- 9. Singh N, Husain S, De Vera M, et al: *Cryptococcus neoformans* infection in patients with cirrhosis, including liver transplant candidates. Med Baltimore 83:188, 2004
- Munoz P, Giannella M, Valerio M, et al: Cryptococcal meningitis in a patient treated with infliximab. Diagn Microbiol Infect Dis 57:443, 2007
- Tuxen AJ, Yong MK, Street AC, et al: Disseminated cryptococcal infection in a patient with severe psoriasis treated with efalizumab, methotrexate and ciclosporin. Br J Dermatol 157: 1067, 2007
- Wilson ML, Sewell LD, Mowad CM: Primary cutaneous cryptococcosis during therapy with methotrexate and adalimumab. J Drugs Dermatol 7:53, 2008
- Lortholary O, Poizat G, Zeller V, et al: Long-term outcome of AIDS-associated cryptococcosis in the era of combination antiretroviral therapy. AIDS 20:2183, 2006
- Chiapello L, Iribarren P, Cervi L, et al: Mechanisms for induction of immunosuppression during experimental cryptococcosis: Role of glucuronoxylomannan. Clin Immunol 100:96, 2001
- Muslimani A, Francis NS, Gopalakrishna KV, et al: Disseminated cryptococcosis in a patient with idiopathic CD4+ T-lymphocytopenia. Clin Adv Hematol Oncol 6:446, 2008
- Busch MP, Valinsky JE, Paglieroni T, et al: Screening of blood donors for idiopathic CD4+ T-lymphocytopenia. Transfusion 34:192, 1994
- Centers for Disease Control and Prevention: Unexplained CD4+ T-lymphocyte depletion, persons without evident HIV infection. JAMA 268:1254, 1992

J Oral Maxillofac Surg 69:226-229, 2011

- Singh N, Gayowski T, Wagener MM, et al: Invasive fungal infections in liver transplant recipients receiving tacrolimus as the primary immunosuppressive agent. Clin Infect Dis 24:179, 1997
- Mehrabi M, Bagheri S, Leonard MK Jr, et al: Mucocutaneous manifestation of cryptococcal infection: Report of a case and review of the literature. J Oral Maxillofac Surg 63:1543, 2005
- 20. Prendiville S, Bielamowicz SA, Hawrych A, et al: Isolated cryptococcal sphenoid sinusitis with septicemia, meningitis, and subsequent skull base osteomyelitis in an immunocompetent patient. Otolaryngol Head Neck Surg 123:277, 2000
- Behrman RE, Masci JR, Nicholas P: Cryptococcal skeletal infections: Case report and review. Rev Infect Dis 12:181, 1990
- 22. Liu PY: Cryptococcal osteomyelitis: Case report and review. Diagn Microbiol Infect Dis 30:33, 1998
- 23. Warnke PH, Becker ST, Springer IN, et al: Penicillin compared with other advanced broad spectrum antibiotics regarding antibacterial activity against oral pathogens isolated from odontogenic abscesses. J Craniomaxillofac Surg 36: 462, 2008
- 24. Mylonas AI, Tzerbos FH, Mihalaki M, et al: Cerebral abscess of odontogenic origin. J Craniomaxillofac Surg 35:63, 2007
- 25. Saag MS, Graybill RJ, Larsen RA, et al: Practice guidelines for the management of cryptococcal disease, Infectious Diseases Society of America. Clin Infect Dis 30:710, 2000
- 26. Lanternier F, Lortholary O: Liposomal amphotericin B: What is its role in 2008? Clin Microbiol Infect 14:71, 2008 (suppl 4)

Temporomandibular Joint-Related Symptoms as Initial Presentation of Lung Carcinoma in a Patient With Takayasu's Arteritis

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Metastases to the temporomandibular joint (TMJ) are rare and have been described only in several case reports. We describe a case of 1 patient who had had Takayasu arteritis for 30 years who presented with TMJ-related symptoms (pain and swelling in the preauricular area). Additional examinations revealed a lung mass and liver metastasis. The liver biopsy specimen was diagnostic for lung carcinoma. We suggest

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that metastasis should be considered when evaluating a patient with TMJ-related symptoms. Metastases to the TMJ are also discussed and reviewed.

Report of a Case

A 49-year-old man was admitted to the emergency department because of pain and swelling in the left preauricular

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© 2011 American Association of Oral and Maxillofacial Surgeons 0278-2391/11/6901-0033\$36.00/0 doi:10.1016/j.joms.2010.04.023

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area of 1 month's duration. Before his referral, he had been treated with antibiotics, nonsteroidal anti-inflammatory drugs, and carbamazepine. Within the previous few days, he had also experienced chest pain, vomiting, and nasal bleeding.

The patient had been diagnosed with Takayasu arteritis at 17 years of age, for which he had been treated with low-dose oral steroids (10 mg prednisone). Five years before the



FIGURE 1. Left preauricular swelling (arrows). Tabib et al. TMJ-Related Symptoms and Lung Carcinoma. J Oral Maxillofac Surg 2011.

present admission, his ascending aorta had been replaced because of an aortic aneurism. In addition, he had developed ischemic heart disease and had undergone several heart catheterizations. His medical history also included a transient ischemic attack a few years before the present admission.

The physical examination revealed a nearly 3-cm swelling in his left preauricular area that was tender on palpation (Fig 1). He had no restriction in his mouth opening or lateral movements of the mandible. The intraoral examination revealed denture use, without any changes in occlusion or pain while palpating the area of his left lateral pterygoid muscle. An enlarged lymph node was palpated over his left clavicle. His laboratory tests revealed elevated lactate dehydrogenase (4,460 U/L) and abnormal levels of liver enzymes (aspartate aminotransferase -225 U/L, alkaline phosphatase -288 U/L, and gamma-glutamyl transpeptidase -670 U/L; alanine aminotransferase and bilirubin levels were within the normal range). A panoramic view revealed complete osteolysis of the left mandibular condyle (Fig 2). A computed tomography scan revealed a 3.5-cm mass that had replaced the condyle (Figs 3A-C), liver metastases (Fig 3D), enlarged chest lymph nodes, and a large mass at the left lung. An attempt to biopsy the mandibular mass was not successful. The liver biopsy specimen was diagnostic for small cell lung carcinoma. The patient started undergoing chemotherapy. Ten days later, he was hospitalized in the intensive care unit because of neutropenia and lung infection. However, a few weeks later, he died.

Discussion

Most oral malignancies are a primary neoplasm, and metastatic disease is unusual (1% to 8% of oral malignancies).¹ Metastasis to the TMJ is considered extremely rare and has been described only in several case reports. In a recent review of metastatic tumors to the oral and maxillofacial region, of 796 cases, 39 (13.8%) were at the condyle.² Primary cancers that have been reported to metastasize to the TMJ have included breast adenocarcinoma,^{1,3-5} renal cell carcinoma,⁶ testicular teratoma,⁷ prostate cancer,⁸⁻¹⁰ rectal carcinoma,¹¹ hepatocellular carcinoma,¹² and melanoma.¹³ Other primary cancers that have been reported have included uterine cancer, pancreas can



FIGURE 2. Panoramic view showing complete osteolysis of left TMJ *(circle).*

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FIGURE 3. Computed tomography scans of the temporomandibular joint showing the mass and osteolysis of the joint. A, Axial section; B, coronal section; C, 3-dimensional reconstruction; and D, coronal section demonstrating liver metastases. *Tabib et al. TMJ-Related Symptoms and Lung Carcinoma. J Oral Maxillofac Surg 2011.*

cer, and melanosarcoma.¹⁴ Two cases have been reported of metastatic spread to both mandibular condyles.^{1,7} Except for the present case report, metastasis of lung carcinoma has been reported only 3 times in English-language medical studies.¹⁴⁻¹⁶

Several explanations are possible for the low incidence of metastasis to this site, including the paucity of red marrow, the separate blood supply of the condylar portion of the mandible, and the presence of a limiting osseous plate.^{6,17} The difficulty in differentiating clinically between common TMJ dysfunction syndrome and metastatic involvement of the condyle might have contributed to the infrequent reports of metastasis to the TMJ.¹¹ The typical symptoms of

metastasis to the TMJ include severe TMJ pain, swelling, and trismus,^{1,3,6} which have appeared in 50% of cases.^{10,14} Rarely, it could manifest as a pathologic fracture.^{3,9,11} On physical examination, preauricular swelling and tenderness has sometimes been noted,^{7,15} with a severe opening limitation¹ or deviation of the mandible.^{6,7,11} The panoramic imaging findings will initially be normal,¹⁷ but might reveal a radiolucency of the TMJ,^{1,7,11} condyle erosions,^{1,15} or fracture of the condylar neck.⁹ Radiopacities were noted in 1 case of breast carcinoma metastasis.⁵

In several patients with metastasis to the condyle, TMJ-related symptoms were the presenting symptom of the tumor.^{8,14,15,17} In others, it was a complication of a known cancer,^{3,5,7,9,11} even 19 years after its first presentation.¹ Sometimes condylectomy is required to re-establish mandibular function and to assess the histopathologic nature of the lesion.^{5,11} Metastasis to the TMJ, an indication of widespread disease, has been associated with a poor prognosis (usually survival of several months).^{3,5,7,9,11,14,15,17}

In addition to the rare manifestation of lung carcinoma as TMJ syndrome, our patient had Takayasu arteritis, a rare vasculitis of the large vessels.¹⁸ Several reports have established the association of vasculitis, particularly of the small vessels, and different malignancies.¹⁹⁻²² In these cases, the vasculitis appears in a temporal relationship to the tumor diagnosis and represents a paraneoplastic syndrome. In the case of large-vessel vasculitis (usually giant cell arteritis), such an association has not usually been found,^{23,24} but other authors have found an association.^{25,26} However, the effect of long-standing vasculitis, specifically Takayasu arteritis, and the appearance of cancer should be better addressed.

We have presented a rare case of TMJ-related symptoms as the presentation of metastatic lung carcinoma. Although not directly proven, the mass at the TMJ was most probably metastasis of the lung cancer. It might be efficient to consider this rare possibility in the differential diagnosis when evaluating a patient with TMJ symptoms. Therefore, a complete examination, laboratory tests, and proper imaging are mandatory when examining patients with TMJ symptoms.

References

- Miles BA, Schwartz-Dabney C, Sinn DP, et al: Bilateral metastatic breast adenocarcinoma within the temporomandibular joint: A case report. J Oral Maxillofac Surg 64:712, 2006
- Shen ML, Kang J, Wen YL, et al: Metastatic tumors to the oral and maxillofacial region: A retrospective study of 19 cases in West China and review of the Chinese and English literature. J Oral Maxillofac Surg 67:718, 2009
- Webster K: Adenocarcinoma metastatic to the mandibular condyle. J Craniomaxillofac Surg 16:230, 1988

- Mizukawa JH, Dolwick MF, Johnson RP, et al: Metastatic breast adenocarcinoma of the mandibular condyle: Report of case. J Oral Surg 38:448, 1980
- Mace MC: Condylar metastasis from mammary adenocarcinoma. Br J Oral Surg 15:227, 1978
- Johal AS, Davies SJ, Franklin CD: Condylar metastasis: A review and case report. Br J Oral Maxillofac Surg 32:180, 1994
- Porter SR, Chaudhry Z, Griffiths MJ, et al: Bilateral metastatic spread of testicular teratoma to mandibular condyles. Eur J Cancer B Oral Oncol 32B:359, 1996
- Thatcher SL, Dye CG, Grau MJ, et al: Carcinoma of the prostate metastatic to the mandibular condyle mimicking a parotid tumor. J Oral Maxillofac Surg 44:394, 1986
- Catrambone RJ, Pfeffer RC: Significant postoperative hemorrhage following biopsy of a prostate tumor metastatic to the mandibular condyle: Report of a case. J Oral Maxillofac Surg 48:858, 1990
- deBoom GW, Jensen JL, Siegel W, et al: Metastatic tumors of the mandibular condyle. Review of the literature and report of a case. Oral Surg Oral Med Oral Pathol 60:512, 1985
- Giles DL, McDonald PJ: Pathologic fracture of mandibular condyle due to carcinoma of the rectum. Oral Surg Oral Med Oral Pathol 53:247, 1982
- Yoshimura Y, Matsuda S, Naitoh S: Hepatocellular carcinoma metastatic to the mandibular ramus and condyle: Report of a case and review of the literature. J Oral Maxillofac Surg 55:297, 1997
- Nortje CJ, van Rensburg LJ, Thompson IO: Case report: Magnetic resonance features of metastatic melanoma of the temporomandibular joint and mandible. Dentomaxillofac Radiol 25:292, 1996
- 14. Owen DG, Stelling CB: Condylar metastasis with initial presentation as TMJ syndrome. J Oral Med 40:198, 1985
- 15. Peacock TR, Fleet JD: Condylar metastasis from a bronchogenic carcinoma. Br J Oral Surg 20:39, 1982
- Gerlach KL, Horch HH, Feaux de Lacroix W: Condylar metastasis from bronchial carcinoma: Case report. J Maxillofac Surg 10:250, 1982
- Rubin MM, Jui V, Cozzi GM: Metastatic carcinoma of the mandibular condyle presenting as temporomandibular joint syndrome. J Oral Maxillofac Surg 47:507, 1989
- Johnston SL, Lock RJ, Gompels MM: Takayasu arteritis: A review. J Clin Pathol 55:481, 2002
- Garcia-Porrua C, Gonzalez-Gay MA: Cutaneous vasculitis as a paraneoplastic syndrome in adults. Arthritis Rheum 41:1133, 1998
- Mitsui H, Shibagaki N, Kawamura T, et al: A clinical study of Henoch-Schonlein purpura associated with malignancy. J Eur Acad Dermatol Venereol 23:394, 2009
- Hutson TE, Hoffman GS: Temporal concurrence of vasculitis and cancer: A report of 12 cases. Arthritis Care Res 13:417, 2000
- 22. Pertuiset E, Liote F, Launay-Russ E, et al: Adult Henoch-Schonlein purpura associated with malignancy. Semin Arthritis Rheum 29:360, 2000
- Huston KA, Hunder GG, Lie JT, et al: Temporal arteritis: A 25-year epidemiologic, clinical, and pathologic study. Ann Intern Med 88:162, 1978
- 24. Myklebust G, Wilsgaard T, Jacobsen BK, et al: No increased frequency of malignant neoplasms in polymyalgia rheumatica and temporal arteritis: A prospective longitudinal study of 398 cases and matched population controls. J Rheumatol 29:2143, 2002
- Haga HJ, Eide GE, Brun J, et al: Cancer in association with polymyalgia rheumatica and temporal arteritis. J Rheumatol 20:1335, 1993
- 26. O'Connor MB, Murphy E, O'Donovan N, et al: Takayasu's arteritis presenting as a dissecting aortic aneurysm history: A case report. Cases J 1:52, 2008