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Case Report

CD10-positive malignant spindle cell tumor of the lip in a child: a malignant myoepithelioma?

Tadashi Terada

Departments of Pathology, Shizuoka City Shimizu Hospital, Shizuoka, Japan

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Abstract: A 9-year-old girl consulted to our hospital because of lower lip tumor. Excision of the tumor was performed. Histologically, the tumor consisted of cellular spindle and round cells with hyperchromatic nuclei and nucleoli. Mitotic figure and apoptotic bodies were scattered. Immunohistochemically, the tumor cells were strongly and diffusely positive for CD10 and vimentin. The tumor was focally positive for S100 protein, α-smooth muscle actin, PDGFRA, HER2/neu, p53, and CD68. The Ki-67 labeling was 20%. In contrast, the tumor cells were negative for pancytokeratins (AE1/3, CAM5.2, WSS, KL-1, HNF116), cytokeratin (CK) 5/6, CK34βE12, CK7, CK8, CK14, CK18, CK19, CK20, EMA, desmin, CD34, melanosome, KIT, p63, myoglobin, CD45, CD56, GFAP, D2-40, CEA and synaptophysin. The Histologies, positive p53 protein, and Ki-67 labeling of 20% suggested low grade malignancy. Although histological type was unclear, the author diagnosed this tumor as malignant myoepithelioma arising from the lip minor salivary gland because of positive reaction for S100 protein, α-smooth muscle actin, and CD10. The patient is now free from tumor 8 years after the first presentation.

Keywords: Malignant myoepithelioma, CD10, girl, lip, immunohistochemistry

Introduction

It is well known that CD10 is expressed in various normal cell types and various tumors including B-cell neoplasm, renal cell carcinoma, and hepatocellular carcinoma. In the breast, CD10 is expressed in myoepithelial cells and myoepithelioma [1]. In the salivary glands, normal myoepithelial cells do not express CD10 [2]. However, CD10 is expressed in epithelial-myoepithelial carcinoma [3]. Recently, a report described that CD10 is expressed in myoepithelioma, pleomorphic adenoma, and malignant myoepithelioma [2]. The author herein reports a case of probable malignant myoepithelioma of the lower lip in a 9-year-old girl.

Case report

A 9-year-old girl complained of a tumor of the lower lip and consulted to our hospital. Physical examination revealed an elevated tumor measuring 1 x 1 x 0.7 cm. An excision of the tumor was performed. Histologically, the tumor consisted of cellular spindle and round cells with hyperchromatic nuclei (**Figure 1A** and **1B**). Mitotic figure and apoptotic bodies were scattered (**Figure 1B**). Histochemically, Azan stain revealed that collagen fibers surrounded a group of tumor cells. Mucin stain (Alcian blue and PAS) showed no mucins.

An immunohistochemical study was performed with the use of Dako Envision method, as previously described [4-7]. Immunohistochemically, the tumor cells were strongly and diffusely positive for CD10 (**Figure 2A**) and vimentin (**Figure 2B**). The tumor was focally positive for S100 protein (**Figure 2C**), α-smooth muscle actin (ASMA) (**Figure 2D**), PDGFRA, HER2/neu, p53 (**Figure 2E**), and CD68. The Ki-67 labeling was 20% (**Figure 2F**). In contrast, the tumor cells were negative for pancytokeratins (AE1/3, CAM5.2, WSS, KL-1, HNF116), cytokeratin (CK) 5/6, CK34βE12, CK7, CK8, CK14, CK18, CK19, CK20, EMA, desmin, CD34, melanosome, KIT, p63, myoglobin, CD45, CD56, GFAP, D2-40, CEA and synaptophysin.

The histologies, positive p53 protein and Ki-67 labeling of 20% suggested low grade malignan-

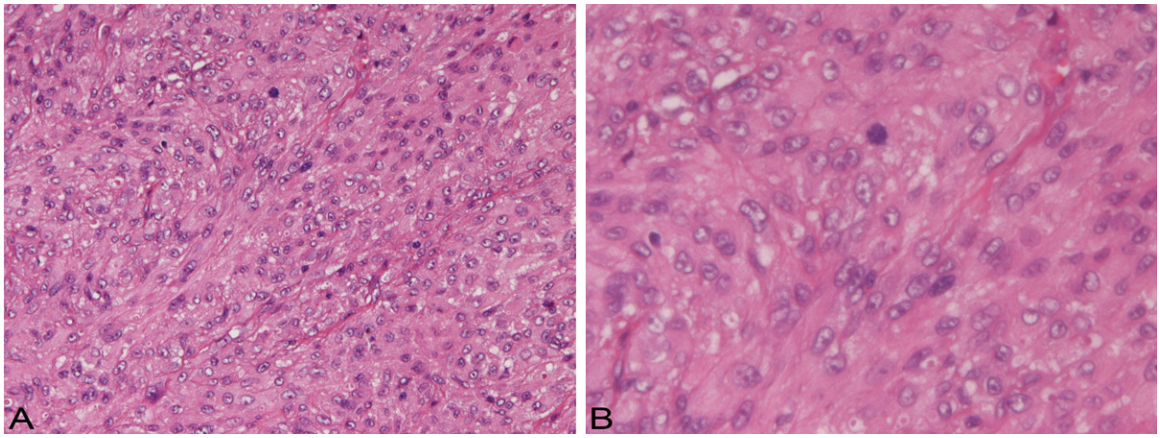

Figure 1. Histological features. A: Diffuse atypical spindle and round cell proliferation is seen in the lip. HE, x100. B: The atypical cells have hyperchromatic nuclei and nucleoli. Some mitotic figures and apoptotic cells are seen. HE, x200.

cy. Although histological type was not definite, the author diagnosed this tumor as malignant myoepithelioma arising from the lip minor salivary gland because of positive reaction for S100 protein, ASMA and CD10. The patient is now healthy and is free from tumor 8 years after the first presentation.

Discussion

The present tumor is histologically compatible with low grade malignant neoplasm. Immunoreactivity for p53, HER2/neu, and Ki-67 (20%) may support this suggestion.

The present tumor diffusely expressed vimentin but not any types of CK, suggesting that the present tumor is low grade sarcoma. However, the present tumor histologically and immunohistochemically does not fit into any types of sarcoma. Leibel and Moinfar [8] described mammary NOS-sarcoma with CD10 immunoreactivity may be a kind of malignant myoepithelioma. CD10, CK5/6, CK34βE12, CK14, p63, S100 protein, ASMA and GFAP are well known to be expressed in the myoepithelial cells or basal cells. The present tumor expressed S100 protein and ASMA, though other antigens were negative. These findings also suggest a myoepithelial differentiation of the present tumor. The negative reaction of cytokeratin in the present tumor does not exclude malignant myoepithelioma because there are a few reports of myoepithelial carcinoma of the salivary glands that are negative for cytokeratin [9-12].

The positive reaction of CD68 in the present tumor has not been described in the literature, but the CD68-positive cells may be tumor-associated macrophages. The positive reaction of PDGFRA in the present case has also not been reported in the literature, suggesting that malignant myoepithelioma may express this tyrosine receptor kinase.

Malignant myoepithelioma is rare. In addition, the present case arose in the lip. The present tumor appears derived from minor salivary gland of the lip. Furthermore, the present case is 9-year-old girl; most malignant myoepitheliomas develop in adults [9-12]. Therefore, the present case is very rare.

The prognosis of the present case was good; no recurrence was seen for 8 years. The biologic behaviors of malignant myoepithelioma are not known because it is very rare. Di Palma and Guzzo [9], who analyzed 10 cases of malignant myoepithelioma of salivary glands, reported that there are two pathways of the development of this tumor. One is de novo malignant myoepithelioma, and another is malignant myoepithelioma associated with pleomorphic adenoma. The de novo tumor is tend to be more aggressive with frequent recurrence and metastasis, while malignant myoepithelioma associated with pleomorphic adenoma is less aggressive and has long clinical history. The present tumor is de novo tumor because pleomorphic adenoma element was absent. The present tumor showed good clinical course. This may be because the present tumor is very small and also because the histology of the present tumor was low-grade.

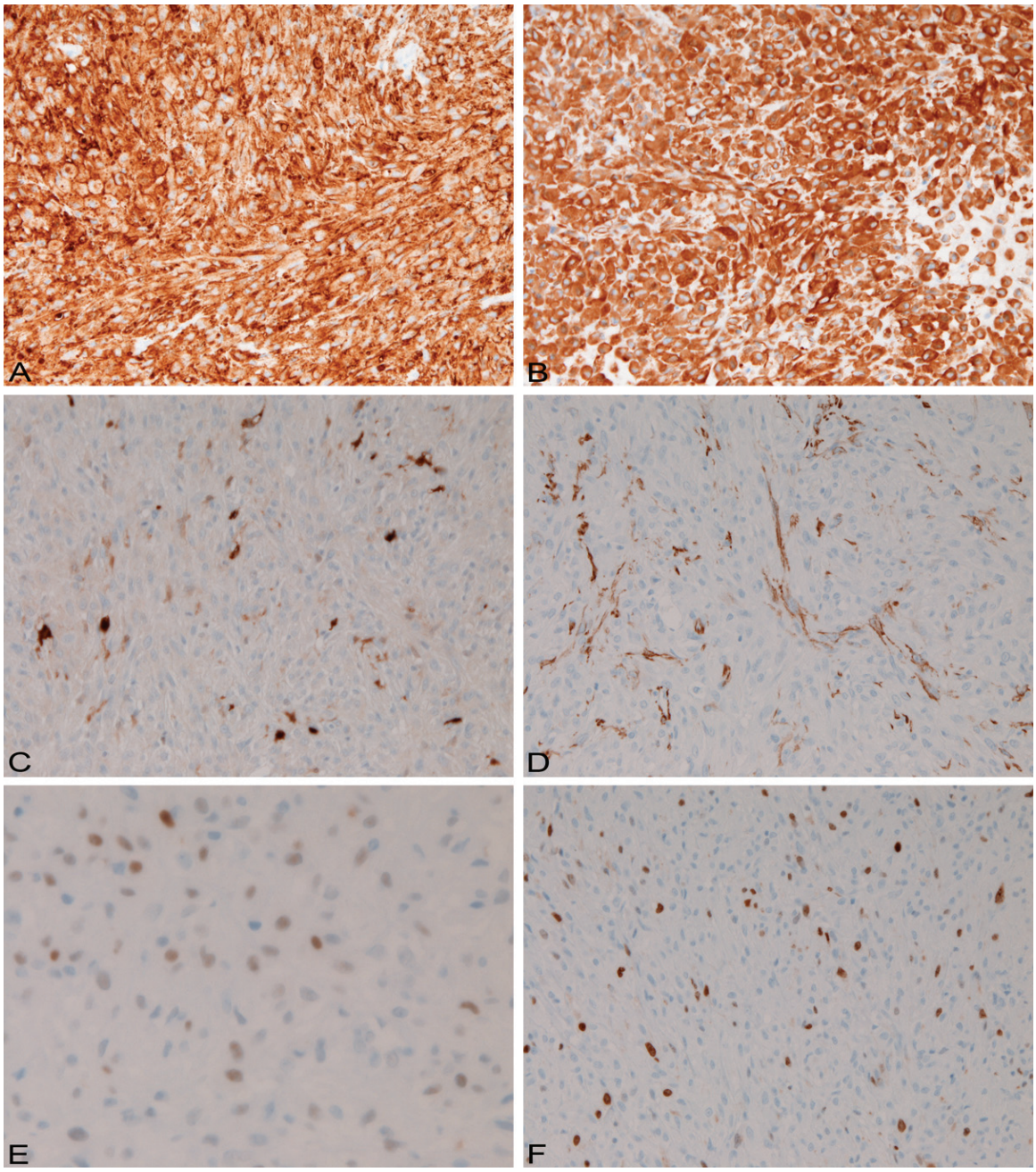

Figure 2. Immunohistochemical features. The tumor cells are diffusely positive for CD19 (A) and vimentin (B). The tumor cells are focally positive for S100 protein (C) and α-smooth muscle actin (D). p53 is positive (E), and Ki-67 labeling is 20% (F). Immunostaining, x200.

In summary, the author reported a case of CD10-positive malignant myoepithelioma or CD10-positive sarcomatoid tumor with differentiation into myoepithelial cells in a 9-year-old girl.

Conflict of interest statement

The author has no conflict of interest.

Address correspondence to: Dr. Tadashi Terada, Department of Pathology, Shizuoka City Shimizu Hospital, Miyakami 1231 Shimizu-Ku, Shizuoka 424-8636, Japan. Phone: +81-54-336-1111; Fax: +81-54-334-1173; E-mail: piyo0111jp@yahoo.co.jp

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