Sclerosing polycystic adenosis of the parotid gland: Diagnosis and management

F. Perottinoa, R. Barnoudb, A. Ambrunc, M. Poupartc, J.-C. Pignatc, O. Merrotc,

a Escartons Hospital, 24, avenue Adrien-Daurelle, 05100 Briançon, France
b Biology and Pathology Center, Croix-Rousse Teaching Hospital, 103, Grande Rue de la Croix-Rousse, Claude-Bernard Lyon-1 University, 69317 Lyon cedex 04, France
c ENT and Head and Neck Surgery Department, Croix-Rousse Teaching Hospital, 103, Grande Rue de la Croix-Rousse, 69317 Lyon cedex 04, France

Available online 24 March 2010

KEYWORDS
Salivary gland;
Polycystic adenosis;
Parotid tumor

Summary
Objective: To describe diagnostic and therapeutic management of a rare parotid lesion: sclerosing polycystic adenosis.
Patient and method: We report a case of persistent right intraparotid tumefaction.
Results: A 68-year-old man was referred with a right parotid nodule of 2 years’ evolution. Cytology diagnosed pleomorphic adenoma, verified on MRI. Conservative subtotal parotidectomy diagnosed sclerosing polycystic adenosis. Over 1 year’s regular follow-up, there were no signs of local recurrence.
Conclusion: Sclerosing polycystic adenosis of the parotid gland is a rare and recently described entity presenting several analogies to the much more frequent cystic mastitis. Although benign and well-delimited, it requires complete exeresis of the parotid, due to a non-negligible risk of recurrence.
© 2010 Published by Elsevier Masson SAS.

Introduction
Sclerosing polycystic adenosis (SPA) is a rare salivary gland lesion, first described in 1996 by Smith et al. [1]. Its anatomo-clinical features are those of benign and often complex mammary lesions such as cystic mastitis. The principle location is the parotid gland, but it has also been reported in the submandibular and accessory salivary glands.

We report a case of SPA of the parotid gland in a 68-year-old man. Optimal management was guided by a review of the literature.

Clinical case
A 68-year-old man was referred to ENT for right parotid nodule of 2 years’ evolution. He had no medico-surgical his-
Examination found an isolated 2 cm-diameter mobile, pain-free juxta-lobular mass, firm on palpation. The facial nerve was fully functional and the neck showed no palpable adenopathy. Ultrasonography found an 18 mm-diameter intraparotid lesion in the posterior part of the inferior pole of the right parotid. The well-contoured hypo-echogenic US aspect suggested pleomorphic adenoma or cystadenolymphoma and cytology suggested pleomorphic adenoma. MRI was in favor of a pleomorphic adenoma with an oval nodular image measuring $19 \times 14 \times 13$ mm with T1 hyposignal and slight T2 hypersignal, strongly enhanced by gadolinium (Figs. 1 and 2). Conservative subtotal parotidectomy was performed without any postoperative complications. Pathology examination found a well-contoured, partially encapsulated nodular lesion comprising hyperplastic acinous and ductal structures, sometimes harboring microcystic dilatations, within abundant focally inflammatory fibrous and sclerous tissue (Figs. 3 and 4). A few sites of atypical epithelial hyperplasia were found, free of invasive or in situ carcinoma. Immunohistochemistry using smooth-muscle anti-actin antibody (DakoCytomation, dilution 1/4000) found a conserved myoepithelial cell layer, confirming benignity (Fig. 5). The two subdigastric ganglions that were harvested showed no invasion. SPA of the parotid gland was diagnosed. At 1 year’s follow-up, the patient was free of local recurrence; regular surveillance was maintained. There were no functional sequelae.

**Discussion**

SPA of the parotid gland is a rare benign salivary-gland lesion, recently described, sharing the anatomopathologic features of very frequent and well-described benign mammary lesions, often grouped under mastopathy, and comprising complex lesions including cystic mastitis. In the international literature, we found 36 cases, with age at diagnosis ranging from 9 to 84 years for a mean of 40 years, and a sex ratio of 1 [1–10]. Mean tumor size ranged from 0.3 to 6 cm. Location was mainly parotid, but also in the submandibular and accessory salivary glands [1,6,8]. Clinical features were non-specific, with progressive development of a generally asymptomatic intraparotid mass, although pain was also reported in some cases [1,3]. One case was discovered secondarily to surgery for disabling chronic juvenile parotiditis [2]. Tumors were single, generally well-delimited and partially encapsulated, and only much more rarely multinodular [6]. An association of submandibular pleomorphic adenoma and parotid oncocytoma was reported, with serendipitous discovery of a SPA [6]. Cytologic puncture is to be interpreted with caution. In the present case, it confirmed benignity; in the few cases found...
in the literature, however, hypercellular aspects and more atypical cells suggested diagnosis of low-grade mucoepidermoid carcinoma [4,5]. Microscopic examination of the sample found sclerotic lobules with dense, hyalinized stromal collagen supporting sometimes cystized ducts. Stroma and epithelial contingents varied according to the tumor, and cystic remodeling was also of variable degree. As in cystic mastitis, duct-type epithelial hyperplasia, with or without more or less severe atypicality, was found in more than half of cases and was generally interpreted as more or less severe dysplasia and in situ carcinoma [3,6,8]. In mammary pathology, it is important to identify dysplastic lesions given the risk of subsequent infiltrating carcinoma. In salivary gland SPA, carcinomatous degeneration has not been reported, but cannot be completely ruled out in view of the small number of cases and the superficial location of generally small tumors. Immunohistochemistry, by using specific antibodies (smooth-muscle anti-actin), can confirm myoepithelial cell-layer integrity in benign lesions; this layer is lost in carcinomatous lesions. The nature of salivary gland SPA remains to be established. It may be a reactional inflammatory pseudotumoral lesion, but the risk of local recurrence and one study reporting it to be monoclonal combine to suggest a truly neoplastic origin [8]. Management is surgical, with conservative subtotal parotidectomy followed by regular prolonged surveillance. This attitude is justified by the risk of multiple, sometimes late recurrence in case of incomplete exeresis, and the risk of carcinomatous degeneration [1,6--8].

**Conclusion**

SPA of the parotid gland is a rare benign salivary gland lesion with histologic analogies to sclerosing adenosis of the mammary gland. Complete surgical exeresis is the reference treatment, to reduce the risk of recurrence and/or evolution. Surveillance should be continued over several years.

**Conflict of interest statement**

The authors have not communicated conflicts of interest.

**References**