A cutaneous angiomyolipoma case mistaken for a parotid mass

Naczyniakowłókniakomięsak skóry błędnie rozpoznany jako guz ślinianki przyusznej – opis przypadku

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ABSTRACT:

Introduction: Angiomyolipoma is a benign tumor consisting of vascular structures, smooth muscle and adipose tissue. It is most commonly found in the kidneys and more rarely in the liver, lung, heart, intestines, nasal cavity, oral cavity and ear. Cutaneous angiomyolipomas are very rare and show some differences with non-cutaneous angiomyolipomas; these are mostly located in the distal extremities and are a result of traumas and HMB-45 antibody negativity and appear more frequently in men. Neither of the cases, except for one presented until today, had cellular atypia; thus, cutaneous angiomyolipomas are considered not to lead to malignant transformation. It is thought that total excision of the mass is indicated in the treatment of cutaneous angiomyolipoma.

Case report: Our case presented to our clinic with a left preauricular mass which was thought to be a parotid mass after an initial physical examination. The MRI revealed that the mass did not originate from the parotid gland. The fine-needle aspiration biopsy (FNAB) was non-diagnostic and the mass was excised. The pathological examination revealed „angiomyolipoma".

KEYWORDS: angiomyolipoma, cutaneous angiomyolipoma, parotid mass

STRESZCZENIE:

Wprowadzenie: Naczyniakomięśniakotłuszczak jest łagodnym nowotworem, składającym się ze struktur naczyniowych, mięśni gładkich i tkanki tłuszczowej. Najczęściej pojawia się w nerkach, rzadziej w: wątrobie, płucach, sercu, jelitach, jamie nosowej, jamie ustnej i uchu. Naczyniakomięśniakotłuszczak skóry występuje bardzo rzadko i wykazuje pewne różnice w stosunku do postaci pozaskórnych, w tym: zazwyczaj lokalizuje się w dystalnych częściach kończyn w miejscu urazu, daje ujemny wynik w badaniu immunohistochemicznym z przeciwciałami HMB-45 i dotyczy częściej mężczyzn niż kobiet. We wszystkich przypadkach opisanych do tej pory, z jednym wyjątkiem, nie stwierdzano atypii komórkowej. W związku z tym uważa się, że naczyniakomięśniakotłuszczak skóry nie ulega transformacji złośliwej. Uważa się, że całkowite wycięcie masy jest właściwym leczeniem w postaci skórnej.

Opis przypadku: W opisywanym przez nas przypadku pacjentka zgłosiła się do naszej kliniki ze zmianą guzowatą lewej okolicy przedusznej, którą po badaniu przedmiotowym uznano za wywodzącą się ze śliniaki przyusznej. Po przeprowadzeniu badania MRI stwierdzono, że masa nie pochodzi ze ślinianki przyusznej. Po niediagnostycznym wyniku FNAB zdecydowano o resekcji zmiany. Badanie histopatologiczne wskazało rozpoznanie „angiomyolipoma".

SŁOWA KLUCZOWE: guz ślinianki przyusznej, naczyniakowłókniakotłuszczak, naczyniakowłókniakotłuszczak skóry

ABBREVIATIONS

FNAB – fine-needle aspiration biopsy
FNAC – fine-needle aspiration cytology
MRI – magnetic resonance imaging
WHO – World Health Organization

INTRODUCTION

Angiomyolipomas are benign mesenchimal tumors commonly found in the kidneys and more rarely in the lung, heart, intestines, nasal cavity, oral cavity and ear [1]. Cutaneous angiomyolipomas, on the other hand, are quite rare. These are located mostly in the distal parts of the extremities and develop due to trauma [2, 3]. In histological examinations cutaneous angiomyolipomas consist of vascular structures, smooth muscle and fatty tissue, as renal angiomyolipomas; however, renal angiomyolipomas are significantly positive for HMB-45 antibodies while cutaneous angiomyolipomas are negative. Renal angiomyolipomas are observed more frequently in women while cutaneous angiomyolipomas are more common in men. Association with tuberosclerosis, found in renal angiomyolipomas, is not observed in the cutaneous form [2, 4].

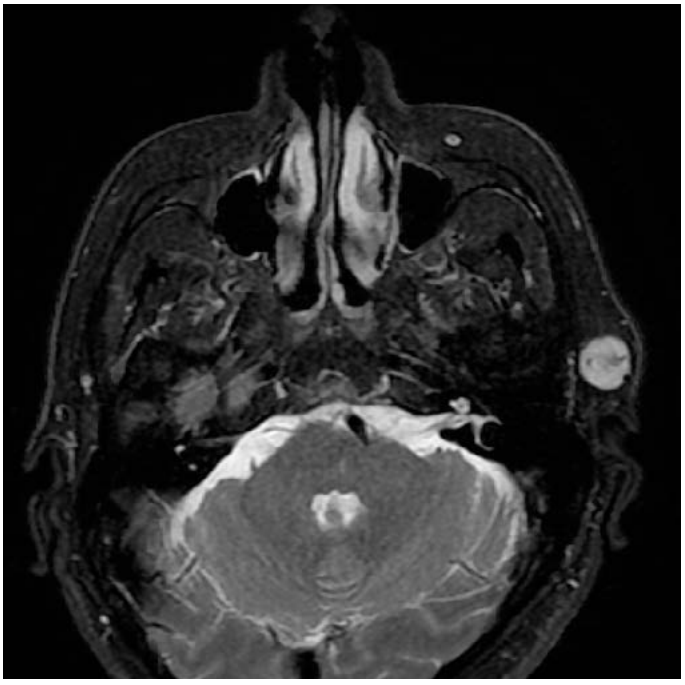

Fig. 1a. T1-weighted magnetic resonance imaging (MRI) images showing a diffusely hyperintense, well-circumscribed ovoid mass of 2.3 x 1.2 cm in size in the preauricular area that was not related to the superficial lobe of the parotid gland.

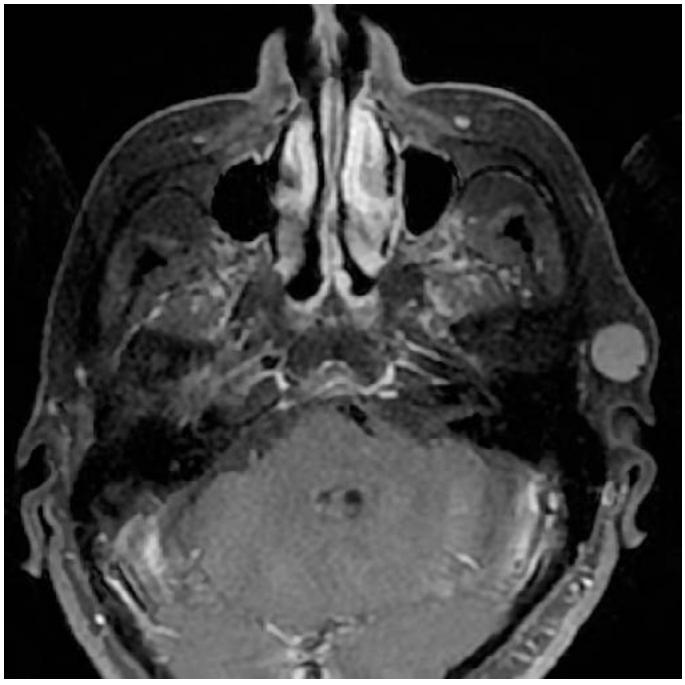

Fig. 1b. T2-weighted magnetic resonance imaging (MRI) images showing a diffusely hyperintense, well-circumscribed ovoid mass of 2.3 x 1.2 cm in size in the preauricular area that was not related to the superficial lobe of the parotid gland.

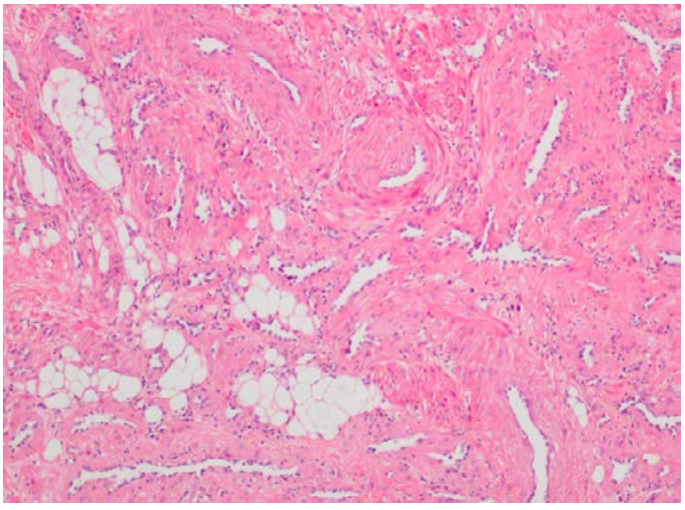

Fig. 2. Microphotography showing numerous vessels, spindle-shaped myoid cells and mature adipocytes (hematoxylin and eosin, original magnification, x100).

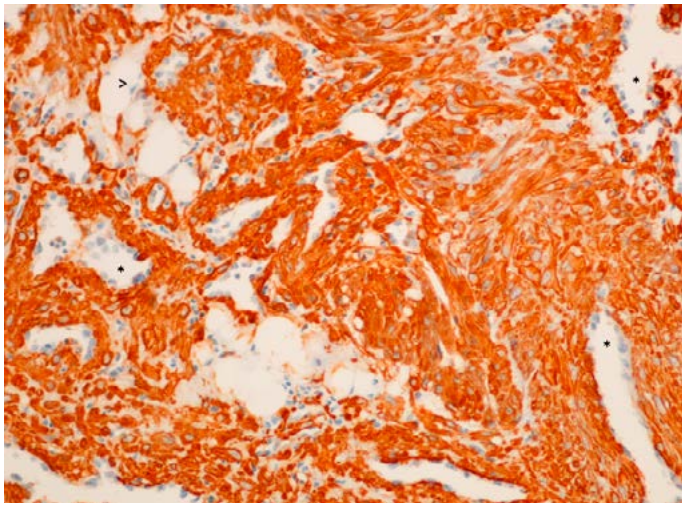

Fig. 3. Smooth muscle actin immunostaining of smooth muscle cells (original magnification, x200). Endothelial cells (asterisk) and adipocytes (arrowhead) showing no staining.

We present a case of a patient that applied to our clinic with a preauricular mass that was thought to be related to the parotid gland at first. X-ray showed that no relation between the mass and the gland. Total excision of the mass was performed, and the postoperative pathology report diagnosed it as „angiomyolipoma".

CASE REPORT

A 62-year-old woman presented to our clinic with a mass in the preauricular area that showed progressive growth for the last 6 months. Physical examination revealed a mobile mass, 2 x 1.5 cm in size, with medium stiffness, and a well-circumscribed margin. There was no other pathological finding. It was thought to originate from the parotid gland, and the initial diagnosis was pleomorphic adenoma, Warthin's tumor or periparotid lymphadenopathy. Neck MRI with contrast and US-guided FNAB were carried out. The T1- and T2-weighted MRI images showed a diffusely hyperintense, well-circumscribed ovoid mass, 2.3 x 1.2 cm in size, in the preauricular area, not related to the superficial lobe of the parotid gland (Fig. 1a.–1b.). FNAC that was performed twice resulted to be non-diagnostic (mixed inflammatory cells rich in leukocytes and hemosiderin pigment); thus, excision of the mass was decided on.

Left modified face-lift incision under general anesthesia was performed to reach the medium-stiff mass, which was dissected, freed

from surrounding tissue and excised. No complications were observed postoperatively.

The postoperative pathological examination with hematoxylin-eosin dye showed vascular structures, smooth muscle bundles and mature adipocyte tissue. Cellular pleomorphism and atypical mitosis were not observed. The HMB-45 antibodies were not found in the immunohistochemical examination (Fig. 2.–3.). During a 6-month follow-up there was no sign of local recurrence. The mass was diagnosed as „cutaneous angiomyolipoma" on the basis of histological and immunohistological findings. Written informed consent was obtained from the patient for this report.

DISCUSSION

Angiomyolipoma is a benign tumor consisting of vascular structures, smooth muscle and adipose tissue. It is most commonly found in the kidneys and more rarely in the liver, lung, heart, intestines, nasal cavity, oral cavity and ear [1]. This is the most common benign tumor of the kidneys, with its prevalence being 0.2–0.6%. This is found sporadically in 80% of the cases, but also observed frequently in tuberosclerosis complex which is a multisystemic disease inherited with an autosomal dominant pattern and characterized by benign tumors of multiple organ systems [5]. As much as 50–80% of tuberosclerosis patients are diagnosed with renal angiomyolipoma [6]. Cutaneous angiomyolipomas are generally reported on as case reports and observed to be located in acral areas. It is believed that this location is due to a trauma [2, 3], not associated with tuberosclerosis [4] Fitzpatrick et al. once reported a series of 8 patients with cutaneous angiomyolipomas all located in the acral areas, in 1990 [7].

Angiomyolipoma is a benign mesenchimal tumor consisting of adipocytes, smooth muscles that partially fasciculate, and thick-walled blood vessels. In order to diagnose a mass as angiomyolipoma, these three components must be present [2]. Although renal and cutaneous types seem histologically the same, they have some different features. HMB-45 antibody is highly positive in renal angiomyolipoma (95%) while it is generally negative in the cutaneous type. The renal form is predominantly seen in females while the cutaneous one in males. Association with tuberosclerosis is present in the renal form [2, 3, 8].

The 4th edition of the WHO Classification of Head and Neck Tumors from 2017 included angiomyolipoma in the group of „benign soft tissue tumors" and considered it as leiomyoma showing vascular differentiation. Besides, it also reported that less than 1% of leiomyomas were found in the head and neck region [9]. There are also cases of angiomyolipoma located in the head and neck region apart from the acral areas, such as nasal tip or ear. Shim Hs et al. in 2014 presented a case of cutaneous angiomyolipoma present in the forehead [10], and Kim Hj et al. in the glabella in 2017 [11]. It is thought that total excision of the mass is necessary in the treatment of cutaneous angiomyolipoma [4]. Buyukbabani et al. presented a case with double recurrence after incomplete excision [12]. Neither of the cases, except for one [13] presented until today, had cellular atypia; thus, cutaneous angiomyolipomas are considered not to lead to malign transformation [8].

Our case presented to our clinic with a left preauricular mass which was thought to be a parotid mass after an initial physical examination. After an MRI scan was performed, it was found that the mass did not originate from the parotid gland. After the FNAB turned out to be non-diagnostic, the mass was excised. The pathological examination revealed „angiomyolipoma". To sum up, cutaneous angiomyolipoma is a rare entity but may be seen in the head and neck region and can be misdiagnosed with a parotid mass or lymphoma; lesions in the skin tissue on the parotid can also be confused with the parotid mass and MRI imaging is helpful in diagnosis.

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