CASE REPORT

Squamous carcinoma arising in a parotid Warthin's tumour

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SUMMARY

Warthin's tumour is the second most common benign neoplasm to affect the salivary glands. It virtually affects the sole parotid gland. A sudden increase in a tumour's size is usually due to a malignant transformation of the tumour. The transformation of the lymphoid stroma into malignant lymphoma is relatively common, while an epithelial malignancy is extremely rare. In this paper, the authors present a case of squamous cell carcinoma arising in Warthin's tumour. The patient underwent enucleoresection of the tumour. Intraoperative frozen section revealed the presence of a cystic component associated with the squamous cell carcinoma areas. In consideration of the result of the intraoperative consultation, the surgeons decided to enlarge the previous resection by removal of a 30×25 mm cuff from the surrounding parotid tissue. Close follow-up was carried out and 12 months after surgery there was no evidence of recurrence or metastatic neoplasm.

BACKGROUND

Warthin's tumour is the second most common benign neoplasm to affect the salivary glands. It virtually affects the sole parotid gland and account for 10% of all parotid tumours; they usually manifest as a slow-growing painless mass.^{1–3} This kind of neoplasia was first described by Warthin in 1929.⁴ It is more common in men and at an average age of 55 years.^{1 5}

Histologically, it is composed of epithelial and lymphoid elements; glandular and papillary cystic structures coexist with lymphoid tissue in the stroma. A sudden increase in a tumour's size is usually due to a malignant transformation of the tumour; the transformation of the lymphoid stroma into malignant lymphoma is relatively common, while an epithelial malignancy is extremely rare.⁵

The epithelial component may evolve into mucoepidermoid carcinoma,⁶ adenocarcinoma⁷ or squamous cell carcinoma.^{1 8 9} The first case of carcinoma arising from Warthin's tumour was described by Ruebner and Bramhall in 1960.⁷ In the literature, there are only two reports of Warthin's tumour transformation into poorly differentiated carcinoma⁹; 32 cases of epidermoid carcinoma arising in Warthin's tumours are described⁵ and 4 cases of squamous cell carcinoma as a malignant component have been listed by Therkildsen *et al.*¹⁰ In this paper, the authors will present a case of squamous cell carcinoma arising in Warthin's tumour.

CASE PRESENTATION

A 60-year-old Caucasian man presented with a slow-growing painless mass in the left preauricular region. He first noticed the swelling 3 years earlier, but did not visit a physician then because of fear of operation. Three months before his first visit to us, he noticed a moderate enlargement of the mass, which compelled him to enter our surgical department. Physical examination revealed an extensive non-tender mass in the left preauricular region, partly fixed to the underlying tissues. The skin over the mass was intact and movable. Neither facial paresis nor local lymphadenopathy were present. Intraoral examination showed no anomalies. The patient was otherwise healthy, with no history of current or previous diseases.

INVESTIGATIONS

Blood count, inflammation indexes and chest X-ray were normal. The patient underwent ultrasound examination that revealed a 50×45 mm solitary tumour mass in the left parotid gland. MR revealed a 46×47 mm lesion with well-defined margins localised in the left parotid gland ([figure 1](#)). Fine-needle aspiration cytology (FNAC) was performed; no malignant cells were found; the cytological features were consistent with Warthin's tumour.

DIFFERENTIAL DIAGNOSIS

Differential diagnosis should include all benign and malign tumours of the parotid gland, including metastases from other primary tumours. The gold standard in obtaining the correct differential diagnosis is the histopathological examination.

TREATMENT

Under general anaesthesia, the patient underwent an enucleoresection of the tumour. The resected mass, measuring 45×48 mm, was macroscopically cystic and solid ([figure 2](#)). An extemporaneous histological examination revealed the presence of a cystic component associated with areas of squamous cell carcinoma. In consideration of the results of the intraoperative consultation, the surgeons decided to enlarge the previous resection and a 30×25 mm cuff from the surrounding parotid mass was removed. The facial nerve was identified and entirely conserved ([figure 3](#)). A definitive histological examination confirmed the presence of Warthin's tumour associated with some areas of low-grade and high-grade dysplasia and squamous cell carcinoma. Resection margins of the first specimen appeared free of neoplasia. The second

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Figure 1 Preoperative MRI showing a 46×47 mm lesion with well-defined margins localised in the left parotid gland.

specimen was analysed: it presented some areas of squamous metaplasia without signs of malignancies.

OUTCOME AND FOLLOW-UP

The postoperative course was uneventful. Close follow-up was carried out and 12 months after surgery there was no evidence of recurrence or metastatic masses.

DISCUSSION

Malignant tumours arising from pre-existing Warthin's tumours are extremely rare. Batsakis *et al*² estimated their incidence to be 0.3% of all Warthin's tumours.

Warthin's tumour, also called adenolymphoma, was first described by Warthin in 1929. Adenolymphoma usually presents with a cystic pattern within a lymphoid stroma. Malignant evolution generally originates from the lymphoid component, while an epithelial malignancy is extremely rare and often associated with a mucorepidermoid carcinoma.⁵

The pathogenesis of the malignant transformation of Warthin's tumour is unknown.⁷

Figure 2 Intraoperative picture showing the parotid mass resected, which macroscopically appeared as a cystic and solid lesion.

Figure 3 The facial nerve was entirely conserved at the end of surgery.

In 1983, Damjanov and colleagues supposed that squamous cell carcinoma in Warthin's tumours arose from a focus of squamous metaplasia.¹¹ In our patient's specimens, pathologists could identify all the three stages of degeneration: Warthin's tumour, squamous metaplasia and squamous cell carcinoma. Damjanov postulated that the transition from the first stage, characterised by the presence of cylindrical cells with keratin cytoskeleton rich in mitochondria, to the last stage, that of squamous cells with few cytoplasmic organelles, may be due to infection, trauma or a large tumour causing ischaemia and necrosis. Ischaemia was thought to be the most probable aetiology for squamous metaplasia.¹

A malignant transformation is usually clinically suggested by a recent rapid enlargement of the mass.

Our patient did not present distant or local metastasis despite some authors reporting metastasis localised at regional lymph nodes as a common manifestation of this kind of tumour. Distant metastases are thought to be extremely rare.³

Other benign and malign tumours of the parotid gland have to be taken into consideration for differential diagnosis. Metastatic tumours to the lymphoid stroma of Warthin's tumour should be considered because cutaneous cell carcinoma and melanoma of the head and the scalp represent the metastatic malignant tumours most often found in the parotid lymph nodes.¹¹

In our patient, the absence of any other localisation of the tumour and the presence in the specimen of epithelial as well as lymphoid components confirmed that our carcinoma arose from Warthin's tumour. Moreover, transitional zones from benign oncocyctic to frankly malignant epithelium were found in our specimen.

The histopathology of Warthin's tumour is highly distinctive and FNAC fails to detect the lesion in up to 20% of cases.⁵ On the other hand, FNAB has good overall accuracy for diagnosing salivary gland neoplasm (87–97%).⁷

Learning points

- ▶ Besides polymorphous adenomas of the salivary glands, Warthin's tumours can also, rarely, degenerate into malignancies.
- ▶ Warthin's tumour degeneration is most often lymphatic, but epithelial degeneration should also be kept in mind.
- ▶ Patients who decide to opt out of surgery for small or deep Warthin's tumours should be advised to perform a periodical echographic or MRI evaluation in order to promptly identify degenerations.

Surgery represents the gold standard in treatment of this kind of tumour: adequate excision of the neoplasm with safe margins, coupled with neck dissection, should it be indicated, according to disease stage.^{12 13}

The long-term prognosis of these patients is not yet clear.^{7 14}

In our case, we did not discover any distant or local lymph node metastasis and the patient was disease free 12 months after complete surgical resection.

Contributors FB performed the surgery and FA took care of the patient during the preoperative and postoperative period.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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