Dental Invasion by Adenoid Cystic Carcinoma of the Oral Cavity

Marco Guzzo, Pasquale Quattrone, Roberto Bianchi, Sarah Colombo
Department of Surgery, Maxillo Facial Surgery and Otolaryngology Unit, Fondazione I.R.C.C.S. Istituto Nazionale dei Tumori, Department of Pathology, Fondazione I.R.C.C.S. Istituto Nazionale dei Tumori, Milan, Italy

Abstract
Cancer progression through the mandible bone is still questioned. Once the tumor has eroded the cortex, it can extend vertically and laterally. It usually spreads in the medullary spaces and finally affects the alveolar nerve. The presence of the teeth has been considered as a relative barrier to cancer infiltration until now. Herein, we report an uncommon case of dental root invasion by adenoid cystic carcinoma of the floor of the mouth.

Keywords: Adenoid cystic carcinoma, oral cancer, salivary gland cancer

INTRODUCTION
Adenoid cystic carcinoma (ACC) accounts for <10% of all salivary gland neoplasms occurring in more than one-third of the cases in minor glands mainly in the oral cavity and sinonasal tract.

The tumors with solid growth patterns show a more aggressive clinical course. The 10-year survival rate ranges between 50% and 70%.

Mandible invasion by oral cavity cancer is a multistep process, better described for squamous cell carcinoma (SCC), and the presence of the teeth is believed to represent a relative barrier to tumor infiltration. Very few data were reported about this argument concerning ACC. To the best of our knowledge, we report the first case of direct tooth infiltration by ACC arisen in the oral cavity. The clinical value of this behavior is not clear, but it highlights the tumor aggressiveness and the questionable role of the teeth.

CASE REPORT
A 64-year-old man showed an infiltrative ulcerated neoplasm of the anterior floor of the mouth which developed about 7 years ago. The lesion had recently become painful, and the patient decided to refer to our department. A first examination showed that the tumor involved the ventral tongue and extended from the anterior floor of the mouth to the buccal vestibule through the mandible bone which appeared largely eroded in the incisal region. The mandible was dentated as showed in Figure 1. The teeth embedded into the alveolar process involved by cancer appeared firm. The tumor was biopsied, and a diagnosis of ACC was rendered. The clinical staging included head-neck magnetic resonance imaging [Figure 2] and total body CT (computerized tomography) scan. The cancer was classified as cT4aN0M0 (UICC 7th edition), and consequently, the patient underwent surgery. A wide resection of the anterior portion of the tongue, the floor of the mouth including a segmental bone resection from the left angle of the mandible to the contralateral premolar area was performed. The large defect was then reconstructed by means of microvascular osteocutaneous left fibular flap.

DISCUSSION
Since McGregor and McDonald,[1-2] many authors have investigated the patterns of invasion and the route of the tumor entry into the mandible by cancer of the floor of the mouth. There are two types of bone involvement by oral cancer.[3] The first one is an invasive/infiltrative pattern in which the tumor advances with fingers and islands into cancellous bone and finally affects the alveolar nerve. The presence of the teeth has been considered as a relative barrier to cancer infiltration until now. Herein, we report an uncommon case of dental root invasion by adenoid cystic carcinoma of the oral cavity.

Address for correspondence: Dr. Marco Guzzo, Maxillo Facial Unit and Otolaryngology, Fondazione I.R.C.C.S. Istituto Nazionale dei Tumori, Via Venezian, 1 Milan, Italy.
E-mail: marco.guzzo@istitutotumori.mi.it

This is an open access article distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 3.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

For reprints contact: reprints@medknow.com

How to cite this article: Guzzo M, Quattrone P, Bianchi R, Colombo S. Dental invasion by adenoid cystic carcinoma of the oral cavity. Ann Maxillofac Surg 2017;7:148-50.
ill-defined radiolucency of the mandible. In the erosive pattern, the bone recedes before a broad pushing tumor margin with an active osteoclastic activity. In this case, the tumor appears as a U-shaped or scalloped excavation on the bone cortex at X-ray image. The role of the teeth in conditioning the mandible infiltration is questioned. McGregor and MacDonald[2] and Bhattathiri et al.[4] suggested that tumors invading the dentate mandible tend to enter the bone through the periodontal membrane; other authors[5,6] such as Brown and Muller, found that the pathway to tumor entry was through the attached mucosa of the alveolar bone[6] in both dentate and edentulous mandible. Recently, Brown et al.[5] reported a large prospective series of 100 previously untreated patients with mandible infiltration by oral SCC. The authors concluded that tumor tends to invade the bone at the point of abutment both in dentate or edentulous mandible. None of these studies reported direct infiltration of the dental tissue by cancer. In the present case, the tumor penetrated the mandible cortex through the lingual aspect of the incisal region with an invasive pattern. Once penetrated into the cancellous bone, cancer widely invades the medullary spaces and finally the cementum and dentin of the dental root [Figures 3 and 4].

Cancer progression through the mandible bone has been investigated mainly for SCC which represents the most common histology among oral cavity malignancies. Few authors reported about ACC as in our present experience. Suei et al.[7] reported on ACC infiltrating the mandible without causing any marked destruction of the bone and few radiographic changes. Nevertheless, the author never described direct dental infiltration. Moreover, Al-Sukhun et al.[8] in a review of 16 cases of central intraosseous ACC of the mandible reported the case of a patient whose ACC was initially misdiagnosed as a periapical lytic/cystic lesion but teeth remained vital and not involved by cancer. Hence, to the best of our knowledge, this is the first report of direct tooth infiltration by ACC arisen in the oral cavity. The clinical significance of this finding remains unknown. Presumably, in the case of our patient, it could be related to the long-standing period before diagnosis (more than 7 years) of this high-grade tumor.

**Figure 1:** Preoperative orthopantomogram radiograph showing an irregular low-grade lytic lesion related to the incisal region of the mandible (arrows)

**Figure 2:** Preoperative magnetic resonance imaging showing cancer involving the tongue and infiltrating the mandible through the incisal region toward the soft tissue of the anterior vestibule (arrows)

**Figure 3:** Mandibular bone and dental root are invaded by adenoid cystic carcinoma with solid and cribriform pattern. The tumor widely invades the mandibular bone and focally the cementum and dentin (H and E, ×4)

**Figure 4:** Mandibular bone and dental root are invaded by adenoid cystic carcinoma with solid and cribriform pattern. The tumor widely invades the mandibular bone and focally the cementum and dentin (H and E, ×10)
Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

References
1. McGregor AD, MacDonald DG. Routes of entry of squamous cell carcinoma to the mandible. Head Neck Surg 1988;10:294-301.
2. McGregor AD, MacDonald DG. Patterns of spread of squamous cell carcinoma within the mandible. Head Neck 1989;11:457-61.
3. Cleary KR, Batsakis JG. Oral squamous cell carcinoma and the mandible. Ann Otol Rhinol Laryngol 1995;104:977-9.
4. Bhattathiri VN, Sudha L, Pillai BR, Sudhakaran A, Sasidharan K, Nair MK. Periodontal space: Major route to bone in oral cancer. Eur J Cancer 1991;27:222.
5. Brown JS, Lowe D, Kalavrezos N, D’Souza J, Magennis P, Woolgar J. Patterns of invasion and routes of tumor entry into the mandible by oral squamous cell carcinoma. Head Neck 2002;24:370-83.
6. Müller H, Slootweg PJ. Mandibular invasion by oral squamous cell carcinoma. Clinical aspects. J Craniomaxillofac Surg 1990;18:80-4.
7. Suei Y, Tanimoto K, Taguchi A, Wada T. Radiographic evaluation of bone invasion of adenoid cystic carcinoma in the oral and maxillofacial region. J Oral Maxillofac Surg 1994;52:821-6.
8. Al-Sukhun J, Lindqvist C, Hietanen J, Leivo I, Penttilä H. Central adenoid cystic carcinoma of the mandible: Case report and literature review of 16 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2006;101:304-8.