CASE REPORT

Vascular changes in hard palate sialolipoma: Sialoangiolipoma or vascular malformation?

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ABSTRACT

Palate sialolipomas are rare. Less than 10 cases located in the hard palate are reported to our knowledge. We report a case of hard palate sialolipoma, peculiar by the intratumor vascular patterns. A 67-year-old man presented with a 1.5 cm lesion of the oral hard palate. The lesion was surgically resected. On microscopy, the lesion, partly encapsulated, consisted of a proliferation of mature adipocytes containing normal minor salivary gland tissue and branching intratumoral vessels of varied size with irregularly thickened wall and papillary projections or tufts. The microscopic features of the tumor we report suggest that vascular malformation-like patterns may occur in sialolipomas of the hard palate. This morphological vascular peculiarity should be acknowledged since it may represent source of hemorrhage.

Key words: Angioma, sialolipoma, palate, vascular malformation

INTRODUCTION

Palatal sialolipomas are rare and were identified as a definite entity in 2001.[1] To our knowledge, less than 10 cases from the hard palate are reported.[1-6] Here, we report a case of hard palate sialolipoma, peculiar by the intratumor malformation-type vascular patterns.

CASE REPORT

A 67-year-old man presented with a median mass of the oral hard palate, noticed since several years. There was no facial malformation. The patient was hypertensive, treated by losartan. The mass was resected under local anesthesia (outpatient). The resected specimen measured 1.5 cm and was analyzed entirely on microscopy. The lesion consisted of adipocyte lobules of varied large size [Figure 1]. Focally, the lesion was surrounded by a fibrous tissue rim mainly at the interface with the oral mucosa (which was not ulcerated). Deeply, there were minor salivary gland lobules, interspersed completely or in part by adipocytes. The salivary gland showed focal lymphocytic infiltrate, a focus of fibrous regression with ductal atrophy as well as zones of ductal basal cell hyperplasia and/or large duct dilatation. Inflammation and fibrosis/fibrous septae in the adipose tissue were very sparse. There was no hemorrhage, necrosis, or adipocyte atypia. However, numerous hemorrhagic areas were seen in marginal zones, probably in relationship with intraoperative vascular section/rupture. Sparse mast cells and mildly atypical fibroblasts were observed between adipocytes. Rare nerves were seen in close association with adipocyte, glandular lobules and large vessels at the periphery of the specimen. There was no striated muscle. Intratumor blood vessels were large, branched with divergent trajects and showed thickened wall. Several small sized vessels were also seen. There were no thrombi. The vascular walls showed tufts or papillary projections on serial cut sections. Smooth muscle actin was expressed in the vessel wall and consisted of a rim of positive cells of varied thickness. Endothelial cells were positive for CD31 and negative for D2.40 (expressed in superficial, pericapsular lymphatics).

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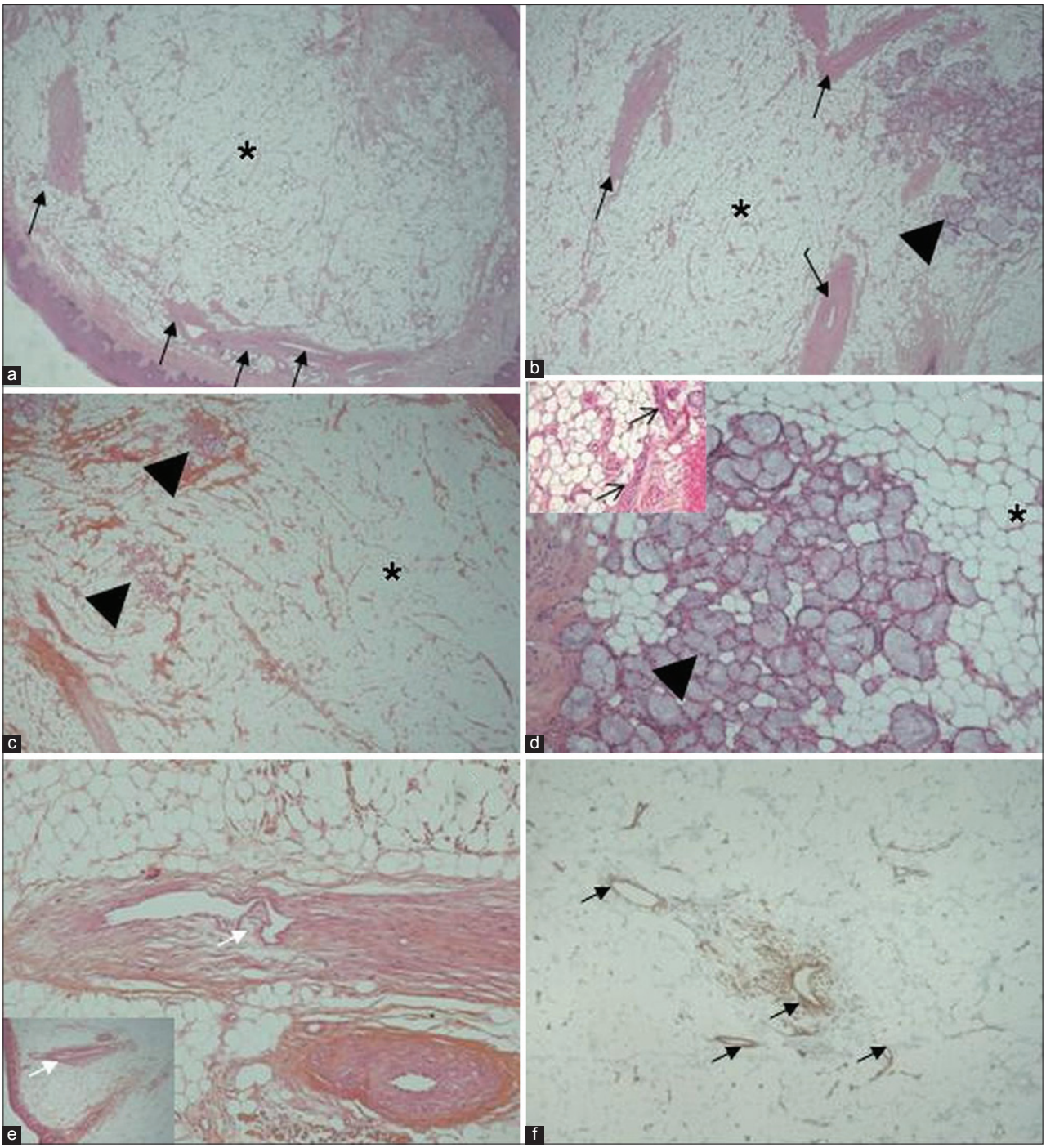

Figure 1: (a) Photomicrograph showing lesional tissue consisting mainly of adipose tissue (asterisk) covered by an intact epithelium with a fibrous band below the epithelium (black arrows: vessel walls) (H&E stain, x25), (b) Lesional tissue showing large, ramified vessels with irregularly thickened wall (black arrows) and salivary gland lobules (arrow head) (H&E stain, x50), (c) Areas showing salivary gland lobules (arrow head) and adipose tissue (asterisk) (H&E stain, x25), (d) High power view showing the salivary gland lobules (arrow head) (H&E stain, x200) (asterisk: adipose tissue) (Inset: Photomicrograph showing adipose tissue and nerves/arrows, H&E stain, x100), (e) Vessel walls showing papillary projections (white arrows) (H&E stain, x200) (Inset: Low power view of the vessel's wall (H&E stain, x25), (f) Vessel wall showing positivity for smooth muscle actin (black arrows) (IHC stain, x50).

DISCUSSION

Here, we report a case of sialolipoma of the hard palate peculiar by the presence of intratumoral malformation-type vascular patterns. Several blood vessels were large, with thickened wall and rare papillary projections or tufts. This latter morphological feature, more frequently reported in vascular malformations than in angiomas when considering the gastrointestinal location, rather suggested a vascular lesion of malformation-type than an associated angioma or

an angiolipoma-type lesion.[7] Moreover, the presence of nerves favored the hypothesis of vascular malformation type lesions.[8] With regard to hard palate angiolipomas, defined as lipomas with a prominent capillary network,[3] these tumors are rare to our knowledge, reported in two cases: one in a young adult man and one in a girl.[3,9,10] They are located laterally on the right or left side, age predilection is mainly between 60-70 years (although a symptom duration of 15 days to 10 years) and measure more than 4-cm in size. The size of hard palate sialolipomas (1–1.8 cm) and their age distribution reported in the literature are closer to that of the present case.[1-6]

Several other adipocyte-abundant lesions are also to be ruled out.[11] A traumatic pseudolipoma was not considered since the evolution was long and reparative signs such as fibrosis and inflammation were limited. However, the association of minor traumas cannot be completely ruled out. The diagnosis of lipomatosis was also not considered due to the presence of capsular tissue neither was an infiltrating lipoma considered, although this lipoma-type can contain large vessels.[11] Another hypothesis is that of entrapped vessels in adipose tissue, difficult to sustain for a low-fat location such as the hard palate. The origin of lipomatous lesions of the hard palate, is similar to that of oral lesions and remains incompletely elucidated. Lipoblastic embryonic cell nests, metaplasia of muscle cells and fatty degeneration can be incriminated as originary events while trauma (with or without soft palate tissues dislocation), infection, chronic irritation or hormone imbalance can be considered as favoring factors.[12,13] Whether an altered oxygenation as a result of intra-tumor malformative vessel network may have an impact on a limited growth although there was a long evolution period remains to be further elucidated.[14] It should also be taken into consideration that losartan-treated patients show a higher body mass index as compared to placebo-treated patients (despite randomization), although losartan does not affect this index.[15] However, adipose hyperplasia or submucosal adipose involution in the context of overweight/obesity may be considered among the hypotheses for histogenesis of fat lesions of the hard palate. Interestingly, a hard palate lipomatous lesion of osteolipoma type is reported to be associated with cleft palate in a 6-year-old boy.[12] Cleft palate patients are also reported to rarely show vascular malformations either *de novo*, reported in pediatric cases[16,17] or as a manifestation after post surgical treatment, reported in a 25-year-old woman.[18] Whether in the present case a subclinical osseous malformation was present remains difficult to be determined due to the lack of computed tomography scan.

CONCLUSION

The microscopic features of the tumor in the present case suggest that malformation-type vascular patterns may occur in sialolipomas of the hard palate. This morphological vascular peculiarity should be acknowledged since it can be a possible source of hemorrhage.

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Conflicts of interest

There are no conflicts of interest.

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