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Review

Orbital invasion of ameloblastoma: A systematic review apropos of a rare entity

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Abstract

Purpose: Ameloblastoma is a non-encapsulated and slow-growing tumor with high recurrence rate. Orbital involvement by this neoplasm is an extremely rare entity. In this study, we present a systematic review on this situation along with clinical and paraclinical features of a case.
Methods: An electronic search was conducted on major medical sources. Data of the cases in the literature in addition to our own case were extracted, summarized, and statistically analyzed.
Results: A total of 36 other cases from 20 relevant studies were also reviewed. Review topics included epidemiology, clinical presentation, pathologic features, differential diagnosis, imaging, treatment, and prognosis. We provided a five-year history of a 50-year-old man with orbital/skull base invasion of plexiform maxillary ameloblastoma.
Conclusions: Maxillary ameloblastoma is a locally aggressive neoplasm, and physicians must be alert to the biologic behavior of this tumor to detect any invasion to critical structures such as orbit and cranium. Orbital ameloblastoma causes significant morbidity and mortality. We advocate meticulous patient follow-up with regular clinical examinations and paraclinical work-up for timely detection of any invasion or recurrence. The best must be done to avoid extensions by aggressive removal of maxillary ameloblastoma.

Keywords: Ameloblastoma; Eye; Vision; Orbit; Review

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Introduction

Ameloblastoma is a non-encapsulated, slow-growing tumor with high recurrence rate.[1−3] This neoplasm was first described in 1879 by Folkson, and the term 'Ameloblastoma' was first used by Churchil in 1933.[4] In the literature, there are some other terms used interchangeably with ameloblastoma such as 'cystosarcoma', 'adamantine epithelioma' and 'adamantinoma'.[5]

The origin of tumor is known to be remnants of odontogenic epithelium, lining of odontogenic cysts, and overlying mucosa.[1−3] The tumor arises from the mandible in approximately 80% of cases, mostly in association with an unerupted tooth. In addition, it may arise from the tuberosity of maxillary sinus in approximately 20% of cases.[6−8] Demographically, the reported age range of cases varies from 20 to 50 years with no sexual preponderance. Ameloblastoma rarely invades the orbit; if it does, it involves the orbit in the elderly with male predilection.[9]

Ameloblastoma is reported to be the most prevalent odontogenic tumor in our country, Iran.[10] However, to the best of our knowledge, there is no report of its invasion to the orbit either by the authors from Iran or other parts of the Middle East. The purpose of the study was to present the clinical and paraclinical features of a patient suffering from orbital ameloblastoma and an outline on previous reported cases. For the first time, we provide a systematic review on epidemiology, clinical aspects, pathology, prognosis, and current treatment modalities of this situation.

Methods

For the literature review process, a thorough electronic search was performed on the PubMed, Medline, Scopus, EMBASE, and web of science databases using the following keywords and terms: 'ameloblastoma', 'odontogenic tumors', 'orbital', 'ocular', 'eye', 'vision', and 'ophthalmic'. A reference list of eligible articles was also reviewed for possible eligibility. No limitation on publication date, study type/design, and language was applied.

Data of the cases in the literature in addition to our own case were further extracted and summarized based on the following items including age, sex, histopathology, initial location of the tumor, sites of extension, ophthalmologic sign and symptoms, management, recurrence, outcome, and disease duration. Furthermore, extracted data were descriptively analyzed in different histopathological categories for the following variables: age, age at diagnosis of ameloblastoma, origin, delay between diagnosis to invasion, location of extension, main ophthalmic presentation, delay between the last therapeutic modality and the date of recurrence and survival. Results were reported as mean ± standard deviation, maximum and minimum values, mode, median, and/or number (percent). Descriptive data analyses were conducted by IBM SPSS version 21.0 software (SPSS, Inc, Chicago, IL).

The study protocol and patient's ophthalmologic visits as well as orbital procedures were performed at Feiz Eye Hospital, located in Isfahan city, in the center of Iran. This university hospital serves as the referral center for at least four adjacent provinces. Our patient signed an informed consent for the publication of his disease data.

Results

Until now, thirty-seven (including our patient) cases with orbital invasion of ameloblastoma have been described in the literature. Previously, it has been proposed that ameloblastoma invades the orbit in 5th−6th decade of life[11]; in our literature review, the mean age of patients with orbital invasion of ameloblastoma was 52.79 ± 20.62 years, ranging from 7 to 81. Interestingly, the mean age of patients varied in different patterns; 51 ± 20.45 years in follicular; 59.5 ± 9.2 years in mixed; 62.8 ± 18.8 years in plexiform, and 63 ± 0.0 years in basal cell-like pattern. Furthermore, similar to other reports,[12,13] we found male preponderance, and the male to female ratio was 2.8:1. In none of the reported patients ameloblastoma developed primarily in the orbit, and all of the cases were secondary due to invasion from either maxillary or mandibular sinuses. The mean delay from onset of disease to orbital invasion was 12.7 ± 13.7 years. The mean delay of invasion varied in different patterns: 17 ± 16.9 years in follicular and 12.33 ± 15.37 years in plexiform. Almost always, invasion occurs unilaterally; however, two cases with bilateral invasion of tumor have been described.[12,14] The most common pattern of neoplasm was follicular followed by plexiform; however, the most prevalent tumor pattern in males and females were plexiform and follicular, respectively. According to patients' histories, the most common complaint at disease onset was decreased or loss of vision followed by proptosis. Most of the patients were managed surgically with or without chemotherapy or radiotherapy, and the mean age of survival was 13.47 ± 12.81 years.

In the literature, we found 20[1,11−29] studies describing 36 individuals with orbital invasion of ameloblastoma. The article publication dates ranged from 1934 to 2017. Table 1 shows age, sex, histopathology, initial location of tumor, sites of extension, ophthalmological signs and symptoms, management, recurrence and outcome of reported cases. Data of patients are further analyzed in Table 2.

Case Report

A 50-year-old man was referred to the oculoplastics service at the Feiz Eye Hospital in January 2014 for the evaluation of progressive inferior lid swelling and diplopia in down gaze. He was visited by a dentist in March 2011 for the extraction of upper third molar tooth. Two weeks following the extraction, a blister in his lingual vestibule adjacent to the extraction site appeared. The dentist evacuated the blister fluid with a surgical blade. Two weeks later, the patient was referred to a maxillofacial surgeon for the recurrence of the blister and computed tomography (CT) in April 2011, showing a mass in the maxillary sinus. At that time, the tumor was resected so that pathologic assessment revealed the diagnosis of plexiform ameloblastoma. Six months later in November 2011, the tumor

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