Auris Nasus Larynx 42 (2015) 496–500

Contents lists available at ScienceDirect

Auris Nasus Larynx

journal homepage: www.elsevier.com/locate/anl

Primary adenoid cystic carcinoma of the nasolacrimal duct treated with proton beam therapy

Kota Wada *, Chiaki Arai, Toshihito Suda, Masato Nagaoka, Eiji Shimura, Sawako Yanagisawa, Hideo Edamatsu

Department of Otorhinolaryngology, Toho University, Tokyo, Japan

ARTICLE INFO

Article history:
Received 18 March 2015
Accepted 27 April 2015
Available online 18 May 2015

Keywords:
Adenoid cystic carcinoma
Nasolacrimal duct
Lacrimal passage
Proton beam therapy

ABSTRACT

Primary malignant tumors of the lacrimal passage, particularly of the nasolacrimal duct, are rare. We describe a 72-year-old woman who presented with lacrimation 5 years previously. She had pain and bloody and purulent lacrimation, and a mass was identified in the inferior meatus. Accordingly, she was diagnosed with primary adenoid cystic carcinoma of the nasolacrimal duct. She was treated with proton beam therapy and showed a favorable response. Owing to the long-term risks of recurrence and distant metastasis, adenoid cystic carcinoma requires sufficient follow-up.

1. Introduction

Adenoid cystic carcinoma is a malignant tumor, histologically characterized by cribriform structure formation and infiltrative proliferation [1]. In the head and neck region, adenoid cystic carcinomas frequently develop in the parotid gland and submandibular gland (the major salivary glands) and in the minor salivary gland in the mouth, and they are most commonly observed in middle-aged or older women [2]. Masses originating in the lacrimal passage frequently develop in the lacrimal sac [3], whereas primary malignant tumors of the nasolacrimal duct are rare. In fact, we have not come across any previous report of adenoid cystic carcinoma originating in the nasolacrimal duct. Herein, we report a patient with primary adenoid cystic carcinoma of the nasolacrimal duct who was successfully treated with proton beam therapy.

2. Case report

A 72-year-old woman presented with lacrimation in her left eye 5 years previously. She had received nasolacrimal duct irrigation at a nearby ophthalmologist. As she had experienced no changes in the symptoms or pain for 5 years, she had been followed conservatively with irrigation. Owing to exacerbation of symptoms including bloody purulent lacrimation, she wished to undergo dacryocystorhinostomy (DCR) and was referred to an ophthalmologist. Subsequently, a mass in the inferior meatus was diagnosed, and she was referred to the Department of Otolaryngology at our institution. There was no nasal congestion, nasal discharge, pain around the lacrimal sac and in the face, or facial swelling. At the initial visit, she had no lacrimal sac swelling or redness but had purulent discharge. Intranasal examination showed a soft, edematous mass near the opening of the nasolacrimal duct and the inferior meatus, as well as blood vessel dilatation and a bleeding tendency (Fig. 1). Computed tomography (CT) of the paranasal sinuses showed a mass, with the center in the nasolacrimal duct, extending toward the lacrimal sac. Although the lacrimal bone was partly lost, no distinct destructions of bone were noted. Magnetic resonance imaging of the paranasal sinuses showed enlargement of the nasolacrimal duct. A mass with relatively low signal intensity was noted on T1- and T2-weighted images; the center of the mass showed high signal intensity on the T2-weighted image, suggesting the presence of an internal aqueous component (Fig. 2). Fluorodeoxyglucose-positron emission tomography/CT (FDG-PET/CT) showed a mild accumulation corresponding to the site of the left nasolacrimal duct (maximum standardized uptake value = 2.1). There was no distinct accumulation suggestive of metastasis in the whole body, including the

* Corresponding author at: Department of Otorhinolaryngology, Toho University, 6-11-1 Omori-Nishi, Ota-ku, Tokyo 143-8541, Japan. Tel.: +81 3 3762 4151; fax: +81 3 3767 9877.
E-mail address: wadakota@med.toho-u.ac.jp (K. Wada).

http://dx.doi.org/10.1016/j.anl.2015.04.008
0385-8146/

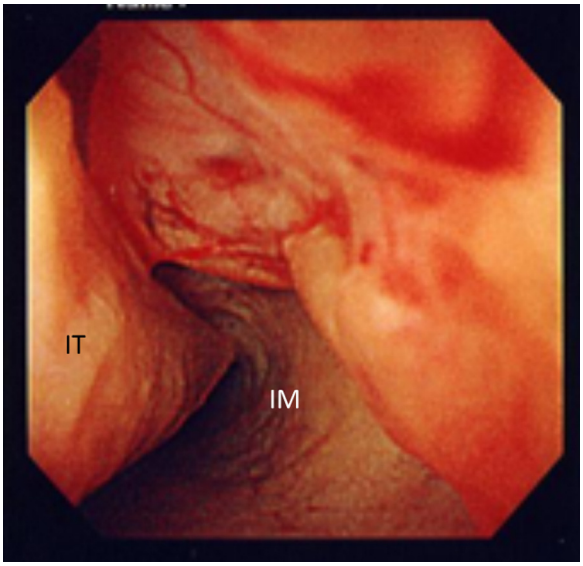

Fig. 1. An edematous mass in the inferior meatus. An edematous mass with blood vessel dilatation is observed at the opening of the nasolacrimal duct in the inferior meatus. IT: inferior turbinate; IM: inferior meatus.

lungs. The pathological examination of the tumor in the inferior meatus showed formation of a combination of small and large cystic spaces and epithelial cells with dysplasia forming a dense cribriform pattern. Accordingly, adenoid cystic carcinoma was diagnosed (Fig. 3).

Although tumor resection with lateral rhinotomy was recommended, the patient declined external nasal incision and wished to receive other treatment. After being informed about the risks and benefits of chemoradiotherapy, proton beam therapy, and CyberKnife therapy, she chose to undergo proton beam therapy. She started to receive 74.8 GyE of proton beam therapy from December 2013 (Fig. 4). Approximately 2 years have passed since the start of the treatment. At the latest follow-up, she showed atrophy and adhesion of the nasal tissues, likely associated with the proton beam therapy; however, she had achieved complete remission, as determined clinically and based on the imaging findings (Fig. 5). CT showed a shadow in the paranasal sinuses. Although secondary sinusitis was suspected, cancer recurrence could not be completely ruled out. Hence, endoscopic sinus surgery was performed to concurrently observe the ethmoid sinus and maxillary sinus internally. As suspected, the shadow was caused by secondary sinusitis. The patient is undergoing regular imaging studies, including head magnetic resonance imaging and chest CT and has not yet shown any signs of tumor recurrence or distant metastasis including brain and lung metastases.

3. Discussion

Nasolacrimal duct obstruction commonly occurs in elderly people; in most cases, it is caused by infection, traumas, and inflammation in the nasal cavity. Therefore, many patients visit ophthalmologists with a chief complaint of lacrimation, and receive conservative treatment with antimicrobial ophthalmic solution and a nasolacrimal duct bougie. Some patients undergo

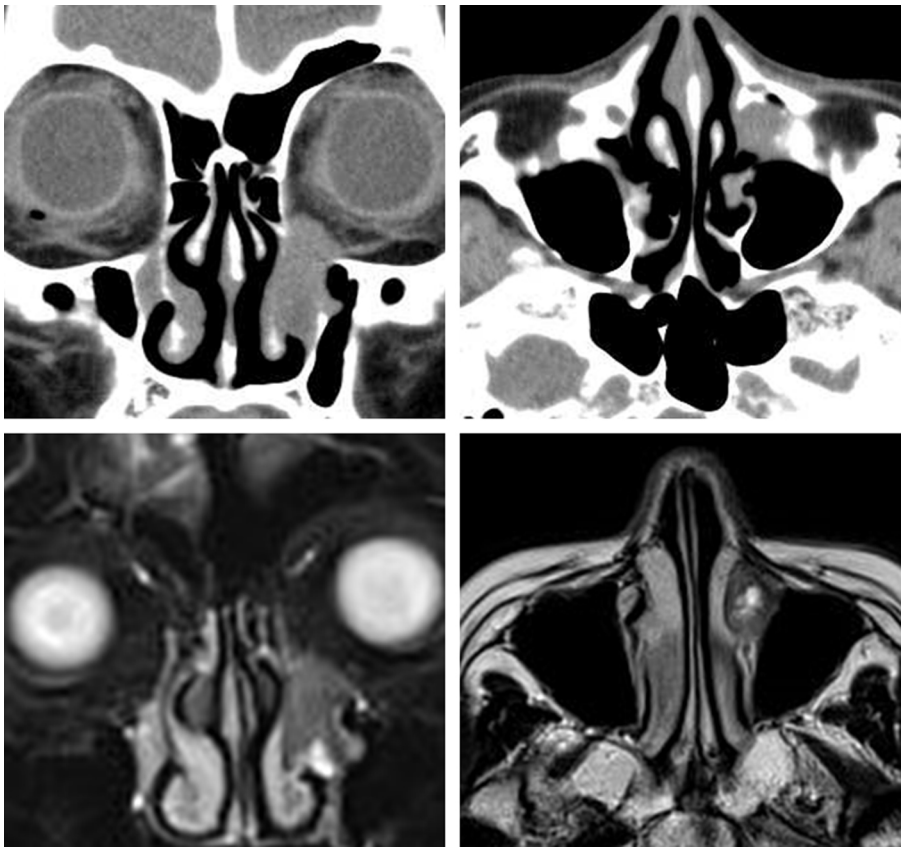

Fig. 2. Computed tomography (CT) and T2-weighted magnetic resonance imaging (MRI) of the paranasal sinuses (before treatment). A CT image in the coronal section shows a mass with its center in the left nasolacrimal duct, extending to the orbital cavity. The axial section image shows enlargement of the nasolacrimal duct as compared with the right. The MRI scan in the coronal section shows a mass with low intensity corresponding to the site of the nasolacrimal duct. The coronal section image shows a high-intensity area internally, suggesting the presence of an internal tear component.