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Case report

Collision tumor of a malignant fibrous histiocytoma and a pleomorphic adenoma in the parotid gland: A case report

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ABSTRACT

We report a rare case of collision tumor comprising a pleomorphic adenoma and a sarcoma, pathologically diagnosed as a malignant histiocytoma, in the parotid gland.

A 57-year-old Japanese woman presented with rapid swelling of the left cheek in March 2002. After the malignancy was detected by aspiration biopsy cytology in the buccal region, it was resected via surgery. Based on histopathological and immunohistochemical examinations, the postoperative diagnosis was a collision tumor comprising a malignant histiocytoma and a pleomorphic adenoma. At pathological diagnosis of the surgical specimen, the tumor was enveloped in normal tissue. The tumor recurred and was treated with radiotherapy. However, it was persistent and subsequently metastasized to the lung. Chemotherapy was delivered, but the patient died from multiple organ failure in October 2002.

In the pathological finding of this sarcoma, histiocytic cells, giant cells, and multinucleated giant cells with severe atypism were regularly distributed, with thin epithelium and fibrous composition. This finding led to a malignant fibrous histiocytoma subtype diagnosis. However, this sarcoma was neither clinically nor histopathologically a typical malignant fibrous histiocytoma. Therefore, we suggest that it was either an undifferentiated high-grade pleomorphic sarcoma or a carcinosarcoma with osteoclast-like giant cells.

1. Introduction

Collision tumors, tumors with different histological types that are present in one lesion and share a boundary or infiltrate, are unusual. Collision tumors are especially uncommon in the oral and maxillofacial cavity [1,2]. We treated a collision tumor comprising a pleomorphic adenoma and an undifferentiated malignant tumor that occurred in the parotid gland.

Malignant fibrous histiocytomas, which were reclassified by the World Health Organization in 2002, appear most commonly in the limbs and retroperitoneum, but rarely in the salivary glands.

Here, we report a case of collision tumor comprising an undifferentiated sarcoma and a pleomorphic adenoma, with some discussion. To our knowledge, this is the first report of a tumor of this type.

2. Case report

A 57-year-old Japanese woman visited the Tokyo Dental College at Chiba Hospital with swelling of the left cheek in March 2002. The swelling of the left cheek was previously noted by an otolaryngologist in 1982, but she received no treatment. The swelling suddenly increased in February 2002 and she consulted a dentist who referred her to our department.

Swelling with elasticity and an indistinct border was found in her left cheek. The surface of her cheek was hot and tender, but a neurological disorder of the facial nerve was not observed (Fig. 1). However, she showed symptoms of trismus with an inter-incisor distance of 23 mm.

A computed tomography scan showed a mass in the left side parotid region and a distinct border surrounding the tissue. The inside of the tumor exhibited heterogeneous low density and fluid repletion was suspected (Fig. 2). It had not metastasized to the cervical lymph nodes.

Anemia was the only disease found on clinical examination.

A parotid gland tumor was suspected and thus aspiration biopsy cytology of the left cheek was performed. A malignancy was detected, but its histologic type was not able to be clearly identified. Biopsy was impossible because the tumor had a cyst-like

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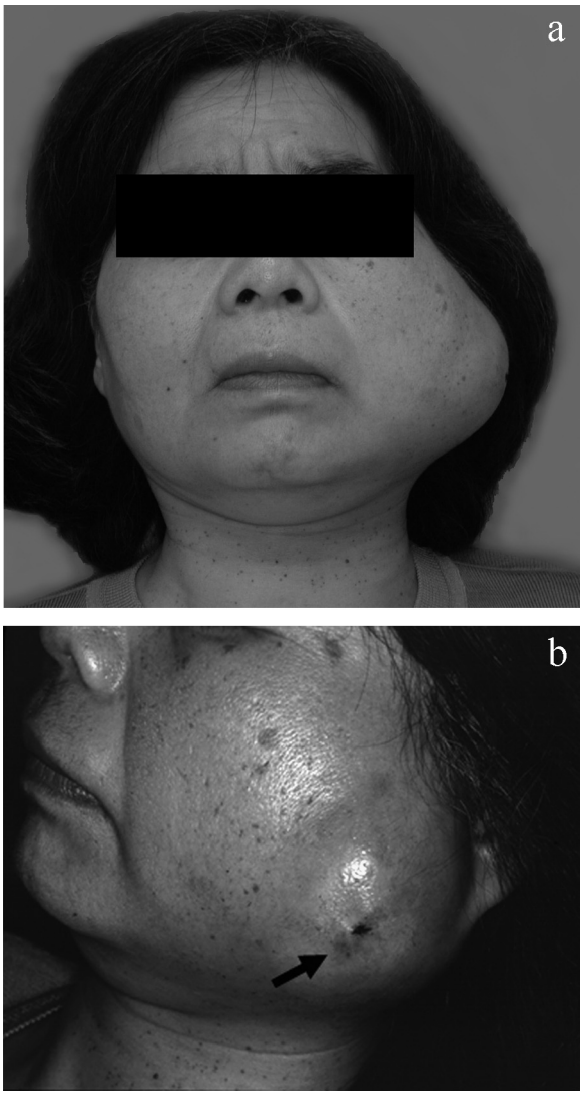

Fig. 1. (a) A facial photograph taken during initial diagnosis. The cheek was swollen to the size of a fist. (b) The arrow shows the puncture site for aspiration biopsy cytology.

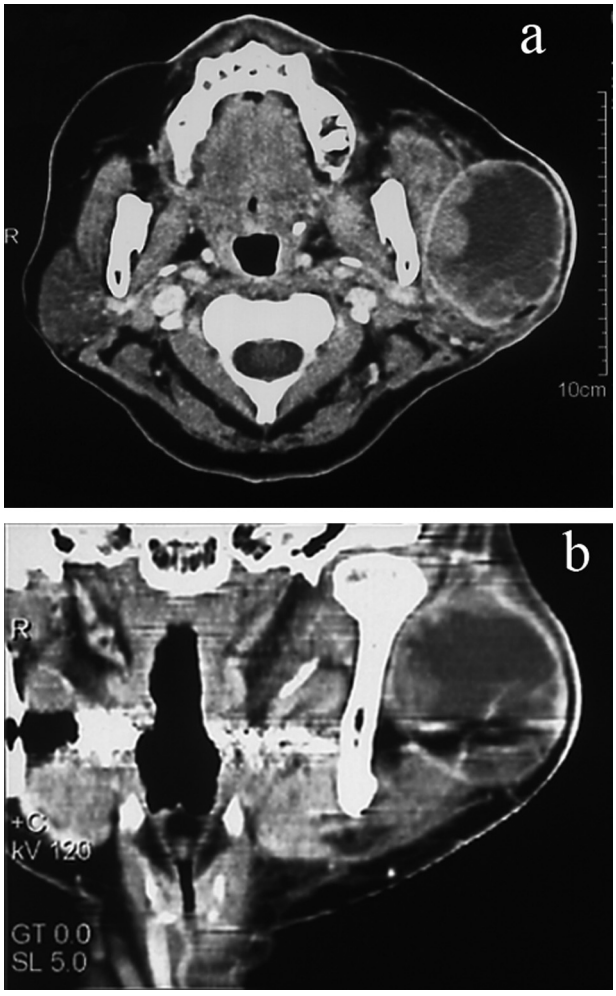

Fig. 2. Preoperative computed tomography images. A tumor mass with distinct boundaries was found in the left parotid gland region. The inside of the tumor was nonuniform and had low density. (a) Axial section and (b) coronal section.

lesion filled with blood or effusion fluid. Based on these results, a malignant salivary gland tumor originating from the parotid gland was diagnosed, and surgical removal was planned.

Tumor resection was performed in mid-April 2002 under general anesthesia. An incision was made along the submandible to the front of the pinna and the tumor including the parotid superficial layer was resected. The tumor was enveloped in a capsule. Results of intraoperative frozen section examination showed no residual tumor in the surrounding area. The mass was 95 mm × 83 mm × 50 mm in size. It was encased by a thick cyst wall and contained a bloody effusion (Fig. 3).

In sections stained with hematoxylin and eosin, spindle-shaped and plasmacytoid cells, duct-like structures and cartilage-like tissue were seen. The tumor was therefore diagnosed as pleomorphic adenoma (Fig. 4). However, part of the tumor had atypical histiocyte-like cells, giant cells, and multinucleated giant cells regularly distributed throughout the tissue, and had few fibrous components (Fig. 5). Furthermore, the tumor was divided with connective tissue and appeared to have distinct sides with different components (Figs. 6 and 7). Therefore, this tumor was diagnosed as a collision tumor comprising a salivary gland tumor and a malignant tumor.

The malignant tumor comprised histiocytic cells, giant-cell-like cells, and multinucleated giant-cell-like cells with atypism in both specimens seen on hematoxylin–eosin staining. It was suspected to be undifferentiated cancer, malignant myoepithelioma, or MFH. However, it was not typical of any tumor type because it did not have an epithelial or fibrous composition. Immunostaining indicated that the tumor was positive for α1-antichymotrypsin (Fig. 8) and negative for keratin and vimentin (Table 1). As a result, it was diagnosed as a malignant histiocytoma, a subtype of MFH, by several pathology specialists.

In mid-May 2002, the patient's left cheek again exhibited swelling, and magnetic resonance imaging scans showed recurrence of the tumor to be the cause.

Because aspiration biopsy cytology indicated malignancy, we resected the tumor to the deep lobe of the parotid gland. The result of histopathological examination of the recurrent neoplasm was similar to that of the primary sarcoma.

Radiotherapy was started in the beginning of June 2002 because the tumor had another recurrence in the lower portion of the pinna behind the resection field. The size of the tumor was reduced by radiation (57.6 Gy), but metastasis was detected in the left internal