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CASE REPORT

Adenocarcinoma arising in a Warthin's tumor

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KEYWORDS

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Summary Carcinoma ex pleomorphic adenoma is a well-recognized entity, while, in rare cases carcinomas may arise from the epithelial component of Warthin's tumor. We present a case of adenocarcinoma arising in a Warthin's tumor located in the left parotid gland in a 49-years-old patient. Chest X-ray, laboratory investigation and thyroid scintigraphy were normal. A ultrasonography and computerized axial tomography showed multiple nodules. A fine needle aspiration biopsy showed typical features of Warthin's tumor. The histology showed the presence of a metastatic adenocarcinoma, that was thyroglobulin and calcitonin negative. The patient underwent a total left parotidectomy, was carefully followed-up, and at a 7 years check-up visit no other primary malignant lesion has manifested.

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Introduction

Warthin's tumor accounts for about 15% of all parotid tumors.¹ Carcinoma ex pleomorphic adenoma is a well-recognized entity,² while, in rare cases carcinomas may arise from the epithelial component of Warthin's tumor.^{3–6} This transformation has been reported in about 30 cases.^{2,7–9} The transformation of the lymphoid component to

malignant lymphoma appears to occur more frequently.⁷ Malignant transformation was seen in 0.1% of Warthin's tumors in Nagao et al. series.² Pathogenesis of malignant transformation of Warthin tumor is unknown.² It must, however, be borne in mind that areas of focal squamous cell metaplasia are not unusual in Warthin's tumor.⁸ Epithelial malignancy in a Warthin's tumor exists in three forms: the most common is a coexistent separate neoplasm, the second is metastasis to the lymphoid component of Warthin's tumor, and the least common is a primary carcinoma arising from the ductal component of the tumor.¹⁰

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Due to the rarity of carcinomas arising from Warthin's tumor, the authors report an additional case.

Case report

A 49-years-old patient was seen in a surgical outpatient Department for a left laterocervical clinically palpable lymph node. Chest X-ray and laboratory investigation were normal. A thyroid scintigraphy was normal. Ultrasonography and computerized axial tomography showed the presence of multiple nodules located in the left parotid gland.

A fine needle aspiration biopsy showed typical features of Warthin's tumor with some cells presenting characteristics suspicious of malignancy. The lymph node was removed and the histology showed the presence of a metastatic adenocarcinoma, that was thyroglobulin and calcitonin negative. The patient underwent subsequently a total left parotidectomy. Microscopically, transitional zones from benign oncocytic to frankly malignant epithelium with pre-existing benign Warthin's tumor were present (Figs. 1 and 2). Salivary gland tissue with multiple foci of well-differentiated adenocarcinoma was present (Fig. 3). The patient underwent a complete and thorough work-up, and no other primary malignant lesions were found. The patient was carefully followed-up, and at a 7 years check-up visit no other primary malignant lesion has manifested. The definitive diagnosis was adenocarcinoma arising in a Warthin's tumor.

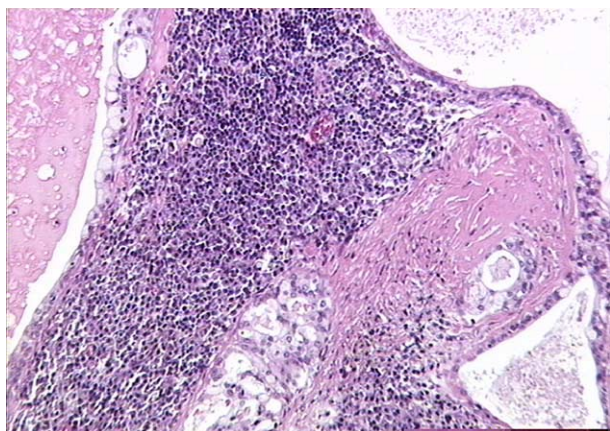


Figure 1 Presence of transitional areas from benign oncocytic to frankly malignant epithelium with lymphoid component (H&E 160x).

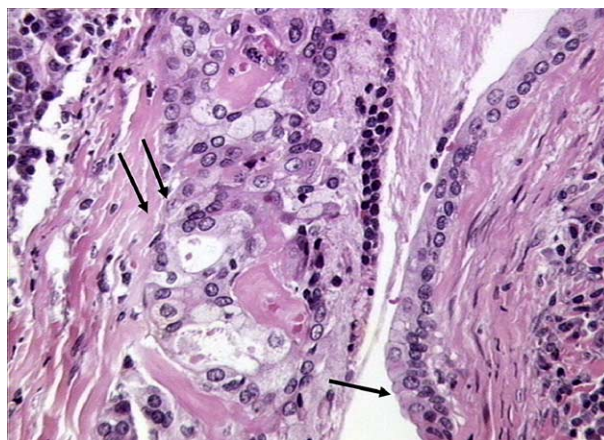


Figure 2 Presence of transitional zones from benign oncocytic (arrow) to frankly malignant epithelium (arrows) (H&E 250x).

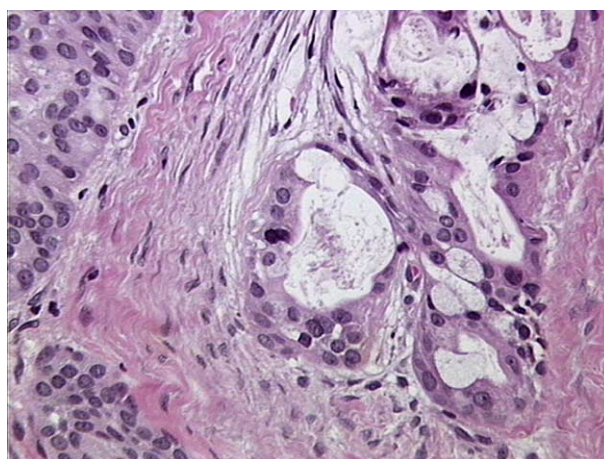


Figure 3 The adenocarcinoma is structured in well-differentiated glandular structures (H&E 400x).

Discussion

The diagnosis of malignant transformation of Warthin's tumor to carcinoma is based on the following criteria:⁷⁻⁹

- (1) presence of a pre-existing benign Warthin's tumor;
- (2) presence of transitional zones from benign oncocytic to frankly malignant epithelium;
- (3) presence of an infiltrating growth in the surrounding lymphoid tissue;
- (4) exclusion of metastasis to lymphoid stroma from an extrasalivary primary carcinoma.

The most common types of carcinomas ex Warthin's tumor are squamous cell carcinoma, mucoep-

idermoid carcinoma, oncocytic adenocarcinoma, undifferentiated carcinoma, and adenocarcinoma.^{2,3,7-9} Of the reported cases, 1/3 metastasized to regional lymph nodes and a single case presented distant hematogenous metastases to lung and liver.^{3,4,8}

A diagnosis of malignancy requires evidence of stromal invasion or local infiltrating growth or cervical lymph node metastases.^{2,8} Moreover, there should be a follow-up period without another primary tumor being identified. A continuous transition can be observed from typical oncocytic tumor cells to goblet cells, metaplastic squamous cells and neoplastic cells with the development of different types of carcinoma.⁸ The first step of the malignant transformation is the replacement of oncocytic cells by neoplastic cells with subsequent infiltration of the lymphoid tissue.⁸ A change in phenotype of cells through metaplasia to squamous cell differentiation is a well-known feature of Warthin's tumor,⁴ and may therefore explain the occurrence of epidermoid carcinomas and mucoepidermoid carcinomas.⁴ The occurrence of adenocarcinomas of variable differentiation in Warthin's tumor is not surprising since oncocytic tumours show glandular/tubular differentiation with formation of luminal surfaces containing microvilli.⁴ Due to the rarity of carcinoma ex Warthin's tumor, a metastatic carcinoma from another site must be excluded^{2,4} and the presence of other salivary gland tumors.^{4,5} Moreover, a prior or coexistent primary malignancy must be excluded.² Another differential diagnosis of carcinoma in Warthin's tumor concern "tumor-associated lymphoid proliferation" and florid squamous and mucous metaplasia in Warthin's tumor.²

Carcinoma in pre-existing cystadenolymphoma must be differentiated from:

- (1) metastases of another carcinoma in the lymphoid stromal component of cystadenolymphoma;
- (2) local coincidence of cystadenolymphoma and another coexistent, separate neoplasm;

- (3) carcinoma arising from the ductal component of cystadenolymphoma.¹¹

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