

Pleomorphic Adenoma Arising from Heterotopic Salivary Gland Tissue in the Neck : A Case Report¹

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We report a rare case of pleomorphic adenoma arising from heterotopic salivary gland tissue in the upper neck. Although it is difficult to differentiate this condition from lymph node diseases-including metastasis- on the basis of radiologic findings alone, it should be included in the differential diagnosis of a solitary unilateral solid cervical mass, particularly one in the upper neck.

Index Words : Neck, neoplasms
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Salivary gland tissue, other than that comprising the three major and the minor salivary glands in the upper aerodigestive tracts, is either heterotopic or ectopic(1). Heterotopic and accessory glands are different, in that the latter are found as detached bodies along a major salivary duct while the former have no direct relation with major salivary glands(1). This tissue is occasionally found in the neck, though tumors arising from it are very rare(2, 3); clinically and radiologically, it is nearly impossible to distinguish them from far more common lymph node diseases including primary or secondary malignancy. Although several reports on tumors of heterotopic salivary tissue have appeared in the literature(2-8), few have been described in the radiologic literature. We present a case of pleomorphic adenoma arising from heterotopic salivary gland tissue in the upper neck.

Case Report

A 52-year-old woman presented with a palpable mass in the right of the neck. Since she had first noticed the mass six years ago, it had grown slowly. She denied any significant past illness and no other complaints

were associated with the mass. Physical examination revealed a 2×2cm movable nontender mass, of rubbery consistency, in the right upper neck anterior to the sternocleidomastoid muscle and below the angle of the mandible. CT of the neck before and after contrast enhancement showed a 1.8×1.5 cm well-margined, ovoid mass with homogeneously low attenuation, posterior to the right submandibular gland and anterior to the internal jugular vein and sternocleidomastoid muscle at the level of the hyoid bone. Within the mass, no calcification was noted, and after the administration of contrast material, there was only subtle enhancement(Fig. 1A). The parotid and submandibular glands appeared normal, and fatty interfaces were preserved between the mass and the glands(Fig. 1B). A 1.5×1.5 cm well-margined calcified mass was found incidentally in the right lobe of the thyroid gland(Fig. 1C). Although it seemed unlikely, the possibility of thyroid cancer, with metastasis to the upper cervical lymph node, could not be entirely excluded by imaging findings alone. Laboratory tests, including the thyroid function test, were all unremarkable.

Both the upper cervical and thyroid masses were surgically removed; a 2×2cm tan ovoid mass was removed from the right neck and several small lymph nodes from the vicinity of the mass. There were clear cleavage planes between the mass and the submandibular or parotid gland. Histologically, the mass proved to be a pleomorphic adenoma. No foci of residual normal salivary tissue or lymphoid follicles were histol-

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Fig. 1. 52-year-old woman with pleomorphic adenoma of heterotopic salivary gland tissue in the neck.
A. Contrast-enhanced CT scan at level of the hyoid bone shows a well marginated, ovoid, poorly-enhancing mass with homogeneously low attenuation (arrows), located posterior to the right submandibular gland (S) and anterior to the internal jugular vein and sternocleidomastoid muscle. No calcification is seen within the mass.
B. CT scan at level of the lowermost tip of parotid gland shows intact parotid (P) and submandibular (S) glands. At surgery, there were clear cleavage planes between the mass and the submandibular or parotid gland.
C. CT scan at level of the supraclavicular fossa shows a calcified mass in the posteroinferior aspect of the right lobe of the thyroid gland. Surgery confirmed a follicular adenoma.

ogically demonstrated in the fibrous capsule of the tumor, and chronic nonspecific lymphadenitis was diagnosed from the small lymph nodes removed; within them, there was no evidence of foci of salivary tissue. Pathologic examination of the thyroid mass revealed a follicular adenoma.

Discussion

Heterotopic and accessory salivary glands are different, in that the latter are found as detached bodies along a major salivary duct while the former have no direct relation with major salivary glands(1). Heterotopic salivary glands have been found in various locations including the hypophysis, cerebellopontine angle, external and middle ears, thyroglossal duct, mandible, thyroid and parathyroid capsules, lymph nodes, upper and lower neck regions, and sternoclavicular joints(1, 2, 6). In the neck, they are most commonly found in the lower third anterior, as far as the sternocleidomastoid muscle, and predominantly on the right side. The pathogenesis of cervical heterotopias still remains unclear, and several different theories have been proposed. Salivary tissue may develop either from heteroplastic changes of normally existing epithelial structures, or from embryonic remnants of branchial apparatus(3); entrapment within lymph nodes during embryonic development explains their inclusion in the periparotid and upper cervical regions (1).

Whatever the pathogenesis of cervical heterotopias might be, neoplasms arising from them are quite rare. A review of the literature disclosed that since the first

report by Pesavento and Ferlito in 1976, less than 30 cases of tumors of cervical heterotopic salivary gland tissue have been reported(2—8). There is wide variation in age, from 8 to 81 years, and a slight female predominance. In contrast to histologically normal heterotopic salivary tissue, tumors were most frequently located in the upper neck, and more than half the cases are believed to have originated from lymph nodes. Malignant and benign tumors were approximately equally common; among these, pleomorphic adenoma is the most common, while the most frequent malignant tumor is mucoepidermoid carcinoma(3, 5, 8). In cases of malignancy, metastatic and primary neoplasms are histologically indistinct(5). A primary tumor should be sought elsewhere in the major or minor salivary glands and if one is not found, these tumors can be justifiably considered to be primary(3, 5, 8).

In the present study, CT characteristics of the heterotopic salivary pleomorphic adenoma were the same as those of a small benign mixed tumor of the major or minor salivary glands. CT, however, offered no specific clue to diagnosis, and this is not surprising. As expected, the usual locations of heterotopic cervical salivary gland tumors are little different from those of cervical lymph nodes or second branchial cleft cysts. Furthermore, the morphological characteristics of pleomorphic adenoma, including the pattern of contrast enhancement seen on CT, are quite similar to those of various kinds of lymph node disease or other granulomatous diseases of the neck.

In summary, we reported a case of pleomorphic adenoma arising from heterotopic salivary gland tissue in the upper neck. Although the radiologic findings are

nonspecific, this condition should be included in the differential diagnosis of a solitary unilateral cervical mass, particularly one in the upper neck.

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경부의 이소성 타액선에 발생한 다형선종: 1예보고¹

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이 증례는 경부의 이소성 타액선 조직에서 발생한 다형선종에 관한 것이다. 이 질환은 편측성 단일 고형성 종괴가 특히 상부 경부에서 발견되고 방사선학적인 검사로는 림프절 전이암을 포함한 다른 림프절 질환과의 감별이 어려운 경우 감별진단의 하나에 포함되어야 할 것이다.

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