Vascular Transformation of Bilateral Cervical Lymph Node Sinuses: A Rare Entity Masquerading as Tumor Recurrence

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Abstract Vascular transformation of sinuses (VTS) is a rare and reactive vasoproliferative disorder infrequently affecting the cervical lymph nodes. It is characterized by effacement of nodal architecture by variable expansion of the subcapsular, intermediate, and medullary sinuses. We report a very rare and unique case of VTS in bilateral cervical lymph nodes along with angiolipomatous hamartoma in a postoperative patient of squamous cell carcinoma of buccal mucosa clinically masquerading as tumor recurrence. To the best of our knowledge, only 15 cases of VTS have been reported in cervical lymph nodes till date and associated angiolipomatous or angiomyomatous hamartoma-like area was noted only in two cases of cervical lymph node VTS.

Keywords Vascular transformation of sinuses · Cervical · Post-operative · Post-radiotherapy · Hamartoma

Introduction

Vascular transformation of sinuses (VTS) is a rare and acquired condition in which the vasoproliferative process is restricted to the lymph node sinuses and does not involve the parenchyma, capsule, or perinodal fibroadipose tissues [1]. It is usually an incidental finding and has been associated with efferent lymphatic and/or venous obstruction by tumor or thrombus or as a result of radical surgery or radiotherapy. However, similar changes have been produced by experimental occlusion of efferent lymphatics and veins, but obstruction may not be the sole cause, as such changes have been seen in cases lacking obvious impediments to vascular drainage [2]. It is very important to differentiate it from malignant vascular tumors of lymph nodes; especially from Kaposi's sarcoma (KS) [3]. Most of the VTS cases were diagnosed incidentally in excised lymph nodes during a variety of surgical procedures mostly for neoplastic lesions. Intraabdominal lymph nodes are the commonest to be affected by VTS, cervical being relatively rare [4]. We report a very rare and unique case of VTS in bilateral cervical lymph nodes along with angiolipomatous hamartoma in a postoperative patient of squamous cell carcinoma of buccal mucosa clinically masquerading as tumor recurrence.

Case Report

A 56-year-old “gutkha” chewer male presented with bilateral cervical lymphadenopathy for last 2 months. The patient was a diagnosed case of moderately differentiated squamous cell carcinoma (SCC) in the right buccal mucosa (Fig. 1) for which he underwent wide excision of tumor mass along with segmental mandibulectomy with unilateral

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modified radical neck dissection 8 months back. The tumor had TNM stage T4N1M0 with metastasis in a single right submandibular lymph node. The tumor displayed similar pTNM staging after histopathological examination. He received bilateral post-operative conventional radiotherapy of 59.4 Gy with a daily fraction size of 1.8 Gy. On examination, three discrete, nontender, firm mid to lower cervical lymph nodes with somewhat restricted mobility—one on left side and two on the right were found. The largest lymph node measured approximately 2.5 cm × 2 cm and smallest being 1.5 cm in diameter clinically. Clinician suspected it to be a case of regional recurrence (cervical node metastasis) and asked for FNAC (fine needle aspiration cytology). One lymph node on right side displayed the feature of metastasis of SCC (Fig. 2) but the FNAC of rest of the lymph nodes displayed a poor yield with scanty lymphocytes, very few histiocytes in a hemorrhagic background without any metastatic deposit. The patient underwent lymph node excision biopsy. The two lymph nodes which were negative for metastasis in FNAC, demonstrated diffuse and almost complete effacement of nodal architecture by variable expansion of the subcapsular, intermediate, and medullary sinuses having a morphologic spectrum of slit or cleft like spaces embedded in sclerotic stroma, rounded or dilated vascular spaces, inter-communicating plexiform channels and even solid areas with plump and spindled cells (Fig. 3). Sinusoidal transformation was seen in various stages of development. Maturation of vascular channels from relatively solid areas in central region to well-formed ectatic vascular channels near capsule was also noticed. There was marked atrophy of lymphoid population with occasional lymphoid follicles with or without germinal centers (Fig. 4). Although the fibrous capsule of lymph nodes was conspicuously thickened, it remained uninvolved by the vascular proliferation. Prominent thick-walled blood vessels were observed in the perinodal fibrofatty tissue. The narrow cleft like vascular spaces were mostly empty or having very few red blood cells, but the dilated and larger vessels contained either lymphatic fluid, blood, or a mixture. There were noticeable extravasation of red blood cells, hemosiderin deposition and conspicuous stromal sclerosis including perisinusoidal hyalinosis (Fig. 5). The vascular channels were lined by flatted as well as plump epithelioid endothelial cells (Fig. 6). However, nuclear atypia or increased mitotic activity was absent in vasoformative cells. Intraparenchymal interposed mature adipose tissue alone (Fig. 7) and intimately admixed with sinusoidal and subcapsular vascular channels as in angiolipomatous hamartoma (Fig. 8) was observed in one of these two lymph nodes. Further, deposition of stromal hyaline globule like droplets and nodal or perinodal venous thrombi were also not seen. We had planned for salvage neck dissection but the patient was lost during follow up.

Fig. 1 Moderately differentiated squamous cell carcinoma invading the muscles (H and E, ×100)

Fig. 2 Metastasis of squamous cell carcinoma in lymph node cytology (Leishman stain, ×100)

Discussion

VTS is a reactive process in which the sinusoidal architecture is replaced by an anastomosing network of vascular channels ranging from capillaries to cavernous spaces. The term vascular transformation of sinuses (VTS) was first coined by Haferkamp et al. [5] in 1971. Although lymphovascular obstruction has a major pathogenetic role in most cases of VTS, angiogenic factors produced locally by activated lymphoid cells can also allegedly produce VTS even in the absence of lymphovascular obstruction [4]. VTS itself is a rare lesion and cervical lymph node is a very unusual site for VTS. To the best of our knowledge, only 15 cases have been reported in cervical lymph nodes till date [4, 6–8]. Further, associated angiolipomatous or angiomatous hamartoma-like area was noted only in two cases of cervical lymph node VTS [6, 8].

Fig. 3 Photomicrograph of VTS showing uninvolved capsule, thin walled cleft like vascular spaces and maturation of the spindle cells into well-formed vascular channels towards the capsular aspect in the direction of the arrow (H and E, $\times 100$)

Fig. 5 Photomicrograph of VTS displaying extravasation of red blood cells, hemosiderin deposition, perisinusoidal hyalinosis (*broken arrow*) and dilated vascular channels containing lymphatic fluid (*solid arrow*) (H and E, $\times 100$)

Fig. 4 Thickened fibrous capsule and residual lymphoid follicle with germinal center in VTS (H and E, $\times 100$)

Fig. 6 Plump endothelial cells lining the vascular channels in VTS (H and E, $\times 400$)

The differential diagnoses of VTS include a variety of vasoproliferative lesions occurring in lymph nodes. Mostly the cellular forms VTS may be mistaken for Kaposi's sarcoma owing to histological similarities like narrow vascular channels, spindle cells and extravasation of red cells, but differs from KS by the pure sinusoidal distribution, a lack of overgrowth of atypical spindle cell fascicles, associated intralesional sclerosis, maturation of the spindle cells into well-formed vascular channels toward the capsular aspect, absence of capsular involvement and rarity of eosinophilic hyaline globules [1, 4].

The rare primary nodal hemangiomas and hemangioendotheliomas should be distinguished from VTS by the presence of a well-circumscribed nodular growth, absence of sinusoidal pattern and predilection for hilar and

medullary region of lymph nodes [9, 10]. Further, bacillary angiomatosis, occurring exclusively in the immunocompromised subjects show plump endothelial cells, abundant neutrophils, presence of deeply eosinophilic interstitial granular material and clumps of Warthin-Starry-positive bacilli [1, 9].

In our case radical surgery followed by prolong radiation possibly caused a locoregional lymphovascular impediment leading to the rare occurrence of VTS in cervical lymph node clinically mimicking tumor recurrence. Further coexistence of VTS with angioliomatous hamartoma added to the rarity of the case.

It is very important for both the pathologist and clinical colleague to be aware of this unusual lymph node entity in an unusual site. It should not be mistakenly over diagnosed

Fig. 7 Interposed mature adipose tissue in lymph node of VTS (H and E, ×100)

Fig. 8 Angiolipomatous hamartoma like area in VTS (H and E, ×100)

as hemangioendothelioma, angiosarcoma or Kaposi's sarcoma. Physician should be alert with the finding of VTS reported specially in an uncommon site as there is possibility of finding clinically detectable or occult primary malignancy or even a tumor recurrence in adjacent lymph node as in this case. This mandates prompt clinical caution in such a setting, thorough diagnostic work up to search for

primary pathology followed by its immediate treatment, thus indirectly influencing prognosis.

To the best of our knowledge, the literature regarding the management of VTS in head and neck cancer are very scarce. Most likely the treatment of a case of SCC may not change with the presence or otherwise of VTS. Nevertheless, the authors strongly believe that VTS shouldn't merely be deemed of as only a rare diagnostic entity that develops secondary to a primary pathology but should be dealt with extra caution as a diagnostic problem which can masquerade as tumor recurrence clinically and also play a game changing role in having a strong indirect impact on prognosis as mentioned above.

Conflict of interest The authors declare that they have no conflict of interest.

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