

《症例報告》

Angiomyolipoma-like lesion of the nasal cavity with abundant stromal mucin deposition

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Abstract : Abstract: In this paper, we report a rare tumor arising in the nasal cavity here.

Keywords : angiomyolipoma-like lesion, mucin, nasal cavity.

Introduction

In this article, we present a rare case of angiomyolipoma (AML)-like lesion arising in nasal cavity with an unusual morphology.

Case Report

A 69-year-old Japanese man noticed the tumor on the anterior portion of the nasal cavity several years ago. As the size of the tumor gradually increased, he visited Department of Otolaryngology, our hospital. On the suspicion of benign tumor, the tumor was surgically resected. Macroscopically, the tumor measuring 15x12mm was well-circumscribed and the cut surface of the tumor showed whitish color without necrosis or hemorrhage. Histologically, the tumor consisted of smooth muscle bundles closely adjacent to vascular structures and mature fat cells (Figure 1A, B). In the stroma, abundant mucin was identified and Alcian blue stain highlighted abundant mucin (Figure 1C). Immunohistochemically, smooth muscle cells were positive for alpha smooth muscle actin, but negative for Melanosome (HMB45), Melan A, S-100 protein and TFE3. Ki-67 index was less than 1%. Vascular endothelial cells showed the positivity for CD31 and CD34.

Discussion

The relationship between angioleiomyoma with adipose metaplasia and “so-called” mucocutaneous AML is debatable to date [1]. Actually, about sixteen cases of nasal cavity angioleiomyoma have been reported, and one case of vascular leiomyoma with abundant adipose cells has also been reported [2, 3]. However, it seems so difficult for pathologists to determine whether adipose tissue component shows neoplastic or metaplastic process. It is possible that these two concepts may show the sequential spectrum or be actually identical to each other. Accordingly, I made a conclusive diagnosis as AML-like lesion of the nasal cavity.

To the best of my knowledge, there are seven cases of nasal cavity “so-called” AML [4-10]. Clinicopathological features of these cases are summarized in Table 1. Most importantly, this disease is not associated with tuberous sclerosis clinically and has no expression for Melanosome (HMB45) immunohistochemically. Accordingly, “so-called” AML of nasal cavity and skin essentially differs from that of other anatomic sites including kidney and liver. Because of this reason, the term of “mucocutaneous” AML has been previously proposed. Pathologists should pay attention in

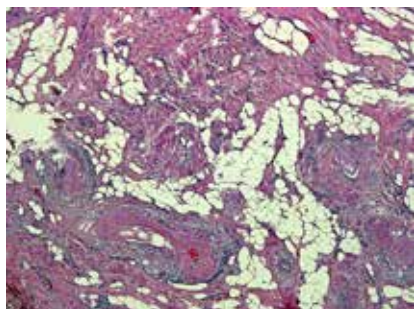


Figure 1A

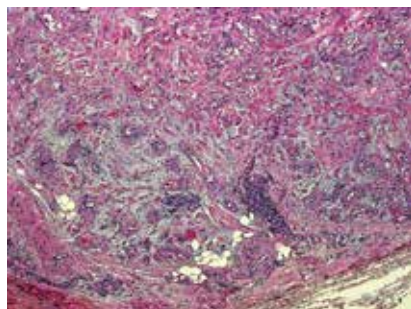


Figure 1B

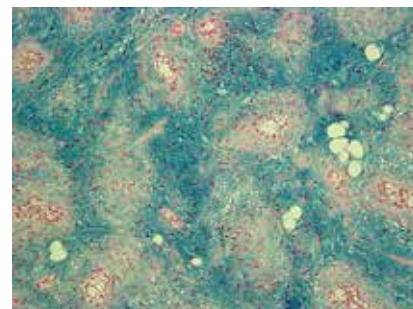


Figure 1C

Figure 1. Microscopic findings. (A) The lesion consists of smooth muscle cells, abortive vessels and mature fat cells. (B) The stroma contains abundant mucin. (C) Alcian blue stain highlights abundant stromal mucin deposition.

making a final diagnosis because perivascular epithelioid cell tumor (PEComa) can occur in nasal cavity [10]. However, I propose the usage of another diagnostic term such as AML-like lesion, instead of AML, until the relationship between mucocutaneous AML and angioleiomyoma with adipose metaplasia will be clarified in order not to cause the confusion between “true” AML suggesting PEComa and mucocutaneous AML. In order to clarify the difference between true AML and mucocutaneous AML in clinical, pathologic and molecular genetic aspects, further investigations in a large scale study will be desirable in the future. Additionally, I found abundant stromal mucin deposition in this tumor. This finding was never previously described in AML-like lesion. Hence, stromal mucin deposition should be added in the histologic spectrum of nasal cavity AML-like lesion, although myxoid change may be occasionally observed in angioleiomyoma.

The author declares no conflict of interest.

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Table 1. Summary of previously reported angiomyolipoma (-like lesion) of the nasal cavity

| Case | Age, Sex | TS | Size (mm) | Mucin | HMB45 | Follow-up (month) and outcome | Reporter |
|------|----------|----|-----------|-------|-------|-------------------------------|----------|
| 1 | 52, M | - | 40 | - | - | 12, AWOD | Dawlatly |
| 2 | 64, M | - | 20 | - | - | 24, AWOD | Gatalica |
| 3 | 66, M | - | 20 | - | - | 24, AWOD | Watanabe |
| 4 | 88, F | - | 20 | - | - | 6, AWOD | Watanabe |
| 5 | 45, M | - | 15 | - | - | 1, AWOD | Tardio |
| 6 | 52, M | - | 30 | - | - | ? | Erkiliç |
| 7 | 54, M | - | ? | - | - | ?, AWOD | Moreira |
| 8 | 69, M | - | 15 | + | - | 2, AWOD | Kuroda |

M, male; F, female; TS, tuberous sclerosis, AWOD, alive without disease.