



Lymphoepithelial Cyst and Warthin Tumor of the Parotid Gland: Is It Really Essential to Distinguish Between Them?

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Abstract

Warthin tumour and lymphoepithelial cysts are both benign lesions with different risk factors but same same clinical behavior, and due to the similarities between these two entities differential diagnosis can be challenging. A 66 year-old Caucasian male, smoker, came to our attention with a cystic lesion located within the superficial lobe of the left parotid gland. Medical history was negative for other diseases. Warthin tumor was suspected, and patient underwent enucleation with preservation of the peripheral branches of the facial nerve. Definitive histopathological examination was consistent with lymphoepithelial cyst with reactive lymphadenopathy. It has been hypothesized that lymphoepithelial cysts may be an alternative histogenetic pathway of Warthin tumor, and due to the similarities between these two entities differential diagnosis can be challenging. Nevertheless, the distinction is not fundamental, since surgical treatment for lymphoepithelial cyst and Warthin tumor is identical. Prospective correlation studies may determine whether FNA can be safely deferred in select cases.

Keywords Warthin tumor · Lymphoepithelial cyst · Parotidectomy

Lymphoepithelial cysts are rare benign tumors that derive from salivary duct cysts, which develop secondary to a lymphoid response or as a consequence of cystic degeneration of salivary inclusions, especially in patients infected with HIV. Warthin tumor is more frequently encountered (25–32% of all benign tumors arising in the parotid gland) and is related to smoking habits, developing within an intra- or periparotid node [1–3]. Typically, a lymphoepithelial cyst presents in the fifth decade of life as a painless slowing growing mass without an obvious cause of obstruction. Sporadic/simple LECs (SLECs) of the parotid gland have no

association with underlying disease or with either Sjögren syndrome or HIV infection [3, 4]. These two histological types usually do not recur or metastasize. However, in the case of HIV infection or long-standing Sjögren syndrome, in which benign lymphoepithelial cysts are most often observed, a lymphoepithelial cyst can act as a precursor or harbor a MALT lymphoma [5–7]. It has been hypothesized that it may be an alternative histogenetic pathway of Warthin tumor, and due to the similarities between these two entities, differential diagnosis can be challenging [1, 3]. On MRI, cystic parotid tumors usually contain a mixture of solid and cystic components, while SLECs are typically unilocular cystic lesions without solid components. However, Warthin tumor can produce purely cystic lesions, and differentiating them from SLECs can be difficult [4]. Both lesions show low signal on T1-WI and moderate-to-high signal on T2-WI; nevertheless, in the case of a lymphoepithelial cyst heterogenous peripheral enhancement after contrast administration can be observed [3]. In our case, 66 year-old Caucasian male with a 30 pack-year smoking history came to our attention with a swelling in the left parotid gland; medical history was negative for prior radiotherapy, immunosuppression (such as HIV infection), autoimmune

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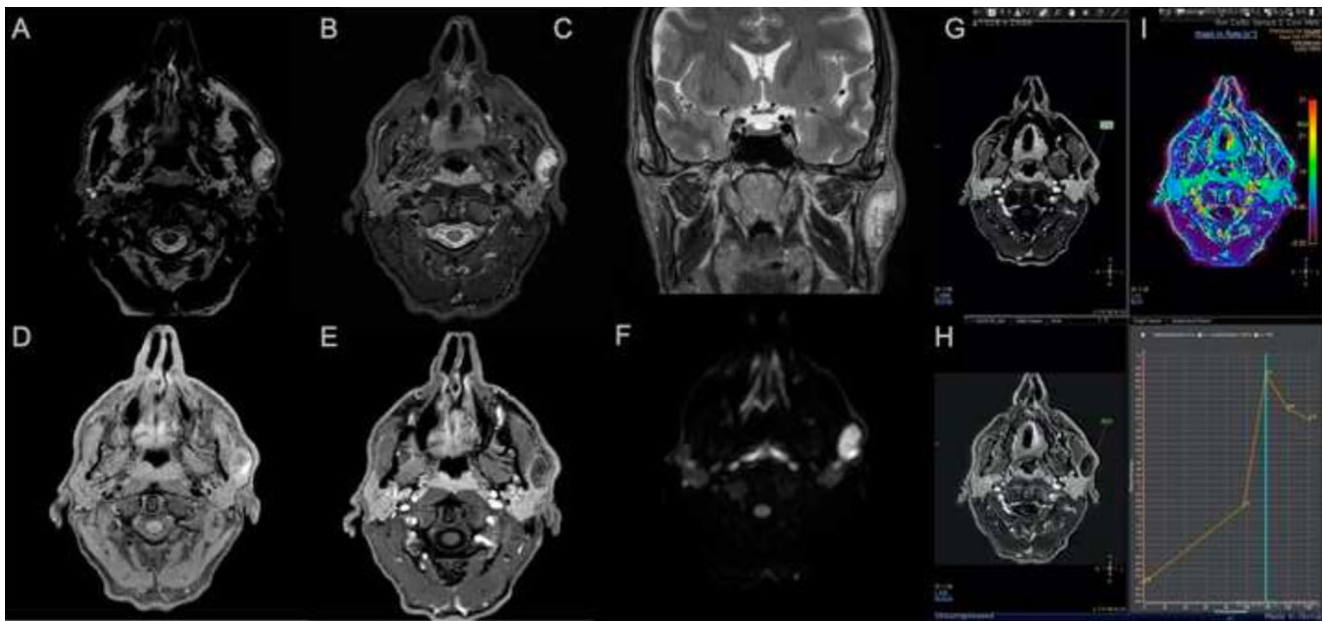


Fig. 1 MRI demonstrates an ovoid lesion located within the superficial lobe of the left parotid gland, consisting of a predominantly fluid component with a solid mural nodule along the lateral wall. The lesion appears hyperintense on axial and coronal T2-weighted images (A–C) and on the T2 fat-suppressed sequence (B). Its features are further

illustrated on pre-contrast T1-weighted imaging (D), post-contrast T1-weighted imaging (E), and on the DWI sequence (F). Dynamic contrast-enhanced T1-weighted imaging shows the enhancement curve of the lesion's nodular component (G, H), characterized by rapid wash-in followed by wash-out (I)

diseases, or occupational exposure. MRI showed an ovoid lesion with benign characteristics located within the superficial lobe of the left parotid gland, consisting of a predominantly fluid component with a solid mural nodule along the lateral wall (Fig. 1). Due to localization (superficial lobe) and anamnesis, a Warthin tumor was suspected, and FNA was considered superfluous. FNA is not always conclusive because the material may be insufficient; furthermore, in the case of a Warthin tumor, FNAC may be responsible for an inflammatory flare, with pain and an increase in the volume of the lesion [8]. The patient underwent enucleation with preservation of the peripheral branches of the facial nerve. The post-operative period was uneventful; nevertheless, definitive histopathological examination was consistent with a lymphoepithelial cyst with reactive lymphadenopathy. Six months after surgery, no recurrence of the disease has been observed, and the patient was prescribed an annual follow-up with ultrasound.

There are no fully reliable, standalone objective MRI features that can definitively separate a solitary, purely cystic lymphoepithelial cyst from a heavily cystic/necrotic Warthin tumor [4].

Effectively, FNA biopsy can be useful in some cases by revealing the presence of inflammatory cells in a proteinaceous background, but it remains nonspecific. On the other hand, the distinction is not fundamental, since surgical treatment for lymphoepithelial cyst and Warthin tumor is identical: partial conservative parotidectomy in case of tumor

arising in the context of the parotid gland, or enucleation if the tumor arises in the superficial lobe. Both lesions can present as unique or multifocal; the main differences relate to risk factors, while surveillance strategies are the same [8–10]. In this case, MRI characteristics and superficial lobe location enabled definitive surgical planning without FNA, and imaging provided sufficient information for surgical planning. Prospective correlation studies may determine whether FNA can be safely deferred in select cases.

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Declarations

Conflict of Interest All authors declare that this is an original article have no conflicts of interest. No financial funds have been received.

Ethical Approval All Authors declare that they have no financial interests. All procedures involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent has been obtained by the patient.

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