Intranodal angiolipoma

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ABSTRACT

تعد الأورام الشحمية الوعائية من الأورام المألوفة في مناطق الأنسجة الرخوة، ولكن يندر ظهورها في أماكن الجسم الأخرى. يصف هذا التقرير حالة نادرة لمريض رجل يبلغ من العمر 64 عاماً، وقد كان يشكو من تورم في المنطقة الأربية اليمنى. اتضح من الفحص المخبري بأن الورم عبارة عن ورم شحمي وعائي في العقدة اللمفية الأربية ومكون من خلايا شحمية وأوعية دموية. ومن هذا نستخلص بأن الورم الشحمي الوعائي يمكن إضافته ضمن الأمراض التي تسبب التورم المحدد الموضع في العقد اللمفاوية.

Angiolipomas are not uncommon tumors of the soft tissue, but are rarely found in other parts of the body. We report a case of intranodal angiolipoma in a 64-year-old man who presented with right inguinal swelling. Histopathological examination showed a tumor composed of mature adipose tissue and prominent vascular component, which is consistent with angiolipoma. We conclude that angiolipoma can be added to the list of conditions or diseases in the differential diagnosis of localized lymphadenopathy.

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Angiolipoma is a relatively common benign soft tissue tumor that occurs mostly in adults. It is rare in children and in patients older than 50 years.[1] The forearm is the most common site for angiolipomas, followed by the trunk, thigh, and upper arm. Multiple angiolipomas are more common than solitary tumors, and account for approximately two-thirds of the cases. Although angiolipomas are subcutaneous tumors, they are also described in other unusual sites such as the breast, gastrointestinal tract, thyroid, bone, spinal region, brain, knee joint, oral cavity, nasopharynx, parotid gland, heart, and lymph node.[2-9] There is only one report describing intranodal cellular angiolipoma in the literature.[2] The purpose of this paper is to report a rare case of intranodal angiolipoma in an adult male presenting as inguinal lymphadenopathy with a review of the literature.

Case Report. A 64-year-old man presented to the surgical clinic with swelling in the right inguinal region. On examination, a localized firm tender lump was found in the right inguinal area. The overlying skin was normal with no signs of inflammation or redness. The testicles and other external genitalia were normal. The other inguinal region showed no evidence of swelling. Laboratory investigation showed a total white cell count of $4.3x10^9$/l, hemoglobin count of 139 g/l, and platelets of $260x10^9$/l. The differential white cell count showed: neutrophils - 38.1%; lymphocytes - 39.8%; monocytes - 11.4%; eosinophils - 10.3%; and basophils - 0.4%. His serum biochemistry and abdominal ultrasound were normal. The patient was referred to the fine needle aspiration (FNA) clinic for diagnostic cytology. The FNA cytology of the lump showed fragments of fibrofatty tissue and aggregates of lymphocytes, and no definite diagnosis was made. However, the lump was later excised and sent for histopathological examination. The specimen consisted of 2 nodules measuring 1.5 cm and 0.7 cm surrounded by fibrofatty tissue. Microscopically, the nodules were lymph nodes, which were almost totally replaced by mature adipose tissue admixed with proliferating capillaries of variable size (**Figure 1**). These capillaries were surrounded by spindle cells (**Figure 2**). Some of the vascular channels were occluded with fibrin. Immunohistochemistry was carried out and showed that the spindle cells were positive for CD 34 (endothelial cell marker), and negative for smooth muscle actin and desmin (smooth muscle spindle cell markers). Angiolipoma was diagnosed based on the characteristic microscopic and immunohistological appearances of this mass. There was no evidence of

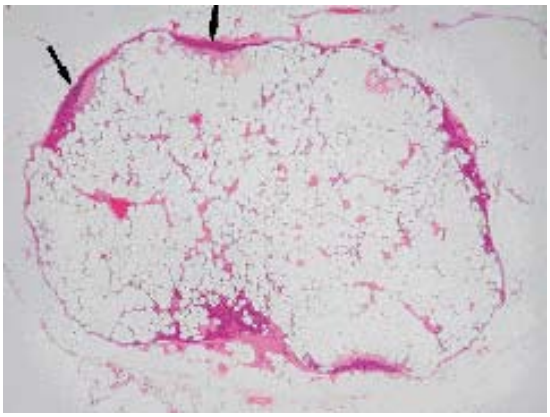

Figure 1 - Low power view of one of the lymph nodes showing almost complete replacement by the well defined angiolipoma. The arrows show residual lymphoid tissue of the node (Hematoxylin & Eosin stained slide x40).

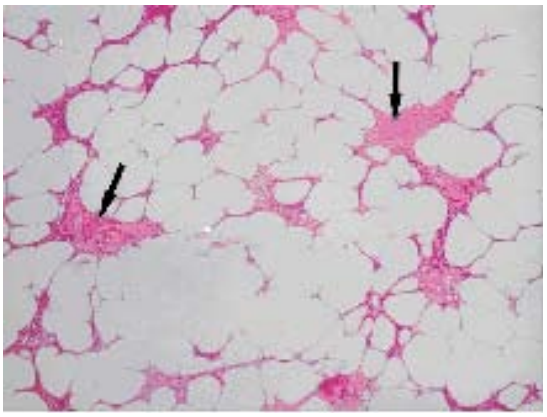

Figure 2 - Microscopic features of angiolipoma composed of mature adipose tissue and proliferated capillaries. Note the capillary lumen filled with fibrin thrombi (Hematoxylin & Eosin stained slide x200).

generalized lymphadenopathy, or similar soft tissue tumors elsewhere in the body.

Discussion. Angiolipomas are not uncommon soft tissue tumors usually found, as well-defined subcutaneous nodules with characteristic yellow cut surface. They usually occur in young adults and account for approximately 5-17% of all lipomatous tumors.[10] The age distribution of angiolipoma breast in the series of Kryvenko et al[3] was 25-80 years. Thyroid angiolipoma has been reported in women in their 70's.[7] The age of our male patient was 64 years. Angiolipoma is rare in other sites of the body, but has been described in other organs. The largest series (52 cases) of extra-soft tissue angiolipomas involving the female breast was described by Kryvenko and colleagues.[3] Gastrointestinal angiolipomas have been reported in the esophagus, stomach, duodenum, small bowel, colon, and rectum.[4] Although extremely rare, angiolipoma has also been described in the thyroid gland,[7] and as intraventricular cardiac angiolipoma[8]. Raghavendra et al[9] reported a case of spinal angiolipoma associated with coarctation of aorta and aortic hypoplasia. Angiolipoma has also been described in the head and neck including the oral cavity and upper lip. Nishimori et al[5] described a case of intra-articular angiolipoma of the knee. Whereas Yu et al[6] reported a case of intraosseous angiolipoma of the cranium. Intraosseous angiolipomas involving the mandible, and mandible and ribs have also been described. The first intranodal angiolipoma in the English literature was reported by Kazakov et al[2] who described focal solid proliferation of endothelial cells in their case. These features were patchy and less pronounced in our case. The mean size of angiolipoma varies according to the studied cases in the breast series was 7.00 ± 3.62 mm for cellular, and 19.61 ± 7.58 mm for low vascularity angiolipomas.

Angiolipomas may have variable clinical presentations according to their location in the gastrointestinal lesions included dysphagia, epigastric discomfort, weight loss, intestinal obstruction, gastrointestinal bleeding or anemia.[4] The thyroid lesion presented with a history of a nodular in the context of a multinodular goiter.[7] Lee et al[8] described recurrent attacks of left sided hemiparesis in the intraventricular tumor, while intracranial angiolipoma was presented in right parietal as swelling for 11 years[6]. Spinal angiolipoma in a 14-year-old patient presented with subarachnoid hemorrhage.[9] The patient in our study presented with gradual enlargement of a right inguinal lump that showed intranodal angiolipoma.

Microscopically, angiolipomas are composed of mature fat cells separated by a branching network of capillaries. The proportion of the fatty tissue to vascular elements varies in different cases. Moreover, the capillary proliferation is usually found in the subcapsular areas.[1] Likewise, the vascular channels in angiolipomas show fibrin thrombi, an important feature, which is not seen in the classical lipomas. These microscopic features are similar to those found in our case. Histologic differential diagnoses of angiolipoma include angioleiomyoma, and hemangioma with partial involution. Smooth muscle bundles are characteristically absent in angiolipoma and hemangioma, while angioleiomyoma is devoid of adipose tissue component. Based on the immunohistochemistry panel findings in our case, which showed CD 34 positive, SMA and desmin negative, the diagnosis of angiolipoma was confirmed. The differential diagnosis of intranodal angiolipoma includes ordinary lipomas in hypovascular lesions, where the presence of vascular microthrombi is in favor of angiolipoma.[1] Cellular

angiolipoma is sometimes difficult to differentiate from Kaposi's sarcoma, but the latter shows positive cellular immunoreactivity for human herpes virus (HHV)-8.[1] Treatment of angiolipoma depends on its localization and in general, a wide resection is usually curative.

In conclusion, angiolipoma is a slowly growing benign tumor commonly found in the soft tissue and rarely in other parts of the body. We report a case of intranodal angiolipoma as a rare cause of lymph node enlargement. Physicians and pathologists should be aware of this lesion, and angiolipoma can be added to the list of conditions or diseases in the differential diagnosis of localized lymphadenopathy.

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