Collision tumor of the palate: A rare case report

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Abstract
This case report presents an unusual swelling of the palate in a 61-year-old male patient. Histopathologically, it revealed features of two separate adjacent lesions, a spindle cells lesion showing diffuse immune-positivity for S-100 protein and focal positivity to glial fibrillary acid protein and an osseous lesion with numerous trabaculae of bone, adipocytes and myxoid tissue confirming the diagnosis of collision tumor of benign peripheral nerve sheath tumor and osteoma. Extensive search of English literature shows no reported cases of peripheral nerve sheath tumor with osteoma. The probable histogenesis of this collision tumor is discussed in detail. The purpose of this case report is to document this rare case in the literature so as to increase the awareness of this entity.

Keywords: Collision, intra-oral, palate, peripheral nerve sheath tumor, S-100

Introduction
Collision and hybrid tumors represent the occurrence of two or more distinct synchronous primary tumors, benign or malignant, appearing in the same anatomic region. Hybrid tumors are composed of two or more different tumoral entities in a single neoplasm that arise within a definite topographical region,[1] whereas collision tumors are lesions that originate in different regions but coalesce in a particular area.[2] We report a rare case of swelling of the palate in a 61-year-old male patient, which was characterized histopathologically by the presence of two different components. Peripheral lesion shows spindle cell tumor positive for S-100 and glial fibrillary acid protein (GFAP) and a distal tumor consists of trabaculae of bone encompassing proliferating adipocytes and myxoid tissue. To the best of our knowledge, collision benign tumors of the palate has never been reported though few cases of hybrid salivary gland carcinomas of the palate have been infrequently seen in the medical literature.[3] The majority of these tumors represent between carcinomas and sarcomas or lymphomas and rarely between two types of carcinomas. Benign peripheral nerve sheath tumor (BPNST) and osteoma are clearly different in terms of histopathological and immunohistochemical examination.

Case Report
The present case report is about a 61-year-old male patient who presented with a growth on the palate, which was present since many years. It was slowly growing and intermittent in nature. Intra-oral physical examination revealed a non-tender, non-mobile, oval, nodular mass on the middle of the palate. The lesion measuring about 2 × 1.5 × 1.5 cm, was well circumscribed and pedunculated [Figure 1a]. The lesion had a smooth intact surface and was firm to hard in consistency. No cervical lymph nodes were palpable. Medical history was unremarkable and hematologic as well as biochemical parameters were within the normal limits. Family history and personal history were not contributory.

Based on the above findings the lesion was provisionally diagnosed as fibroma and differential diagnosis of peripheral osteoma, fibrosed pyogenic granuloma, BPNST was given. The lesion was excised with the patient consent under local anesthesia and was fixed in 10% neutral buffered formalin. Grossly, the dome shaped specimen appeared tan and cut surface has yellowish hue [Figure 1b].

Microscopically, the lesion was well-circumscribed and revealed lobules of adipose tissue and calcified structures separated from the surface epithelium by fibro-cellular connective tissue stroma [Figures 2a and b]. Mass of moderately cellular spindle cells, bound on the deeper aspect by fibrous tissue were evident [Figure 2c]. Histopathologically, these calcified structures revealed the presence of mature bone with entrapped osteocytes in lacunae. Abundant myxoid tissue with a small collection of lipid laden macrophages were also seen [Figure 2d]. Immunohistochemical analysis revealed...
S-100 and GFAP positivity of the spindle cells while actin was positive around the blood vessels but negative for spindle cells [Figures 3a-d]. The lesional tissue was also negative for desmin, CD-57, human melanoma black-45. Mitotic figures and necrosis were not present. Based on the histopathological and immunohistochemical findings final diagnosis of collision tumor of BPNST with osteoma was made. Treatment involved surgical excision of the lesion [Figures 4 and 5]. Patient was followed up for 2 years and showed no signs of recurrence.

**Discussion**

A collision tumor composed of two different types of neoplasms derived from clearly different origins of two neoplastic clones that have arisen from different cell types in close proximity to each other.\(^4\)
The present collision tumor revealed a unique combination of both peripheral osteoma and BPNST components within the single tumor. Microscopic evaluation of our case reveals a peripheral tumor divided into parts subepithelially below the capsule the stroma was highly cellular made up of fascicles of spindle cells and distal lesion was composed of trabeculae of bone with fatty fibromyxoid stroma. Immunohistostaining reveals positivity for S-100, vimentin and glial fibrillary acidic protein of these spindle cells supports the presence of Schwann cell which are neuroectodermal in origin, confirming the diagnosis of peripheral nerve sheath tumor. The presence of osteoma confirms the mesenchymal origin. Therefore, this tumor is made up of two tissue of different origin suggesting it as collision tumor. It raises an interesting question regarding the nature and pathogenesis of this entity and their relationship.

Though uncommon, the presence of peripheral nerve sheath tumor with other lesions such as basal cell carcinoma (BCC), perineuroma, plexiform neurofibroma and melanoma have been reported. The combinations more frequently described are peripheral nerve sheath tumors (PNST) and perineuroma and BCC and neurofibroma. Peripheral nerve sheath tumors are characterized by neoplastic proliferations with Schwann cell differentiation. Schwannomas are benign neoplasms of Schwann cell origin. Our case represent cellular schwannoma where the gross appearance is characteristic, in the form of well-circumscribed masses with degenerative changes and variable admixture of compact spindled areas and hypocellular, microcystic. A well-formed collagenous capsule is a consistent finding, as well as hyalinized vessels. By immunohistochemistry, schwannomas typically show diffuse, strong expression of S-100 protein and abundant pericellular collagen type IV, consistent with the presence of a continuous pericellular basal lamina. GFAP is expressed in a subset of schwannomas. Cellular schwannoma is defined as a schwannoma composed almost entirely of a compact, fascicular proliferation of well-differentiated, cytologically bland Schwann cells, lacking Verocay bodies.

Clinically, the peripheral osteoma of the jaw bones are slow growing and appear as unilateral, pedunculated mushroom-like masses which are usually asymptomatic. The peripheral osteoma is a tumor of mature bone with two different types, the compact or “ivory” osteoma, usually has a sessile base, normal-appearing dense bone with minimal marrow spaces and occasional haversian canals and the cancellous osteoma is usually pedunculated and resembles the bone of origin. It will contain trabeculae of bone and fibrofatty marrow with osteoblasts. The surface can be irregular or smooth, with cortical bone at the margin. Histopathological the present case has features of cancellous osteoma as it was pedunculated and composed of trabeculae of bone lined by osteoblasts with fibrofatty marrow.

Moreover, BPNSTs are rarely altered by an admixture of lipocytes, bone and myxoid tissue. Though lipocytic and osteoid alteration of schwannoma has been reported in few cases with both osteoid and lipocytes being an integral part of the tumor, but in this case the osteoid and fat cells were clearly demarcated from the spindle cells suggesting the presence of two tumor entities.

A rare tumor with metaplastic bone formation that may occasionally mimic PNST is the ossifying fibromyxoid tumor (OFT) of soft parts. This rare neoplasm usually arises in superficial locations and is characterized by bland cytology, fibromyxoid stroma, peripheral ossification in the form of a shell and frequent S-100 expression. A peculiar finding in OFTs is the presence of an incomplete shell of metaplastic lamellar bone located just beneath the fibrous pseudocapsule. In some cases, extensive bone formation can even be observed in central areas of the tumor. In our case, though bone trabeculae presenting variable degrees of mineralization were more prominent but they were present adjacent to the highly cellular CT stroma and complete rim of bone enclosing a fibrofatty stroma was evident deeper but not on the periphery of the spindle cell tumor. Therefore the diagnosis of OFT can be excluded.

Jones et al. believe that if the light-microscopic features of a given soft-tissue neoplasm reveal an unencapsulated tumor composed of two or more mature mesenchymal tissues, with no one mesenchymal tissue predominating at the expense of the other tissues, the neoplasm should be diagnosed as a benign mesenchymoma. However, the reported lesion was partially encapsuled and a clear demarcation could be seen between the fascicles of spindle cells and bony trabeculae, which were intermingled with adipocytes, myxoid tissue and foamy histocytes. Therefore, this case does not favors the diagnosis of benign mesenchymoma.

The histopathological features of the reported lesion include the presence of neuroectodermal (S-100 and GFAP positive Schwann cells) and mesenchymal (osteoid, adipocytes and myxoid). It can be debated that, several features of the tumor described in this report suggest that it likely originated from the pleuripotent neural crest cells hence could be referred as ectomesenchymoma. The term “ectomesenchymoma” was coined for tumors consisting of ectodermal components (usually neuronal cells and S-100 positive spindle cells) in combination with embryonal rhabdomyosarcoma. Thorough review of the literature reveals that ectomesenchymoma is usually malignant and one of the major component is the presence of skeletal or smooth muscle tumor, but our case was negative for both smooth muscle actin and desmin.
The present case was followed-up for recurrence free period of 2 years. Therefore, surgical excision appears to be the treatment of choice. In summary, a group of several unusual neoplasms composed of a mixture of tissues, appear in both benign and malignant forms. Most of these arose in the soft parts. The neoplasm, in the present case was a peripheral lesion on the hard palate. The relative rarity of such neoplasm in intra-oral location demands proper investigation so as to understand its nature and to characterize this rare type.

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