

A rare case of oncocytoma of the parotid gland with aggressive features and malignant potential: a case report

Abstract

We report a rare case of an oncocytoma of the parotid gland with borderline histological features meaning with possible aggressive behavior. A 45 year old male presented to the Otolaryngology department with a unilateral small swelling in the right parotid region. CT and MRI studies revealed a right parotid mass and moderate enhancement following contrast media injection. The patient was treated with superficial parotidectomy with complete tumor resection. Postoperative course was uneventful. Histological results revealed a rare case of an oncocytoma with possible malignant potential. Close clinical and radiological follow-up in this case is of paramount importance. This case is unique due to the few existing references in literature.

Keywords: oncocytoma, parotid gland, borderline features, malignant potential

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Nikolopoulos Elias,¹ Ploumidou Katherine,¹
Gazalidou Maria,² Giannakou Niki²

¹Department of Otolaryngology Athens Medical Center (Omilos Iatrikou Athinon) Greece

²Department of Pathology Athens Medical Center (Omilos Iatrikou Athinon) Greece

Correspondence: Katherine Ploumidou, Department of Otolaryngology Head and Neck Surgery Athens Medical Center (AMC), Distomou 11 Amarousio 151-25 Athens Greece, Greece, Tel +30693272273, Email catplou@yahoo.gr

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Introduction

Oncocytic neoplasms belong to a group of infrequent tumors of the salivary glands just about 0,5%-1,5% and according to WHO they are histologically categorized in three distinct types, to be specific oncocytosis, oncocytoma and oncocytic carcinoma.¹

Oncocytomas are scarce benign epithelial tumors that most commonly occur in adults between the sixth through the eighth decades of life. They are painless tumors and distribution between genders is equal.² Majority of tumors occurs in the parotid gland and in addition it is the most common salivary gland that contains these neoplasms at a percentage of 78-80% in its superficial lobe. Oncocytes may be found individually or in groups in minor salivary glands. They are also found in the pancreas, in the respiratory tract, in thyroid, in parathyroid, pituitary and adrenal glands. Those originating from minor salivary glands have less well defined borders and are not encapsulated.² Alternative designations for the tumor are oncocytic adenoma, oxyphilic granular-cell adenoma and oxyphilic adenoma.³

An oncocytoma is characterized pathologically as a well defined delineated encapsulated mass constituted of layers of oncocytes (small round nucleus, microgranular eosinophilic cytoplasm rich in mitochondria).⁵ The surface is usually pink to rust in color. The tumor usually has a granulated appearance due to the abundant and hyperplastic mitochondria, a characteristic which cannot be observed in other tumors. An oncocytoma does not contain any lymphoid tissue.²

The clinical traits of oncocytomas bear resemblance to those of other benign and low grade salivary gland tumors making clinical diagnosis very difficult and demanding. Patients characteristically present a solitary slow growing painless parotid mass and manifest usually on MRI imaging T1 and T2 hypointensity with homogeneous contrast enhancement.³ The recommended treatment of choice is thorough

surgical excision either with superficial or total parotidectomy. The range and dimension of excision is imposed by preoperative clinical, radiological examinations (CT and MRI) and intraoperative findings. In addition a follow-up MRI at 12 and 24 months is usually suggested in order to evaluate patient progression. Radiation therapy is not recommended because oncocytes are radioresistant.³⁻⁵

Case report

A 45 year old male was presented at the Otolaryngological Department with a small swelling in the right parotid region. No pain or signs of infection were present. Clinical examination revealed a well defined soft mass with no palpable cervical lymph nodes. CT and MRI showed a well circumscribed mass with a major probability of a benign tumor in the superficial lobe of the right parotid gland. The patient underwent superficial parotidectomy with complete tumor excision and preservation of the facial nerve under general anaesthesia.

Pathological examination of the tissue sections revealed a 1,5 cm circumscribed neoplasm with tan color partially encapsulated, covered by healthy parotid tissue. A lymph node 2,1 cm was also present but had non specific reactive changes. The neoplasm had a nested pattern of growth suggesting an oncocytoma. It composed of cells with vesicular nuclei, with prominent nucleoli and abundant granular acidophilic cytoplasm. (Figure 1) (Figure 2). Enhanced mitosis, cytological atypia, moderate nuclear pleomorphism and necrosis were noted. Focally an abnormal circumference with connective tissue and fibrous elements was also present suggesting that this neoplasm has aggressive histological features with malignant potential. Immunohistological staining pattern showed that the neoplastic cells had a Ki67 positive at 8% (Figure 3), positive for ker 8/18 (Figure 4), positive-negative for p63 while were negative for ker 14, SMA, CEA, TTF-1, CD31, calcitonin, EMA, GFAP, and S100 staining.

This is not a typical case of an oncocytoma but an oncocytic neoplasm of uncertain malignant potential. The diagnosis of an oncocytic carcinoma was excluded due to insufficient presence of major diagnostic criteria for malignancy.

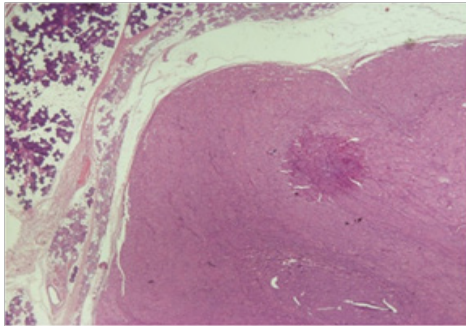


Figure 1 Ex 2.5: A circumscribed neoplasm covered by healthy tissue.

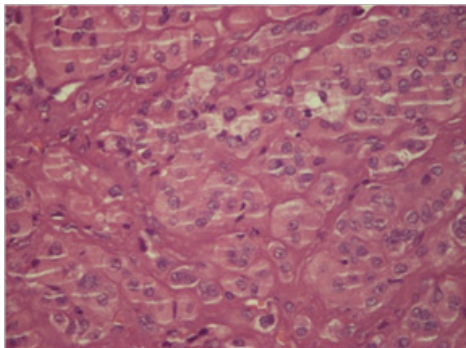


Figure 2 HEx 40: Nested pattern of neoplasm composed of cells with vesicular nuclei and abundant granular acidophilic cytoplasm. Vesicular nuclei with prominent nucleoli.

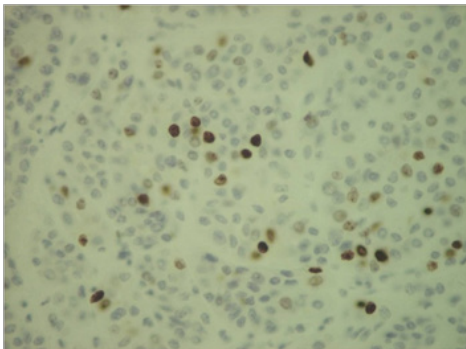


Figure 3 Ki67: Positive at 8% of neoplastic cells.

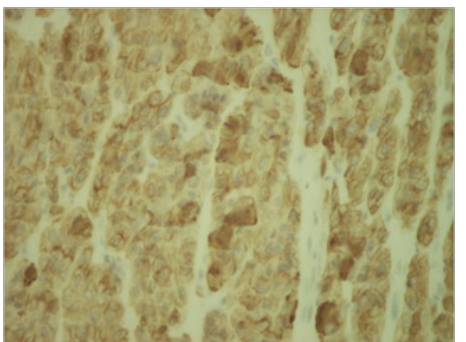


Figure 4 Ker 8/18: Positive at the neoplastic cells.

Discussion

Oncocytomas chiefly affect the parotid salivary gland. They are usually limited and delineated with a size of over 3cm.⁶ Their growth rate is typically slow. Although malignant transformation and local recurrence of oncocytomas is unusual and uncommon, clinical follow-up is very important.⁵ They are composed of layers of oncocytes, epithelial cells which exhibit specific cytological characteristics namely a central pycnotic nucleus and a granular abundant eosinophilic cytoplasm filled with numerous mitochondria of various sizes. Diagnosis by fine needle aspiration cytology (FNAC) may be very difficult and sensitivity in detection of oncocytic neoplasms is reported to be only 29%.^{5,7,8} Even though FNAC has been used as a primary screening tool for salivary gland lesions with high levels of sensitivity and specificity, salivary glands do have the tendency for having overlapping morphological features, therefore differentiating oncocytoma from oncocytic carcinoma can be difficult on cytology as the former can appear cytologically malignant looking, while the latter can look deceptively benign. Furthermore, differentiating oncocytosis from oncocytoma in cytology is difficult not to mention unattainable. Histopathology still remains the gold standard to presume the precise diagnosis.⁷ Specifically the nature of the tumor cells is usually confirmed with special methods, such as histochemical or immunochemical stains. Presence of pleomorphism, necrosis, perineural spread, frequent mitosis, intravascular invasion and distant metastasis to cervical nodes, kidneys, lungs and mediastinum as well as extensive invasion and destruction of adjacent structures are major signs of a high grade malignant tumor.⁹

According to the WHO two criteria are necessary in order to clinch the diagnosis of oncocytic carcinoma. The first consists of the pathological recognition of tumor cells as oncocytes and the second consists of the pathological finding of local infiltration and metastasis and presence of connective tissue. Oncocytic carcinoma shows a greater mitotic reactivity, activity and more nuclear pleomorphism compared to a benign oncocytoma.¹⁰ It has also been reported that using an anti-mitochondrial antibody when performing immunohistochemistry can identify with high sensitivity and specificity the mitochondria on light microscopy. Oncocytic carcinomas pathologically display immunoreactivity for mitochondria and are characterised by atypical accumulation of these organelles. This atypical accumulation is a compensatory mechanism to intrinsic functional defects of the mitochondria. The defective mitochondria produce high amounts of reactive oxygen species which predispose to genome instability and cellular transformation. Management and treatment of oncocytic carcinomas is still elusive and not well established due to the low incidence of this tumor type. Current treatment consists of surgical intervention with facial nerve sacrifice followed by immediate grafting and radiotherapy as well as prophylactic neck dissection. Efficacy of radiotherapy still remains unclear.^{9,11}

In this specific case while the tumor exhibits cytologic atypia, abnormal circumference and increased mitotic activity, atypical mitotic and invasive features were not identified. Even though the major diagnostic criteria for malignancy were not satisfied, the presence of moderate nuclear pleomorphism and an abnormal circumference, indicate that the neoplasm is compatible with a tumor of borderline (uncertain) malignant potential. Due to the nature of this neoplasm, close clinical and radiological follow-up of the patient is of paramount importance. This is probably one of a few cases of

oncocytoma with borderline features and malignant potential that has been reported in literature.

Conclusion

In summary we present a rare case of oncocytoma with aggressive features and malignant potential. Oncocytomas should be evaluated using CT and MRI studies in order to assess the extent of their presence but the final diagnosis should be confirmed histopathologically and by immunostaining. Further close follow-up of the patient is recommended and is of paramount importance.

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Conflict of interest

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