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Extraparotid Warthin Tumor in Upper Cervical Lymph Node Accompanied by Primary Cervical Tuberculosis

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Warthin tumor (WT) accounts for about 5% to 10% of benign parotid neoplasms.¹ Approximately 8% of WTs occur in an extraparotid localization; the most common site is the periparotid and upper cervical lymph nodes.² Extraparotid Warthin tumor (EPWT) arising in sites other than a cervical node is highly uncommon. The following is a clinical report about an EPWT in the upper cervical lesion (level II) associated with primary cervical tuberculosis. We report a unique case of EPWT with primary tuberculosis, which, to the best of our knowledge, has not been reported before. This report was approved by the institutional review board of our center.

Case Report

An 81-year-old man who was a heavy smoker visited our clinic, presenting with an upper lateral neck mass that had been present for the past 7 months. Physical examination revealed that the mass was 3 × 2.5 cm in size, rubbery hard, slightly movable, and nontender on right level II area. Enhanced neck computed tomography (CT) showed a well-margined, heterogeneously enhanced mass, just posterior to the right submandibular gland (Figure 1). In fine needle aspiration cytology (FNAC), histiocyte and some lymphoid cells were observed. We initially pre-diagnosed the patient with branchial cleft cyst (type II), lymphangiomata, Castleman disease, or cystic malignant lymphadenopathy, and the initial plan was excisional biopsy. Under general anesthesia, the surface was slightly in adhesion with the submandibular gland. But the mass was not connected to the parotid gland or submandibular gland. Low-power microscopic specimen findings showed that the lymph node was mostly replaced by the WT and caseating granulomas (Figure 2). No acid-fast bacillus was noted on Ziehl–Neelsen staining. The paraffin-embedded tissue with polymerase chain reaction (PCR) was positive. Histopathological examination revealed WT mixed with primary tuberculosis. The patient underwent antituberculous medications for 9 months. There has been no recurrence at 18 months.

Figure 1. Contrast-enhanced axial CT scan shows well-margined heterogeneously enhanced mass, just posterior to the right submandibular gland.

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Discussion

WT is characterized by papillary growth, cystic formation, and lymphocytic infiltration. WT is the second most common neoplasm in the parotid gland; it tends to occur bilaterally and multifocally and to synchronize with other salivary neoplasms.\textsuperscript{1} The occurrence of WT is strongly associated with smoking and autoimmune disease (ie, diabetes mellitus, Hashimoto thyroiditis, autoimmune hyperthyroidism, and hypothyroidism).\textsuperscript{3} The histogenesis of WT is not fully revealed, but the heterotopic theory is generally accepted. First, tumors develop from the heterotopic salivary ducts from the preexisting intraparotid or paraparotid lymph tissue. Second, T- and B-cell components of WT are polyclonal and the lymphoid tissue is not neoplastic but rather could be a delayed hypersensitivity reaction to proliferating ducts and secretions.\textsuperscript{4} Recently, monoclonal antibodies were used to show that the proportion of T to B cells in WT was near that of a normal lymph node, thus strengthening the theory of salivary organogenesis over previous theories.

Encapsulation of the parotid gland develops at the 14th week of gestation, which is late compared with other salivary glands. The pathogenesis of EPWT in an upper cervical lesion is known to arise because of the late encapsulation of the parotid gland, enabling the mixing of undifferentiated lymphoid stroma with an organizing meshwork of branching salivary ducts.

Compared with other salivary neoplasms, the diagnostic rate of WT with FNAC is slightly lower (61%-83%) because it is confirmed in the coexistence of oncocyte and lymphocyte. Because WT usually has no pseudopod and malignant change is extremely rare (0.06%-3%), the surgical dissection is performed close to the tumor capsule. Tuberculosis in the salivary gland is presented mainly in the parotid gland, and it is estimated that infection results from the tooth or tonsil. The coexistence of WT and mycobacterial infection is extremely rare, with less than 20 cases reported in the English-language literature. WT is often accompanied by various types of inflammation, and it is important to distinguish tuberculosis from other inflammations. It may be confirmed by paraffin-embedded block with PCR. When FNAC combined with PCR is performed, the sensitivity and specificity in the diagnosis of cervical tuberculosis may be increased to more than 90%. Tuberculosis shows positive in Ziehl–Neelsen staining only if there are more than 10,000 organisms per milliliter of mycobacterium tuberculosis. In secondary lesions such as the lymph node and salivary gland, the number of bacteria is insufficient.\textsuperscript{5} Gallium-67 scan may be helpful in the differential diagnosis between neoplastic and inflammatory disease.

Author Contributions

Jun Yeon Hwang, design, acquisition of data, analysis and interpretation of data, revision; Seung Woo Kim, conception and design, acquisition of data, analysis and interpretation of data, revision, final approval; Si Chang Yang, design, acquisition of data, analysis and interpretation of data, final approval; Choon Dong Kim, design, acquisition of data, analysis and interpretation of data, revision, final approval.

Disclosures

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