Case Report

Multiocular Extraadrenal Myelolipoma in the Presacral Region Diagnosed by Fine Needle Aspiration: A Report of a Case

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A large tumor was incidentally identified by whole body CT scanning. It was formed by two components: a low-density area with wall partitions and a high-density area that was well-encapsulated. Fine-needle aspiration (FNA) was performed under colonoscopy for definitive diagnosis. Histological examination revealed that the tumor consisted of mature adipose tissues and hematopoietic marrow including all three myelopoietic cell lines. This observation indicated that the tumor was extraadrenal myelolipoma.

Myelolipoma is a benign tumor. If it is correctly diagnosed and show no symptoms, surgical resection will be unnecessary. Although multiocular extraadrenal myelolipomas is quite rare, and no cases had been reported until now, it is important that these diseases are considered and diagnosed pathologically before surgery is performed.

Key Words: extraadrenal myelolipoma, FNA, multiocular extraadrenal myelolipoma

Introduction

Extraadrenal myelolipoma is a rare benign tumor. Myelolipoma usually occurs within the adrenal gland, so that extraadrenal myelolipoma cases have accounted for 15% of all myelolipoma cases⁵. Reported extraadrenal myelolipomas were found in thoracic cavity, mediastinum, liver, stomach, retroperitoneum, lung, pelvis, and mesentery²⁻³. About 50% of extraadrenal myelolipomas were detected in the presacral region. Most extraadrenal myelolipomas were accidentally detected during medical examinations because they rarely present symptoms. On the other hand, sausage-like myelolipomas sometimes compress neighboring organs, and thus cause symptoms such as dyspnea and acute renal failure⁶⁻⁸. Generally, extraadrenal myelolipomas are uniformly single, well-circumscribed masses. Only a few bilateral extraadrenal myelolipomas have recently been reported⁹.

In this study, we reported the multiocular extraadrenal myelolipoma as the first case as far as we know. Although this tumor was initially considered liposarcoma by CT and MRI, it was correctly diagnosed by fine needle aspiration (FNA).

Case Report

A 69-year-old female had suffered from rheumatic arthritis for 25 years, and had been treated for hyperthyroidism and diabetes mellitus for several months. She had been taking prednisolone for rheumatic arthritis for a long time. In October
Fig. 1 A CT scan of the sacral space. The sausage-like tumor was formed by two components: a low-density mass that has partition walls and a high-density mass that was well-encapsulated. Although rectum was compressed by the tumor, no invasion to neighboring organs or lymph node swelling was observed.

2004, she felt chest pain and consulted another hospital. A large tumor and a dissecting aneurysm were pointed out by whole body CT scanning. The large tumor was found in the presacral region and was found to consist of two components: a low-density mass which had partition walls and a high-density mass which was well-encapsulated (Fig. 1). Subsequent MRI examination revealed that the tumor was $110 \times 74 \times 44$ mm (Fig. 2). On endoscopic ultrasonography (EUS), the tumor was well-demarcated (Fig. 3). Data from laboratory tests were within normal limits. No tumor or lymph node was detected by palpating abdominal region and digital examination of the rectum. Our case showed the typical imaging appearances of adrenal myelolipoma. However, extraadrenal myelolipoma is quite rare, and liposarcoma shows imaging appearances similar to myelolipoma. Therefore, we expected that the tumor would be liposarcoma.

After admission to our hospital, FNA was performed with a 22G needle under colonoscopy. No tumor invasion was observed in the rectal wall. For definitive diagnosis, we took 3 pieces from the tumor. A histological examination revealed that
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Fig. 4 A histological examination of the specimen obtained by FNA. The tumor consisted of mature adipose tissues and hematopoietic marrow including all three myelopoietic cell lines (H&E, ×400).

agement of adrenal stroma in response to curious injuries due to hormonal causes. The imaging, pathologic, and histological appearances of extraadrenal myelolipoma are identical to those of adrenal myelolipoma. While adrenal myelolipoma is often associated with other adrenal diseases including nonfunctioning adenoma, extraadrenal myelolipoma is not. However, Fowler et al. reported that extraadrenal myelolipoma might arise from groups of disjunctive hematopoietic stem cells.

Extraadrenal myelolipomas tend to arise in patients older than 40 and be accompanied by chronic, debilitating diseases or endocrinopathies. In reality, our case had contracted rheumatic arthritis, diabetes mellitus, and hyperthyroidism. She had been treated with prednisolone for a long time. So far, two cases with treatment of prednisolone were reported. According to these results, extraadrenal myelolipoma might be related to exogenous steroid treatment.

It is clinically important that extraadrenal myelolipoma be differentiated from other disease such as mass-forming extramedullary hematopoiesis, lipoma, liposarcoma, and myelolipoma. In our case, it was difficult to distinguish extraadrenal myelolipoma from well-differentiated liposarcoma by CT and MRI. In the end, FNA led to the final diagnosis. Romanowsky staining may facilitate the diagnosis. Otherwise, the Wright-stained imprint helps to establish the final diagnosis of this disease.

FNA was a convenient method to diagnose the tumor pathologically. However, it can introduce bacteria of the rectum into the tumor and blood flow, resulting that the patient would fall into septic status. To avoid this complication, CT-guided needle biopsy should be recommended if possible.

Myelolipoma is a benign tumor. If correctly diagnosed and no symptoms are seen, then a surgical excision is not necessary. In particular, because our case presented many complications, it was important that we should refrain from unnecessary sur-

Discussion

Although its etiology remains unclear, two theories have been proposed. Cina et al. reported that extraadrenal myelolipoma might originate in small foci of ectopic adrenal tissues, which may undergo
gery. Antonio et al, reported that the follow-up schedule for adrenal myelolipoma should include monitoring every 6-12 months or whenever symptoms arise. Therefore, we recommend annual follow-up for extraadrenal myeloma. Although multilocular extraadrenal myelolipoma is quite rare, and no cases has been reported to date, we must have this disease in mind and attempt to obtain pathological diagnosis prior to surgical intervention.

References

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