Case report

A ciliated cyst as a component of pleomorphic adenoma of the parotid gland

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A large cyst, lined with ciliated cells (ciliated cyst), was found as a component of a pleomorphic adenoma of the left parotid gland in a 34-year-old Japanese man. The dimensions of the tumor were 25×33 mm and it consisted of a solid mass with a centrally located cyst 20×23 mm, which contained yellowish-brown material of muddy consistency. The tumor was encapsulated and the solid mass had features typical of a pleomorphic adenoma. The inner surface of the cyst was lined with ciliated cells and scattered mucus-secreting cells in addition to cuboidal-to-flattened cells. In some regions, the neoplastic cells of the pleomorphic adenoma were exposed to the cavity of the cyst. A histological transformation from ciliated cells to pleomorphic adenomatous cells was also observed. These findings indicated that the ciliated epithelium was an element of the tumor. This is the first report, to our knowledge, of a ciliated cyst derived from some elements of a pleomorphic adenoma of the salivary gland.

Key words: ciliated cyst; parotid gland; pleomorphic adenoma

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Cystic lesions of the salivary gland can be categorized into two groups, depending on whether or not they are related to a neoplasm. Several benign and malignant tumors, including pleomorphic adenoma (1-4), Warthin's tumor (2, 3, 5), and mucoepidermoid carcinoma (3, 5), are known to be associated with the formation of cysts, whereas non-neoplastic cysts include retention, branchial cleft, and epidermal inclusion cysts (2, 3, 5-9). Formation of a cyst that originates from a pleomorphic adenoma is relatively rare, and most such cysts are degenerative pseudocysts without lining cells. By contrast, a true cyst with lining cells, resulting from a cystic dilatation of the glandular elements of a pleomorphic adenoma, has been reported in a few cases (2-5). Whatever its cause or type, a large cyst originating from a pleomorphic adenoma is rare (2-4). To our knowledge, there are no previous reports of a large cyst lined with ciliated cells (a ciliated cyst) as an element of a pleomorphic adenoma.

Case report

A 34-year-old Japanese man had been aware of a mass in the left parotid region for 3 years without any pain or facial palsy. The patient was admitted to a local hospital for excision of the mass. Past and family histories of the patient were non-informative. At admission, a large elastic hard mass was palpated in the left parotid region, without invasion, as observed also by CT and MRI. Radiographic examinations, including sialography, gallium scintigram, computed tomography (CT) and magnetic resonance imaging (MRI), revealed that the tumor, located in the left parotid gland, was a well-demarcated mass that measured 25×33 mm, with an associated cyst (Fig. 1). Abnormal concentrations were absent on the gallium scintigram, and no communication between the cyst and the salivary duct was observed on sialography. The patient underwent surgery after a clinical diagnosis of benign parotid tumor had been made. At surgery, the tumor was found in the superficial lobe of the left parotid gland and was well demarcated, without invasion, as observed also by CT and MRI.
MRI. Left superficial parotidectomy was performed, and the patient remains well, without recurrence, 26 months after surgery.

Pathologic findings

On macroscopic examination, the tumor was clearly demarcated and consisted of a peripheral solid mass with a central smooth-surfaced cyst (Fig. 2). The cyst measured 23×20 mm and contained yellowish-brown material of muddy consistency.

On microscopic examination, the tumor was found to be encapsulated and the cyst contained degenerative material with cholesterol crystal clefts (Fig. 3). The solid part of the tumor had features typical of a pleomorphic adenoma, namely, an intimate admixture of cuboidal to spindle-shaped myoepithelial cells, glandular structures with production of mucin, and foci of squamous epithelium in a fibromyxoid stroma (Fig. 4). Both chondroid and osteoid tissues were absent. Most of the inner surface of the cyst was lined with ciliated cells and scattered mucous cells (Fig. 5), but some parts of the cyst were lined by a single layer of cuboidal-to-flattened cells to which some of the neoplastic cells of the pleomorphic adenoma were exposed. A histological shift from ciliated cells to some elements of pleomorphic adenoma was also detected (Fig. 6). There was no lymphoid tissue in the wall of the cyst.

Discussion

The frequency of cystic lesions of the salivary gland, which are found under both neoplastic and non-neoplastic conditions, is 7.4% to 8.7% of all such lesions (2,3,9,10). The size of most of these cysts is small, and cysts that are large enough to be observed macroscopically are relatively rare (2).

Some authors have classified cystic lesions of the parotid gland morphologically into two types: neoplastic and non-neoplastic cysts (2,3). There are several types of non-neoplastic cyst of the parotid gland, including lymphoepithelial cyst, retention cyst or salivary duct cyst, epidermal inclusion cyst, and hamartomatous or heterotopic cyst. Among neoplastic tumors, War-
thin's tumor and pleomorphic adenoma are known to form cysts. Pieterse & Seymour (2) reported formation of a cyst in two of 87 pleomorphic adenomas of the parotid gland (2.2%). Maynard (3) demonstrated that 42% of parotid solitary cysts were associated with neoplasms, including pleomorphic adenoma, Warthin's tumor, mucoepidermoid carcinoma and squamous cell carcinoma, and pleomorphic adenoma accounted for 40% (4/10) of these cases. Two reasons for formation of cysts in pleomorphic adenomas have been proposed. A cyst can result from degeneration within a tumor or, alternatively, cystic dilatation of a glandular element can occur (10). In the first case, degeneration or necrosis of some part of the tumor causes cyst formation. Such a cyst has no lining cells or is exposed to tumor tissue. In the second case, since a pleomorphic adenoma is usually an encapsulated mass without any communication with the salivary duct, secreted material accumulates within a glandular structure. As a result, the glandular structure becomes cystically dilated. This type of cyst is usually lined by flattened-to-cuboidal epithelial cells. Although one case of an epidermal inclusion cyst in a pleomorphic adenoma has been reported (1), an epidermal inclusion cyst could be considered to be a cystically dilated glandular structure with squamous metaplasia. In the present case, since the cyst was located in the central region of a pleomorphic adenoma and since we observed a histological shift from ciliated cells to pleomorphic adenoma, the ciliated cells should be considered to be an element of the pleomorphic adenoma. We suggest, therefore, that the ciliated epithelium was the result of the metaplasia or differentiation of neoplastic cells from some element of the pleomorphic adenoma. Ciliated cells are known to occur in large salivary ducts, and even to be a prominent feature, especially when the goblet cells of the duct increase in number and produce abundant mucus during chronic inflammation (10). Of the salivary gland tumors, Warthin's tumor has been reported to represent ciliated cells on examination by scanning electron microscopy (11). Embryologically, the parotid gland is detected in the embryo at the seventh week of gestation as a furrow in the floor of the alveolobuccal groove, located adjacent to the angle of the mouth. The original cells of the parotid gland are the same as those of the nasal cavity because cells consistent with parotid outgrowth originate from the ectodermal epithelial cells on the stomodeum. Thus, it is not so strange that differentiation or metaplasia that results in formation of cells resembling those of the respiratory epithelium can occur in a pleomorphic adenoma.

References