Malignant Transformation of a Benign Oncocytoma of the Submandibular Gland: A Case Report

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Oncocytic carcinoma arising in the submandibular gland is an extremely rare tumor and only 11 cases have been reported previously. We report on a 51-year-old man with a previously benign oncocytoma in the submandibular gland that transformed from a benign morphology to malignant cellular atypia and mitosis. To our knowledge, the current report is the first published case of a malignant transformation from benign oncocytoma to oncocytic carcinoma of the submandibular gland. The proliferative activity of the tumor cells was evaluated immunohistochemically using antibodies against Ki-67.

Key Words: benign oncocytoma, Ki-67 immunohistochemical stain, malignant transformation, oncocytic carcinoma, submandibular gland
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Salivary gland tumors are rare, comprising less than 3% of all neoplasms of the head and neck region. The majority of salivary tumors are located in the parotid gland (70%), followed by the minor salivary glands (22%) and the submandibular glands (8%) [1]. Oncocytic tumors comprise only 1% of all salivary gland tumors [2]. Oncocytic carcinomas are even more uncommon, representing 11% of all oncocytic salivary gland neoplasms, 0.5% of all epithelial salivary gland malignancies and 0.18% of all epithelial salivary gland tumors [3]. Oncocytic carcinoma arising in the salivary glands, first described by Bauer and Bauer in 1953, is a rare, predominantly oncocytic neoplasm [3,4]. Moreover, oncocytic carcinoma arising in the submandibular gland is an extremely rare tumor and only 11 cases have been reported to date [5]. We report a 51-year-old man with a previously benign oncocytoma in his submandibular gland that transformed from a benign morphology to malignant cellular atypia and mitosis. We performed immunohistochemical staining with antibodies against Ki-67 to qualify mitosis separately for clarity of the tumor. This study was approved by our Institutional Review Board (IRB 09801-008).

Case Presentation

A 51-year-old man presented with a recurrent mass in his left submandibular gland in August, 2008, and a prior diagnosis of benign oncocytoma in the same location. In February, 2005, he underwent total tumor ablation for the benign oncocytoma in his left submandibular gland. Pathological examination of the resected tissue of the earlier specimen revealed an encapsulated tumor (Figure 1A) composed of uniform oncocytes with fine chromatin and indistinct nucleoli (Figure 1B). Mitotic figures were rarely found.

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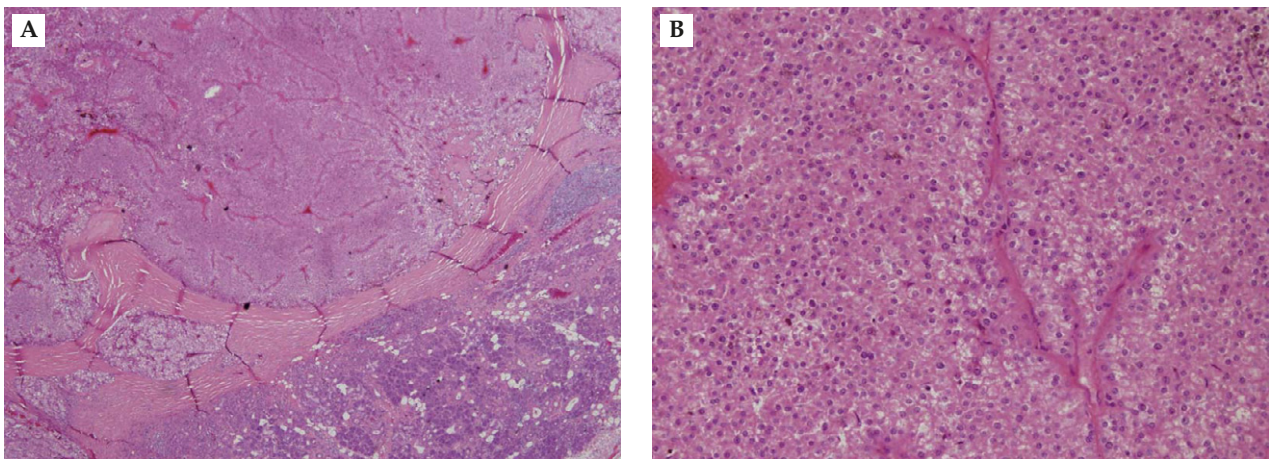

Figure 1. *(A) The tumor was well encapsulated (hematoxylin and eosin; original magnification, 100×). (B) Uniform oncocytes with abundant eosinophilic granular cytoplasm, fine chromatin, indistinct nucleoli and rare mitoses (hematoxylin and eosin; original magnification, 400×).*

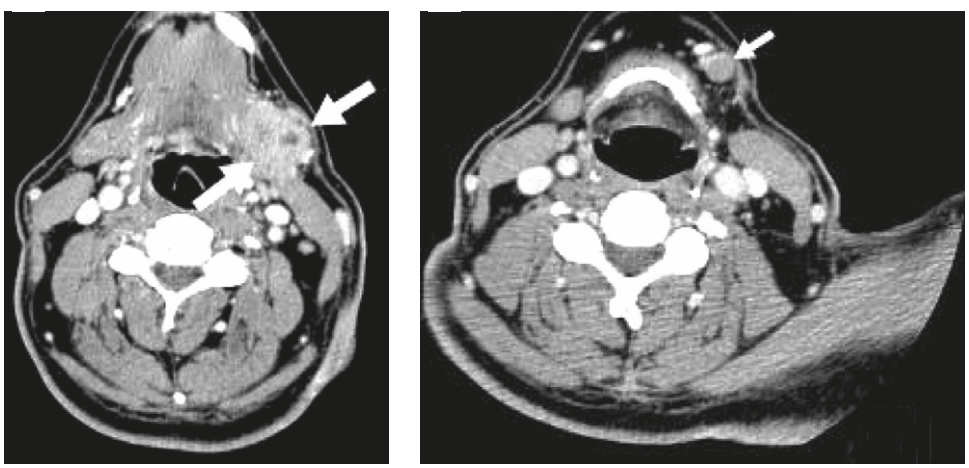

Figure 2. *(A) Axial computed tomography revealed a mass with central necrosis in the left submandibular region (arrows). (B) One enlarged lymph node was found anterior to the mass (arrow).*

At the current presentation, he had a painless swollen mass in the previously resected area. This lesion had been slowly growing for 4 months. On clinical examination, the mass was about 3.0 × 2.5 cm in size, hard, rubbery and fixed. A palpable cervical lymphadenopathy was also found. The facial nerve and other cranial nerves functioned normally. Neck sonography revealed one well defined heterogeneous mass lesion in the left submandibular space, measuring 3.3 × 2.8 cm in size. Several lymph nodes surrounding the tumor were swollen, with the largest one measuring about 1.2 × 0.9 cm in size. Computed tomography of the neck demonstrated one enlarged mass lesion about 3.0 cm in diameter in the left submandibular region (Figure 2A) and an enlarged lymph node, of about 1.4 cm in diameter, anterior to the submandibular mass (Figure 2B). Because recurrence of the oncocytoma was suspected, radical tumor resection with unilateral, modified neck dissection (levels I–III) was performed in September 2008.

Microscopically, the resected tumor revealed infiltrating growth and was composed of atypical oncocytes arranged in solid sheets. Marked nuclear atypia, cellular polymorphism and mitoses were observed (Figure 3A). Focal areas of necrosis were also found. The tumor had invaded the surrounding tissue, including the muscular tissue, perineural spaces and lymphatic vessels (Figure 3B). Three of the four lymph