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Case report

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Poorly differentiated carcinoma arising from adenolymphoma of the parotid gland

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Abstract

Background: There is only one previous case report of a poorly differentiated carcinoma arising from an adenolymphoma of the parotid gland (Warthin's tumour). The absence of clinical symptoms, and the aspecificity of the radiological pattern make the diagnosis very difficult.

Case presentation: We here report the case of a 73-year-old man with Warthin's tumour who was brought to our attention because of a swelling in the parotid region.

Conclusions: In this case with an atypical clinical presentation, the intra-operative examination of a frozen section of the parotid mass allowed us to diagnose the malignant tumour correctly and consequently undertake its radical excision.

Background

Adenolymphoma (also called Warthin's tumour: WT) is a benign neoplasm of the salivary glands that almost exclusively occurs in the parotid gland, more often at the inferior pole, and usually affects white men aged 55 years. It has a classical composite histological appearance, with an epithelial component and a lymphoid stroma. The malignant transformation of WT is a very rare event. The lymphoid component may evolve into malignant lymphoma [1], whereas the epithelial component may develop into mucoepidermoid carcinoma [2], adenocarcinoma [3] or squamous cell carcinoma [4–7]. To the best of our knowledge, only Onder [8] has previously reported a case of poorly differentiated carcinoma of the parotid gland arising from WT.

Case presentation

The patient underwent a right superficial parotidectomy and, as an analysis of a frozen section showed the presence of poorly differentiated carcinoma cells, the surgeon extended the exeresis of the gland to the lower pole, preserving the facial nerve which was unaffected by the neoplasm. There were no right facial nerve deficits after the operation. The surgical specimen (3.5 × 2.5 × 1 cm) had a

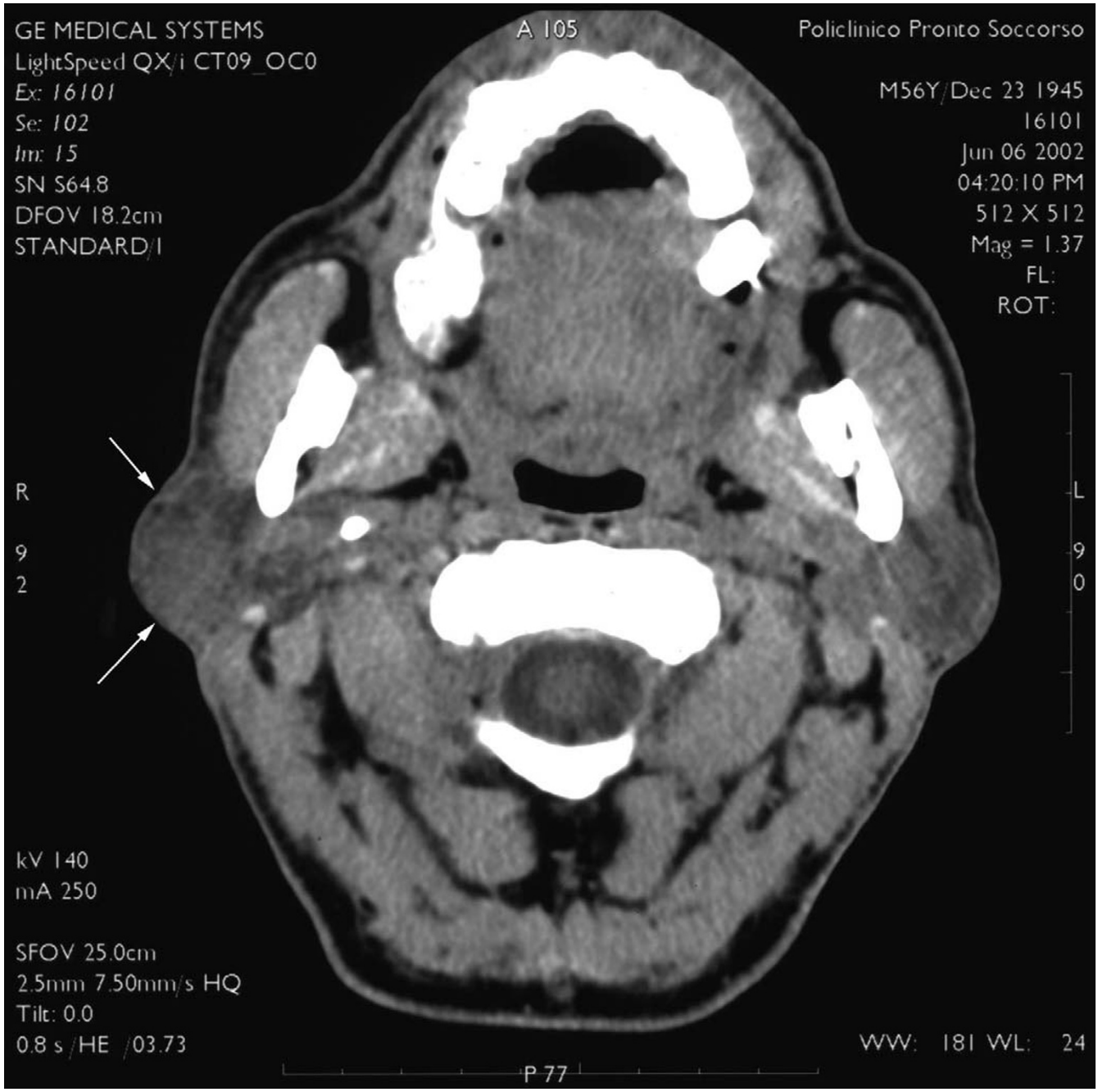

Figure 1
An axial computed tomography scan with contrast medium showing a 3.4 × 2.5 cm mass on the right parotid gland (arrows).

well-circumscribed, spherical, soft grey nodule with a few small cysts (a typical WT); the inner portion consisted of a harder yellow mass measuring 1.7 cm that proved to be a poorly differentiated carcinoma (Fig. 2).

A 73-year-old man was brought to our attention in January 2001 because of a swelling in the right parotid region, which had appeared three years before but had always been neglected by the patient because of its asymptomatic nature. It had apparently grown more rapidly over the previous two months, and a physical examination revealed

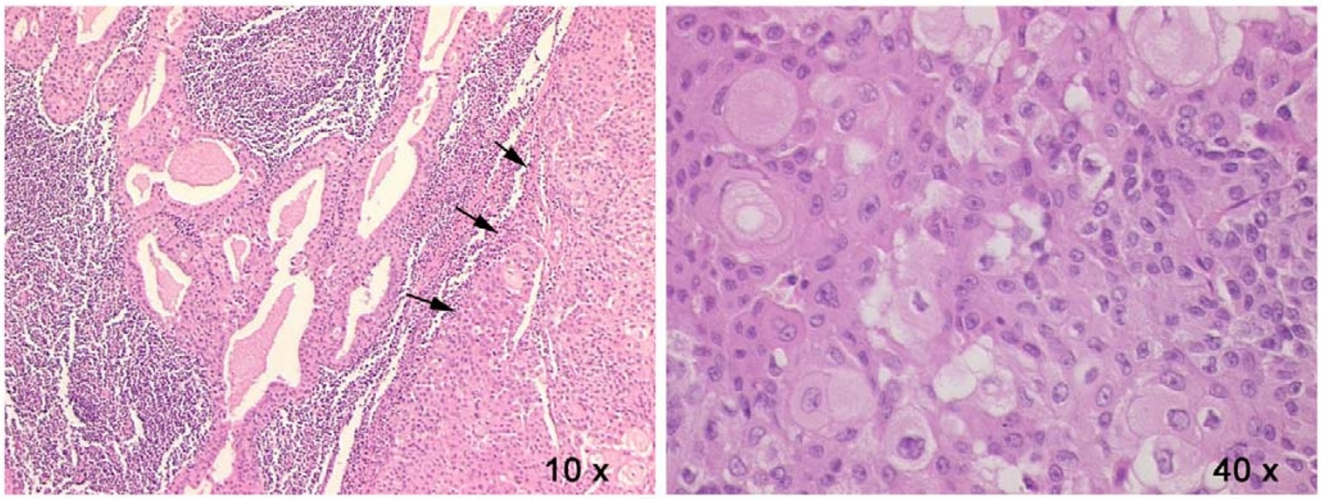

Figure 2
Left panel (enlargement 10×): undifferentiated carcinoma with poorly squamous differentiation (bottom right) very near to a typical Warthin's tumour (upper left). The arrows indicate the transition zone from a benign to malignant neoplasm. Right panel (enlargement 40×): detail of the malignant area. Hematoxylin and eosin stains.

the presence of a voluminous mass with a regular border that was mobile on the superficial and deep planes, and did not hurt upon palpation. There were no palpable cervical lymph nodes.

Three weeks before, the patient had undergone an ultrasound (US) examination of the neck and salivary glands, and a fine-needle aspiration biopsy revealed a mass on the superficial lobe of the parotid. The biopsy specimen suggested Warthin's tumour, but was negative for malignant cells. The computed tomography (CT) scan made using intravenous contrast medium confirmed the presence of a 3.4 × 2.5 cm mass on the right parotid gland. No pathological lymph nodes were identified (Fig. 1).

The grey nodule was histologically characterised by a prominent lymphoid matrix with germinal centres. The surface of this lymphoid tissue was covered by papillations of large epithelial cells forming cystic spaces with oncocytic features. The cells were arranged in two layers: the inner layer consisted of cuboid and polygonal cells containing nuclei with prominent nucleoli; the outer layer consisted of tall and sometime ciliated columnar cells, with centrally located pyknotic nuclei and a peripheral fibrous capsule. The oncocytic epithelial component inside the typical WT showed a hyperplastic-dysplastic change confluent with solid undifferentiated neoplasm (Fig 3a,3b); the malignant epithelial component, which was immunoreactive to an anti-cytokeratin pool (AE1-AE3; Dako), consisted of nests or single cells with a surrounding desmoplastic stroma sometimes infiltrated by small lymphocytes and plasma cells (Fig. 3b,3c). There were two types of intermitted cells loosing the papillary formation. The first were large cells with hyperchromatic nuclei, prominent nucleoli and thick chromatin along the nuclear membrane, abundant cytoplasm and well-demarcated cell membranes; no intercellular bridges or keratin production could be identified in these areas. The second were smaller eosinophil cells containing numerous eosinophilic granules, with oval nuclei, clumped chromatin and a centrally located eosinophilic nucleolus. The presence of mucin was excluded by PAS and PAS diastase staining of the paraffin blocks. Peripherically, small clusters of tumour cells penetrated the fibrous capsule and infiltrated the surrounding salivary gland.

The postoperative course was uneventful. The patient underwent postoperative radiotherapy because of the results of the histological examination. Now, 20 months after surgery, he is well and and has not experienced any recurrence.

Conclusions

Our patient with a poorly differentiated carcinoma arising in WT was asymptomatic and showed no signs, such as the clinical lymph node metastases or facial nerve palsies described by some authors [4,5]. The cytological results were not reliable as the fine-needle biopsy specimen of the parotid mass suggested Warthin's tumour but was negative for malignant cells. As the epithelial tumour

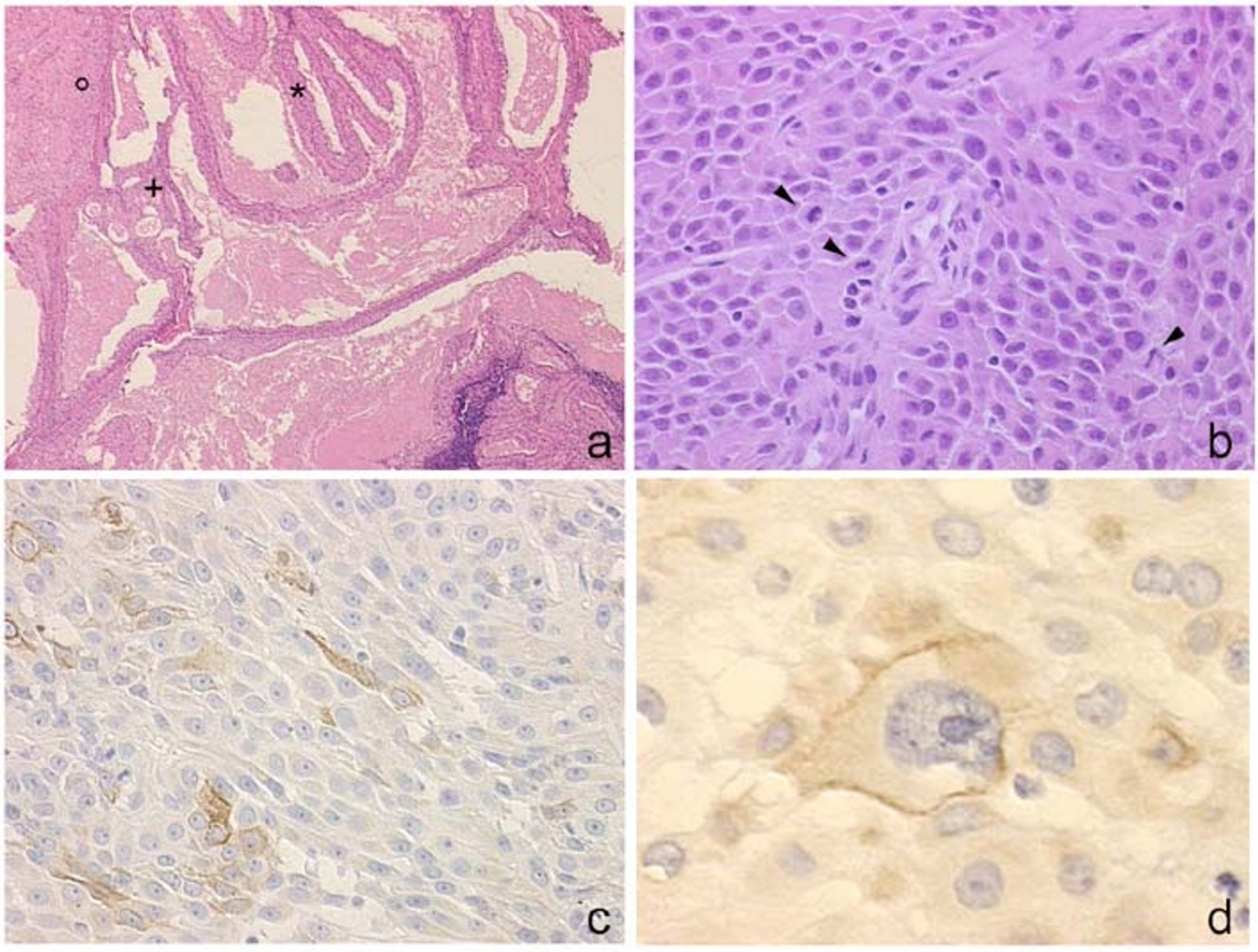

Figure 3
Upper panels, hematoxylin and eosin staining. Panel (a) shows a low resolution (5 ×) image of the oncocytic transition from typical papillae (asterisk) and a hyperplastic-dysplastic state (cross) to malignant transformation (circle). Panel (b) shows a 10× enlargement of the undifferentiated carcinoma with a few atypical mitoses (arrows). Lower panels, immunohistochemistry anti-cytokeratin staining: panel (c) shows a 10× enlargement of the undifferentiated carcinoma immunoreactive to cytokeratin, and panel (d) a detail (40×) of a very atypical cell with cytoplasmic anti-cytokeratin immunoreactivity.

component involved more than 50% of the neoplastic area, the case can be classified as subtype 1 (typical cystoadenolymphoma) according to Seifert *et al*. [9].

Our findings confirm the possibility of a malignant transformation of the epithelial component of a WT. It is worth underlining the diagnostic difficulty caused by the absence of clinical symptoms and the non-specific nature of the radiological signs; furthermore, false negative cytology for malignant cells is not a rare occurrence. In order to ensure a correct therapeutic approach, we recommend the use of intra-operative frozen section analysis in order to exclude malignant transformation when dealing with an atypical clinical presentation. Finally, from the histological point of view, we suggest considering a malignant transformation only when the bulk of the carcinoma is inside the WT and the oncocytic epithelial component simultaneously shows a transition zone from a hyperplastic-dysplastic state to malignancy.

List of abbreviations

Warthin's tumour = WT

Ultrasound = US

Computed tomography = CT

Competing interests
None declared.

Authors' contributions
LP, AP and GS enrolled the patient for clinical and surgical aspects. SF, LC, CA and PB did the histopathological examination and made the diagnosis. UC and MDS participated in the diagnosis and drafting the case report. MMC contributed to setting up the images and editing the case report.

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