Benign Mesenchymoma of the Cheek: A case with immunohistochemical study

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A case of benign mesenchymoma that occurred in the cheek is presented. The tumor consisted of an admixture of such mesenchymal elements as mature fat tissue, blood vessels of varying size and shape with less organized muscular wall, fascicles of smooth muscle cells, and bone. Immunohistochemical examination revealed that many smooth muscle cells comprising the tumor were positive for desmin and α -smooth muscle actin but negative for vimentin. Such an immunohistochemical characteristic is very similar to that of non-vascular smooth muscle cells.

Key words: benign mesenchymoma, cheek, immunohistochemistry

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Introduction

Benign mesenchymomas most frequently involve the kidney and perirenal tissue. Microscopically the tumor is composed of a haphazard mixture of mature fat, fibrous tissue, and tangled blood vessels through which are scattered nests or masses of smooth muscle cells. Occasionally it appears to contain islands of cartilage, bone, and lymphoid tissue as well as other mesenchymal elements (1).

In the head and neck, benign mesenchymoma is a rare lesion. Our review of the British and American literature revealed only two cases reportedly occurring in the cheek (2). An additional case of benign mesenchymoma of the cheek is presented here. In addition, an immunohistochemical study was undertaken to facilitate recognition of the lesion.

Case Report

A 44-year-old man was seen with a painless soft-tissue mass in the right cheek (Fig. 1). This mass was noted by his mother to have been present since infancy. The mass had gradually increased in size by the time he reached the age of 33, but he was without any other symptoms.

Clinical examination revealed a soft-tissue mass, 3 cm in diameter, in his right cheek. In addition, a second mass, 1.5 cm in diameter, was also palpable just in front of the first one. Both were covered with a smooth, intact skin surface exhibiting normal color. There was no adhesion to the overlying skin. Radiographs of the facial bones showed no remarkable changes. A small amount

Fig. 1: A 44-year-old man shown with a painless soft tissue mass in the right cheek (arrowheads).

of bloody fluid could be aspirated from the masses. Tentative diagnosis of hemangioma was made, and then the tumors were excised intraorally under general anesthesia.

At surgery, the masses deep in the right buccal mucosa were well circumscribed from the surrounding

soft tissue but not encapsulated. The boundary between the masses was undefined. Total removal was difficult since a part of the posterior mass extended into the masseter muscle. The clinical impression of the surgeon was that the lesion was an angioliipoma. A follow-up examination conducted 2 years later revealed a residual mass that gradually had increased in size.

Pathological Findings

On gross examination the tumor was composed of two masses that had broad contact and were not separable. The anterior mass, 1.5 cm in diameter, was soft,

Fig. 2: Gross appearance of the tumor. The tumor is composed of two masses. The anterior mass (arrowheads) is soft, yellowish, and lobulated; whereas the posterior one (arrows) is relatively firm and exhibited a mottled appearance.

yellowish, and lobulated; whereas the posterior one, 3.0cm in diameter, was relatively firm and exhibited a mottled appearance (Fig. 2). Microscopically both masses were composed of an admixture of mature mesenchymal elements with fibrous connective tissue stroma, prominent among which were the fat tissue and blood vessels. No cellular atypism was noted (Figs. 3a, b, and c). In the posterior mass, the large vessels appeared to be distorted and the smooth muscle cells of their wall were less organized and often blended in a random fashion with the surrounding fibro-fatty tissue. In addition, a scattering of fascicles of smooth muscle cells without an apparent perivascular arrangement were seen. These mesenchymal components were arranged and intermixed in a haphazard fashion. Foci of lymphoid tissues and osseous tissues were seen in the posterior mass (Figs. 3b and c). On the other hand, the anterior small mass almost exclusively consisted of fat tissue and blood vessels.

Immunohistochemical staining was performed on formalin-fixed paraffin-embedded tissue sections by a streptavidin-biotin staining technique (DAKO CORP., Carpinteria, CA). The series of primary antibodies and their working dilution used in this study are listed in Table 1. A positive reaction for vimentin was observed in most of the cells comprising the tumor, i.e., in fat cells, fibroblasts, endothelial cells, and smooth muscle cells of small- to medium-sized blood vessels (Fig. 4a). The smooth muscle cells were consistently stained with α -SMA antibody and weakly and focally stained with desmin antibody (Figs. 4b and c). However, the smooth muscle cells present in the less organized wall of large blood vessels and those without apparent perivascular

Fig. 3: **a)** Anterior small mass. The mass consists of mature fat cells that are lobulated by thin fibrous septa containing small to medium-sized vessels. **b)** Posterior mass. The mass is composed of an admixture of fibrous connective tissue, blood vessels exhibiting muscular walls, mature fat tissue, and fascicles of smooth muscle. Foci of osseous tissue are noted (arrow). **c)** Posterior mass. The blood vessels are distorted and exhibit less organized smooth muscle cells in the wall. Some of the smooth muscle cells are blended in a random fashion with the surrounding fibro-fatty tissue (H-E, a, $\times 20$; b, $\times 20$; c, $\times 65$).

Table 1: List of primary antibodies used in this study

Antibody against	Type	Dilution	Source
Cytokeratin	polyclonal	1:200	DAKO, Carpinteria, CA, U.S.A.
Vimentin	monoclonal	prediluted	DAKO
Desmin	monoclonal	prediluted	DAKO
α -smooth muscle actin (α -SMA)	monoclonal	1:50	DAKOPATTS, Glostrup, Denmark
Neuron-specific enolase	polyclonal	1:150	DAKO

Fig. 4: Immunohistochemical staining. **a)** Most of the tumor cells, except for less organized vascular smooth muscle cells, show immunoreactivity for vimentin. **b)** Almost all smooth muscle cells are positive for α -smooth muscle actin. **c)** The cytoplasm of the less organized vascular smooth muscle cells is positive for desmin, whereas smooth muscle cells of normal-appearing vessels is negative (a,b, and c, $\times 65$).

arrangement were vimentin-negative, although both reacted intensely with both desmin and α -SMA antibodies (Figs. 4a-c). There was no positive staining of tumor cells for cytokeratin and neuron-specific enolase.

Discussion

The pathological nature of benign mesenchymoma is still full of controversy, as the lesion is construed by some to be a hamartoma and by others as a true neoplasm (2). The majority of mesenchymomas occur in newborn infants or younger patients and occasionally they are associated with developmental anomalies. This has led some pathologists to believe that the mesenchymoma is a type of hamartoma. On the other hand, there is enough reason for supporting the hypothesis of its neoplastic nature. The mesenchymoma sometimes arises and/or continues to enlarge in the

patients after puberty and occasionally appears to contain mesenchymal elements that would be regarded as uncontested neoplasm such as hemangiopericytoma and leiomyoma. In addition, there are malignant counterparts of the benign mesenchymoma. In the present case, the tumor continued to enlarge after normal development had ceased and exhibited an area regarded as an angiolipoma. In addition, a follow-up examination revealed a residual mass that gradually increased in size. These findings would be support our conclusion that our case is a true neoplasm.

During recent years, several cytoskeletal and contractile elements have been shown to be reliable differentiation markers and have been extensively used for tumor identification as well as for studying normal development (3,4). Among the most widely used markers of this type are intermediate filament proteins

and actin isoforms. Desmin is the intermediate filament protein characteristic of the skeletal muscle, cardiac muscle, visceral smooth muscle, and smooth muscle of some blood vessels (4). Alpha-SMA is the actin isoform typical of smooth muscle cells and is prevalent in vascular muscle cells (5). Originally thought to be the only intermediate filament type characteristically present in nonmuscular mesenchymal cells, vimentin has been found in certain smooth muscle cells, e.g., smooth muscle cells of the vascular system, as well (6-8). It has been reported that the majority of the vascular smooth muscle cells are vimentin-positive and desmin-negative or positive for both vimentin and desmin, and that a very small fraction is desmin-positive and vimentin-negative (6-8). The presently studied tumor appeared to contain many smooth muscle cells that were positive for desmin and α -SMA but negative for vimentin. Such an immunohistochemical characteristic is very similar to that of non-vascular smooth muscle cells. For consideration of the pathogenesis of benign mesenchymoma, however, further knowledge should be obtained concerning the immunohistochemical heterogeneity of smooth muscle cells.

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