authors declare that there are no conflict of interest.

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