CASE REPORT

Squamous cell carcinoma arising in unilateral Warthin's tumor of parotid gland

Monika Sharma, Chintamani[1], Sunita Saxena, Usha Agrawal
Institute of Pathology, ICMR, [1]Department of Surgery, Safdarjung Hospital, New Delhi-110 029, India

Correspondence: *Dr. Usha Agrawal, Institute of Pathology, ICMR, New Delhi-110 029, India. E-mail: uburra@yahoo.com*

ABSTRACT
Warthin's tumor is a well-defined benign salivary gland neoplasm consisting of both epithelial and lymphoid components. Malignant transformation is extremely rare and the differential diagnosis of metastasis of an epidermoid carcinoma to Warthin's tumor is important. We present a case with squamous cell carcinoma arising in unilateral Warthin's tumor of parotid gland in a 16-year-old female patient.
Key words: Squamous cell carcinoma, Warthin's tumor, parotid gland

INTRODUCTION

Warthin described papillary cystadenoma lymphomatosum (or adenolymphoma) in 1929. It is a benign salivary gland neoplasm occurring principally in the parotid glands of men in the sixth and seventh decades of life. It accounts for 5 to 10% of parotid tumors.[1] The tumor consists of both epithelial and lymphoid elements. Warthin's tumor is usually asymptomatic or present as a slow-growing painless mass. Malignant transformation of the lymphoid component is relatively common. However, epithelial malignancy in Warthin's tumor is extremely rare, and exists in three forms, as epidermoid carcinoma, adenocarcinoma, and undifferentiated carcinoma.[2] Till date, only 31 cases of epidermoid carcinoma in Warthin's tumor have been reported. This case report presents a case of squamous cell carcinoma arising in Warthin's tumor in the parotid gland.

CASE REPORT

A 16-year-old female presented with a painless lump in right pre-auricular region. It was present for 2 years but had rapidly increased in size over the last 2½ months. On examination, a non-tender well defined lump, 5 x 2 cm in size was seen in the right pre-auricular region pushing up the right ear lobe, most probably arising from the right parotid gland. The lump was fixed to the deeper tissue. Full blood counts, biochemistry and chest x-ray were normal. The clinical impression was pleomorphic adenoma. As the patient couldn't afford MRI, right superficial parotidectomy was performed. The tumor measured 5 x 2 cm and showed both solid and cystic areas on gross examination. On cut section the cystic areas had a necrotic and hemorrhagic appearance. Salivary gland tissue was identifiable adjacent to the tumor. Histological sections from the cystic area stained with Hematoxylin and Eosin showed many dilated cysts filled with secretion and lined by double layered cuboidal oxyphilic epithelial cells arranged in a papillary growth pattern, the papillae projecting into cystic spaces [Figure 1]. The lining epithelium showed a transition zone with increased stratification as well as squamous metaplasia. The underlying subepithelium showed characteristic lymphoid tissue with dense infiltrate of lymphocytes forming lymphoid follicles. The solid areas revealed islands of malignant squamous epithelial cells invading the lymphoid stroma and cells showed atypical features with high N/C ratio, prominent nucleoli and keratinization [Figures 2 and 3]. Vascular embolus was also evident. Immunohistochemical staining for cytokeratin was positive in the Warthin's tumor cells, squamous metaplastic cells and squamous carcinoma cells [Figure 4], and common leukocyte antigen was positive in lymphoid cells [Figure 5]. A section from adjacent areas showed normal salivary gland and interglandular lymph node with features of reactive hyperplasia and no tumor metastasis.

Histopathological features were of Warthin's tumor with a component of squamous cell carcinoma. The patient got a full course of external beam radiation of the neck. A post-operative whole body CT scan showed no other mass or lymphadenopathy. The patient remained disease free on follow up for a further 12 months.

DISCUSSION

Warthin's tumor is a benign tumor exclusively restricted to parotid glands and periparotid lymph nodes. Most cases involve the lower pole although 10% are in the deeper lobe.[3] Warthin's tumor is multicentric in 12-20% of patients and is bilateral in 5-14%.[2] The morphological appearances are characteristic with cystic and solid areas, composed of epithelial and lymphoid component. The cyst and slit like spaces vary in size and shape and papillary structures project into the lumina.

The accepted management is enucleation of the tumor unless it is too close to the facial nerve to be safely enucleated.[4] In the

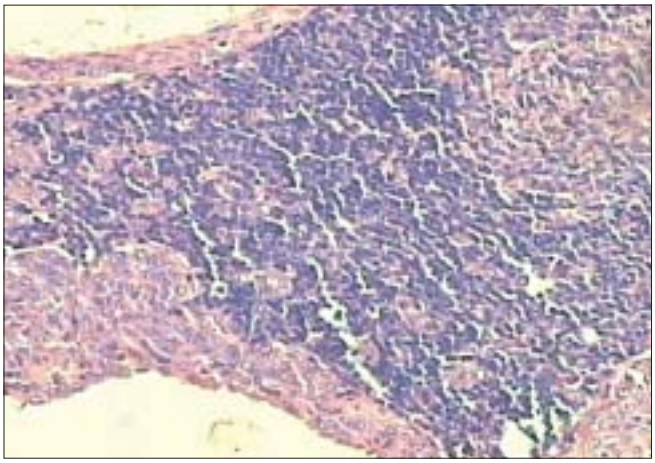

Figure 1: Warthin's tumor showing, lymphoid stroma and squamous metaplasia of lining epithelium in the lower cyst (H and E, 5x)

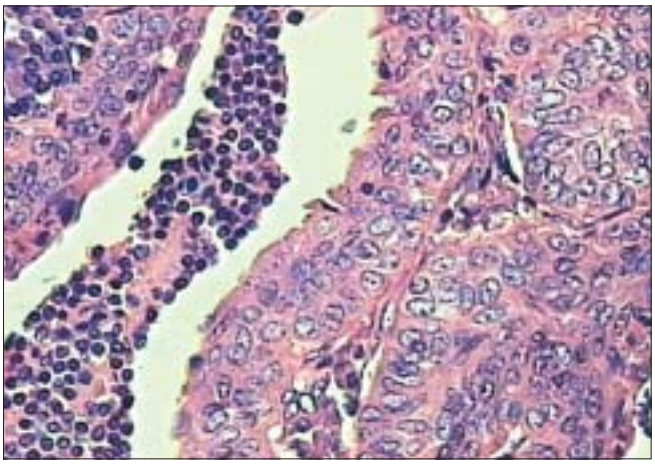

Figure 2: Transitional zone showing squamous metaplasia of the lining epithelium and invasion of squamous cells into lymphoid stroma (H and E, 10x)

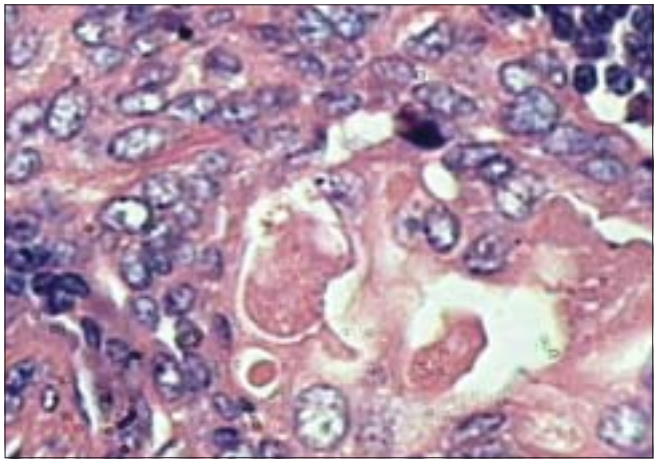

Figure 3: Squamous cell carcinoma with keratinization (H and E, 40x)

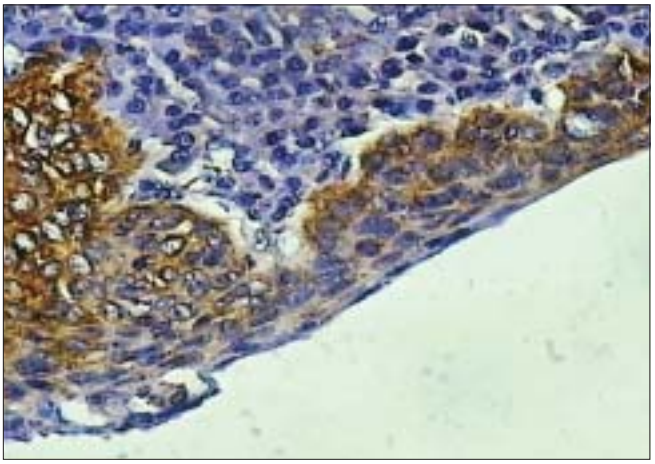

Figure 4: Cytokeratin expression in squamous metaplastic and squamous carcinoma cells (Cytokeratin, 20x)

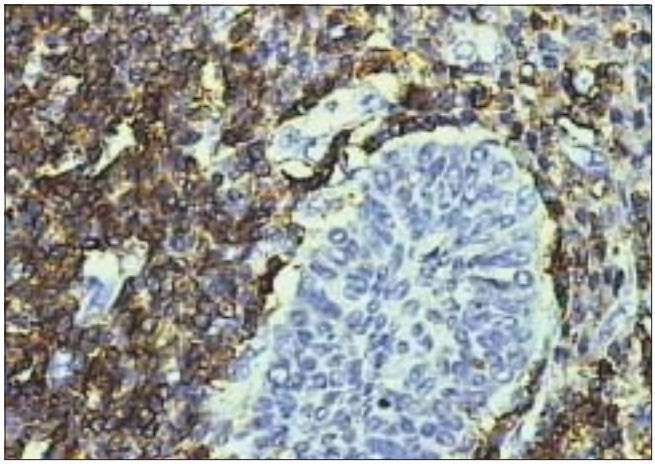

Figure 5: CLA expression in lymphoid cells around the tumor cells (common leukocyte antigen, 20x)

present case the patient underwent superficial parotidectomy as she was not suspected of having Warthin's tumor preoperatively. Most benign tumors of parotid are treated with this procedure.

Malignant tumors evolving from pre-existing Warthin's tumors are extremely rare and their incidence has been estimated to be 0.3% of all Warthin's tumor cases.[2] Until 1999, approximately 30 cases of epidermoid carcinoma arising in Warthin's tumor have been reported in the literature.[5] Damjanov *et al.*, reported a case and demonstrated on an ultrastructural level a gradual transition from cylindrical cells with keratin cytoskeleton and rich in mitochondria to flattened squamous cells with few cytoplasmic organelles. They postulated that squamous carcinomas arise from a focus of squamous metaplasia.[6]

The presence of keratin in the cytoskeleton of a Warthin's tumor cell may provide a clue to the ease with which these cells undergo squamous metaplasia.[7] Malignant change in a Warthin's tumor is suggested clinically by a recent history of rapid enlargement of a longstanding mass.[8] This is noted in the present case as well as in the patients described by McClatchey *et al.*[9] and Damjanov *et al.*[6] McClatchey *et al.*, reviewed the treatment and follow-up of seven cases of malignant change in a Warthin's tumor, of which only one showed evidence of

metastatic spread at any time. This patient was treated with surgery, adjuvant radiotherapy and chemotherapy. The other six cases were treated with surgical excision alone.[9]

The presence of transitional zones from benign oncocytic component to frank malignant epithelium is important to distinguish from metastasis of a primary carcinoma in another site.[6] Most of the reported cases with malignant transformation in Warthin's tumor did not show distant metastasis. However, regional lymph node metastasis was reported to be common. We did not discover either distant or lymph node metastasis in the present case, and she was disease-free at the 12-month follow-up. The long-term prognosis is not clear, however, as patients have been reported to be disease free after about one year's follow-up.

REFERENCES

1. Huvos AG, Paulino AF. Salivary glands. In: Diagnostic surgical pathology. In: Stenberg SS, editors. 3rd ed. Philadelphia: Lippincott Williams and Wilkins; 1999. p. 861-2.
2. Gunduz M, Yamanaka N, Hotomi M, Kuki K, Yokoyama M, Nakamine H. Squamous cell carcinoma arising in a Warthin's tumor. Auris Nasus Larynx 1999;26:355-60.
3. Simpson RH, Eveson JW. Warthin tumor. In: Barnes L, Eveson JW, Reichart P, Sidransky D, editors. Pathology and Genetics Head and Neck tumours. World Health Organisation Classification of Tumours. Lyon: IARC Press; 2005. p. 263-5.
4. Heller KS, Attie JN. Treatment of Warthin's tumor by enucleation. Am J Surg 1988;156:294-6.
5. Bolat F, Kayaselcuk F, Erkan AN, Cagici CA, Bal N, Tuncer I. Epidermoid Carcinoma Arising in Warthin's Tumor. Pathol Oncol Res 2004;10:240-2.
6. Damjanov I, Sneff EM, Delerme AN. Squamous cell carcinoma arising in Warthin's tumor of the parotid gland: A light, electron microscopic, and immunohistochemical study. Oral Surg Oral Med Oral Pathol 1983;55:286-90.
7. Therkildsen MH, Christensen N, Andersen LJ, Larsen S, Katholm M. Malignant Warthins tumor: A case study. Histopathology 1992;21:167-71.
8. Skalova A, Michal M, Nathansky Z. Epidermoid carcinoma arising in Warthin's tumour: A case study. J Oral Pathol Med 1994;23:330-3.
9. McClatchey KD, Appelblat NH, Langin, JL. Carcinoma in papillary cystadenoma lymphomatosum (Warthin's tumour). Laryngoscope 1982;92:98-9.

Source of Support: Nil, **Conflict of Interest:** None declared.