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Adenoid cystic carcinoma of the lacrimal gland

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ABSTRACT

Purpose: This is a retrospective analysis of the 50-year University of Florida experience treating adenoid cystic carcinoma of the lacrimal gland with radiation therapy.

Materials and methods: Between 1965 and 2015, 8 patients with adenoid cystic carcinoma of the lacrimal gland received radiation therapy with curative intent. Four patients received postoperative radiation therapy and 4 received definitive radiation therapy alone. The median follow-up was 3.3 years (range, 0.3 to 11.2 years).

Results: All 4 patients who received postoperative radiation therapy received 74.4 Gy. The 4 patients who received radiation therapy alone received a median dose of 72.3 Gy (range, 70.0 to 74.4 Gy). The overall survival rates at 5 and 10 years were 25% and 13%, respectively. The cause-specific survival rates at 5 and 10 years were 29% and 14%, respectively. The local control and freedom from metastases rates at 5 and 10 years were both 43%. Local recurrences occurred in 50% of patients, and distant metastatic disease occurred in 38% of patients. No patients experienced acute complications of treatment that warranted a treatment break. Two patients experienced bone exposure as late complications of treatment.

Conclusions: The results of this study illustrate the propensity for adenoid cystic carcinoma of the lacrimal gland to recur both locally and with distant metastases despite aggressive local treatment measures. This study also demonstrates the relatively poor outcomes for individuals with this type of tumor.

1. Introduction

Epithelial tumors are the most common type of lacrimal gland tumor, with the most common malignant subtype being adenoid cystic carcinoma [1–6]. Because of the rarity of this tumor, the lack of prospective studies analyzing its treatment, and the limited and mixed results of retrospective studies, the appropriate therapy for local control is under debate. Surgical resection with postoperative radiation therapy (RT) is favored at some institutions, while others recommend aggressive surgical resection alone [3,5,7–14]. Recent studies have analyzed the use of intra-arterial chemotherapy to either preoperatively decrease tumor size or postoperatively increase radiosensitivity of tumor cells; however, no consensus has been reached regarding this treatment approach [15–19]. Despite aggressive treatment, local recurrence and distant metastatic disease are common with adenoid cystic carcinoma of the lacrimal gland. As a result, the 5- and 10-year overall survival rates for adenoid cystic carcinoma of the lacrimal gland are poor [2,6,17,18].

This aim of this study is to update our experience treating patients with adenoid cystic carcinoma of the lacrimal gland with curative RT at the University of Florida.

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2. Materials and methods

Under an institutional review board-approved protocol, we reviewed the medical records of 8 patients with adenoid cystic carcinoma of the lacrimal gland who were treated in our department between December 1991 and March 2008 with curative RT. Patient characteristics are listed in Table 1. The cohort consisted of 2 men and 6 women with a mean age at presentation of 61.75 years (range, 36 to 82 years). Patients were staged according to the 7th edition of the American Joint Committee on Cancer (AJCC) staging system. All patients had a biopsy-proven diagnosis of adenoid cystic carcinoma of the lacrimal gland. Patients presented with the following symptoms: periorbital swelling (50%), diplopia (37.5%), proptosis (25%), ptosis (25%), new onset headaches (25%), decreased visual acuity (12.5%), and nasal obstruction (12.5%).

Of the 8 patients included in the study, 7 presented with a T4 tumor, and 1 presented with a T2 tumor. Only 1 patient had evidence of lymph node involvement and was deemed N1 at the time of treatment. No patients had evidence of distant metastatic disease at the time of treatment. Four patients were deemed surgical candidates and underwent surgical resection followed by postoperative RT. Three patients required orbital exenteration, and 1 received a wide local excision of the tumor with eye preservation. Three patients presented with tumors that were found to be incompletely resectable and underwent definitive RT alone. One patient was considered for surgical treatment; however, the patient declined resection for cosmetic reasons. Five patients had symptomatic clinical evidence of optic nerve involvement, 1 had asymptomatic incidental nerve involvement, and 2 had no nerve involvement. Only 1 patient received induction chemotherapy with cisplatin.

Patients receiving RT alone were treated to a median dose of 72.3 Gy (range, 70.0 Gy to 74.4 Gy). All 4 patients receiving postoperative RT were treated with 74.4 Gy. The mean interval time between surgery and RT was 42 days (range, 34 to 50 days). RT was administered twice daily at 1.2 Gy per fraction to 6 patients and once daily at 1.8 Gy per fraction to 2 patients. At the time of treatment, 1 patient was found to have a positive lymph node. This patient received a dose of 45.6 Gy at 1.2 Gy twice daily to the low neck. All patients received continuous course RT, without unscheduled breaks in treatment.

3. Results

The median patient follow-up was 3.3 years (0.3 to 11.2 years). At the time of analysis, only 1 of the 8 patients was alive, with a follow-up of 11.2 years. Overall treatment outcomes are listed in Table 1. The 5- and 10-year overall survival rates were 25% and 13%, respectively (Table 2). The cause-specific survival rates at 5 and 10 years were 29% and 14%, respectively (Table 2). The local control and freedom from distant metastasis rates at 5 and 10 years were identical at 43% (Table 2). Only 1 of the 4 patients treated with surgery plus RT experienced a local recurrence. However, 3 of these patients experienced distant metastases. Three of the 4 patients treated with RT alone experienced a local recurrence, with none experiencing distant metastases.

Table 1 – Patient Characteristic, Symptoms at Presentation, and Treatment Outcomes.

Patient	1	2	3	4	5	6	7	8
Year Treated	1991	1996	1998	2001	2003	2003	2004	2008
Age (years)	73	82	77	67	62	44	53	36
Sex	F	F	F	M	M	F	F	F
Race	White	White	White	White	Black	White	White	White
Symptoms at Presentation	Decreased visual acuity	Ptosis, diplopia	Proptosis	Periorbital swelling	Diplopia, headache, nasal obstruction	Headache, periorbital swelling, proptosis	Periorbital swelling, diplopia	Ptosis
T Stage	4	4	4	4	4	2	4	4
Lymph Node Involvement	None	None	None	None	None	None	None	N1
Nerve Involvement	Clinical	Clinical	Clinical	None	Clinical	None	Incidental	Clinical
Surgery	None	None	None	WLE	OE	None	OE	OE
RT Dose (Gy)	70.0	74.4	74.4	74.4	74.4	70.2	74.4	74.4
Fractionation	QD	BID	BID	BID	BID	QD	BID	BID
Chemotherapy	None	None	None	None	None	Intra-arterial cisplatin	None	None
Time to Local Recurrence (years)	3.7	1.4	1.92	–	–	–	–	0.9
Time to Distant Metastasis (years)	–	–	–	3.0	3.25	–	–	1.0
Location of Distant Metastasis	–	–	–	Bilateral lungs	Right lung	–	–	Widespread metastatic disease
Length of Follow-Up (years)	4.0	1.8	2.7	3.25	8.0	0.3	11.2	1.5
Late Side Effects	None	None	None	Bone Exposure	None	–	Bone Exposure	None
Status	DWD	DWD	DWD	DWD	DWD	DID	NED	DWD

Abbreviations: M, male; F, female; WLE, wide local excision; OE, orbital exenteration; QD, once daily; BID, twice daily; DWD, died of disease; DID, died of intercurrent disease; NED, no evidence of disease; N/A, not applicable.