Malignant Lesions Presenting as Symptoms of Craniomandibular Dysfunction

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Three cases of carcinoma, seen in a head/neck pain management practice during the past 2 years and presenting as craniomandibular dysfunction symptoms, are reported. Clinicians should always consider a differential diagnosis because of the variety of potential causes of the symptoms presented by the patient. These symptoms act as clues to help to differentiate between a routine or "evil" process. The need for imaging and reimaging, when the clinical symptoms do not correlate with the clinical examination through established guidelines for craniomandibular dysfunction or head/neck pain, is of paramount importance.
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Pain, swelling, or dysfunction that is localized to the region of the craniomandibular structures can result from myriad sources. Appropriate evaluation of this area is made difficult not only by the many possible etiologies, but also by the frequency of this complaint. Specific findings meriting close attention in a patient with craniofacial pain include paresthesias, taste alterations, auditory or vestibular findings, and asymmetric tongue protrusion.

Neoplastic disease is not a common finding but has been the subject of several case reports.[1–7] Symptoms of rapid progression or those involving palpable nontender masses are also cause for concern. The American Academy of Orofacial Pain (formerly American Academy of Craniomandibular Disorders) has put forth these and other guidelines for the management of atypical craniofacial pain.[8]

Three cases of craniomandibular dysfunction (CMD) arising from malignancy are presented, along with a review of the literature. In addition to information obtained through a careful history, physical examination, and follow-up, the presented findings serve as factors which should alert the practitioner to the increased possibility that malignancy is the cause of an atypical craniofacial pain.

Case 1

A 58-year-old white woman, employed as a nurse anesthetist, was referred for a second opinion as to her CMD and head/neck pain symptoms. The past medical history included high blood pressure, rheumatic fever, heart murmur, palpitations, anemia, asthma, airborne allergies, glaucoma, dizzy spells, and hyperthyroidism. Her previous surgical experiences included a hysterectomy, tonsillectomy, and cataract surgery to the right eye.

This patient was in reasonably stable health until approximately 2 months prior to her first visit to a primary physician. She awakened with pain in the left jaw, which progressed both in severity and in area of radiation. Initially causing left otalgia, the pain later encompassed the entire left side of the face within the distribution of the trigeminal nerve. The patient had no prior history of jaw pain and was a nonsmoker. Her evaluation suggested pain at the insertion of the temporalis muscle on the left coronoid process, and she was referred to an oral surgeon. The temporalis insertion was injected with a steroid. She received diazepam and analgesics and was referred for stress counseling. This did not prevent the symptoms from worsening. The patient was then referred by her primary dentist to this practitioner (RD) for a second opinion.

After reviewing her history, a clinical examination was performed. The patient exhibited uniform occlusal contacts, and minimal wear facets were present. The mandibular range of motion recorded a maximum opening of 31 mm, right lateral excursive movement of 6 mm, and a left lateral excursive movement of 7 mm. No joint sounds could be heard upon auscultation. During palpation, the left anterior belly of the temporalis muscle was excessively tender, the left coronoid process was mildly tender, and the remaining muscles of mastication were not remarkedly tender. The patient exhibited an area of brawny swelling in the left temporal region, while the remainder of the head and neck examination was within normal limits. Radiographic examination of the temporomandibular joints (TMJs) exhibited reduced joint space and very early bilateral osteoarthritis. The erythrocyte sedimentation rate was 66.

The differential diagnosis included osteoarthritis, temporal arteritis, or space-occupying lesion. A magnetic resonance imaging (MRI) scan demonstrated a homogenous lesion of 3.4 × 3.0 × 1.7 cm in the sphenoid process and the lateral wall of the orbit (Figs 1a to 1c). According to image studying, the lesion resembled meningioma. The patient was referred for further evaluation. The pathologic diagnosis revealed the tumor to be a primary lung carcinoma, metastatic to the left temporal fossa.

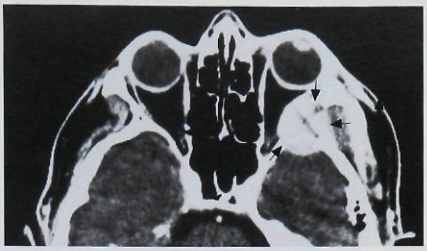


Fig 1a Contrast-enhanced CT image of rounded lesion of a vascular nature (arrows).

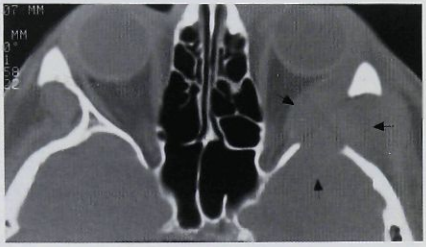


Fig 1b Computerized tomography image with bone window settings.

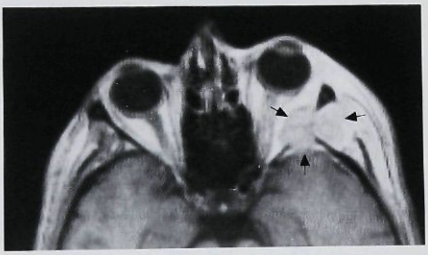
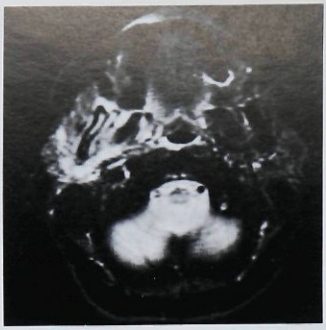
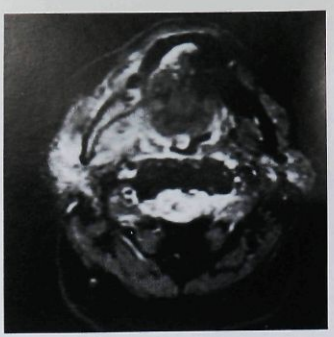


Fig 1c Image showing destruction to the sphenoid wing and lateral orbit caused by the lesion.

Clues

Characteristics leading to in-depth evaluation were the elevated sedimentation rate (66), the brawny swelling in the region of the anterior temporalis muscle, the single muscle being tender to palpation, and the reduced range of motion.





Figs 2a and 2b T2-weighted MRI scan showing abnormal increased signal intensity to the masseter and pterygoid muscles on the right side. The bright, abnormal signal intensity seen at the angle and ramus of the right mandible is consistent with neoplastic invasion from squamous cell carcinoma.

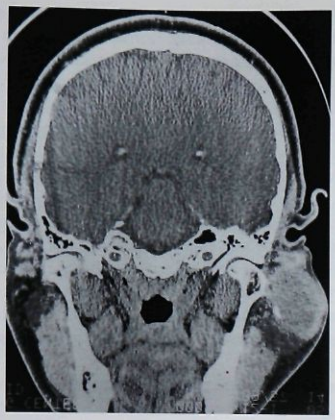
Case 2

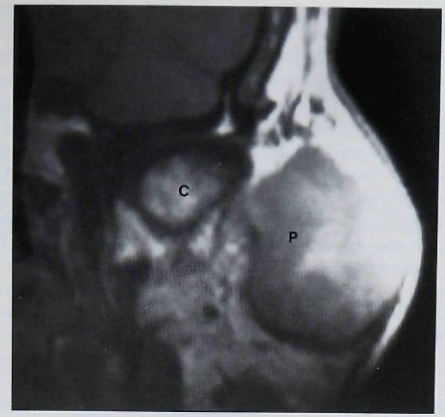
A 55-year-old male smoker was referred for evaluation for possible CMD. His chief complaint was an inability to open his jaw, with a progressive decreasing range of motion. Two years previously, the patient had undergone a parotidectomy, revealing a squamous carcinoma. Panendoscopy with selected biopsies did not reveal a primary site; the patient underwent radiation therapy of 60 Gy to the primary area and 55 Gy to the neck region. Serial evaluations were unremarkable until 1 year later, when an asymptomatic squamous cell carcinoma was identified in the right nasal vestibule. Repeated panendoscopy and selected biopsy were negative, and radiation therapy was used to 66.5 Gy to the primary site. The patient was again followed every 4 to 6 weeks and did reasonably well for 8 to 10 months, when he began complaining of trismus with jaw tenderness on the right side. A computerized tomography (CT) scan showed no lesion in the paranasal sinus, pterygoid plate, or nasal cavity. A diagnosis of CMD was made from the decrease in range of motion, bruxism, and left temporomandibular click. The suspected cause was fibrosis as a result of earlier radiation therapy. This formed the basis for referral to a head/neck pain management practice.

Upon evaluation, the patient revealed a maximum mouth opening of 7 mm, right lateral excursive movement of 5 mm, and left lateral excursive movement of 6 mm. A sporadic soft click could be heard in the left TMJ, but no tenderness was present. An initial diagnosis of decreasing mobility secondary to radiation effect was made based on possible muscle fibrosis and muscle contracture as a consequence of earlier radiation therapy. The patient was advised of routine home exercises to attempt to increase his range of motion. Iontophoresis was performed on the right masseter muscle and led to mild subjective relief; spray and stretch was performed on the temporalis and sternocleidomastoid muscles, although the range of motion was not increased. The patient was seen 2 days later, and he reported that his jaw was sore and his "lip and chin felt puffy." His range of motion, at that time, had decreased to a maximum mouth opening of 5 mm, a right lateral excursive movement of 6 mm, and a left lateral excursive movement of 2 mm. Chin paresthesia with decreased sharp sensation was noted over the right side of the chin and lower lip. Based upon previous history and new neurologic findings, an MRI was performed. It showed a diffuse and ill-defined process of the right parotid gland and infratemporal fossa (Figs 2a and 2b). A repeat CT scan showed bilateral cervical adenopathy that was not clinically evident. A fine-needle aspiration confirmed recurrent squamous cell carcinoma. Despite a skull base procedure and I-125 seeds, the patient died 2 months later of complications resulting from his carcinoma.

Clues

The lack of muscle or joint tenderness, the rapidly decreasing range of motion, the emergence of neu-





Figs 3a and 3b Coronal CT and MRI scans of patient no. 3. Note close proximity of markedly enlarged parotid gland (P) and mandibular condyle (C).

rologic symptoms, and the patient's past medical history are the "red flags" of an atypical CMD patient.

Case 3

A 33-year-old man presented with a fullness in the area of the left TMJ. The patient felt the mass had been present for 1.5 months, and it was first noticed following a blow to the left jaw. The patient smoked a pack and a half of cigarettes a day. The examination revealed a 3 × 2 cm firm mass, located slightly inferior to the lateral pole of the left mandibular condyle. The lesion was slightly mobile and erythematous, but not tender to palpation. The range of motion was recorded as a maximum mouth opening of 42 mm, a right lateral excursion of 5 mm, and a left lateral excursion of 12 mm. The radiographic examination from the submental vertex view was noncontributory. The right and left transcranial radiographic projections revealed glenoid fossae that were of normal morphology, the right and left condyles showed signs of minor sclerosis, and the joint spaces were within normal limits. The firm, enlarged area was not thought to be a traumatically induced condylar fracture, nor a laterally displaced meniscus, because the mass remained stable upon condylar movement. Computer tomography and MRI showed normal joint morphology, but dilation of the parotid gland area with lymphomatous infiltration was apparent (Figs 3a and 3b). The area of parotid involvement was not tender to palpation. The patient was referred for further evaluation. A fine-needle aspiration of the mass yielded a diagnosis of malignant melanoma. Close examination, including dermatologic skin survey, upper airway panendoscopy, and ophthalmologic retinal examination, did not reveal a primary tumor. The patient underwent a parotidectomy, radical neck dissection, and excision of skin above the tumor mass, followed by radiation therapy. He is currently under close observation, with no evidence of persistent or recurrent disease.

Clues

The crucial signs in this case were the lack of tenderness in the parotid gland, the lack of mobility of the mass even though the right lateral excursive movement was markedly decreased, and the fact that the condyle underwent a separate degree of translation.

Discussion

Discomfort arising in the craniomandibular structures and muscles of the head and neck presents a challenging differential diagnosis. Use of the term temporomandibular joint pain or "TMJ" is discouraged, because even this vague description does

not completely encompass the possible disorders presenting in this area.[9] Terms such as myofascial pain dysfunction syndrome (MPD), head and neck fibromyalgia, and craniomandibular dysfunction have been suggested. Practitioners must use clinical judgment based on history, experience, and examination findings to determine whether an exhaustive series of tests is warranted, or if they should proceed with relatively simple treatment strategies, such as stress counseling, anti-inflammatories, and possibly a trial of orthotic splint therapy. While orthotic splint therapy is highly successful and cost effective in the treatment of TMJ disease, it should not delay the investigation of a potentially more serious lesion.[10,11] Among serious etiologies for CMD, none is more ominous than malignant lesions. No simple screening test exists to exclude neoplastic disease. Metastatic disease presenting as myofascial pain has been reported by various authors on a regular basis,[1-7] but is uncommon. The presence of bruxism, opening or closing clicks, crepitus, decreased range of motion, and joint tenderness can be very misleading. The patient with pain in this area having minimal or no findings related to the joint and lacking evidence of routine disease of the paranasal sinuses or parotid gland should be carefully scrutinized. Careful evaluation should be undertaken to exclude unusual disease before labeling such an atypical patient as having "TMJ" or fibromyalgia.[10] The cases presented here illustrate three important mechanisms by which malignant lesions can result in CMD symptoms.

The first case illustrates tumor spread from elsewhere in the body. Development in such an unusual area is most frequently encountered as a part of generalized carcinomatosis, a diffuse spread of metastatic tumor throughout the body generally occurring as a late stage, presumably after the patient's immunologic defenses have been exhausted. It follows that patients presenting with this condition will usually have many other signs in their history or physical examination, especially neurologic signs, to assist the clinician in identifying the cause of pain and dysfunction. Occult metastatic disease (tumors presenting as pain or swelling where cancer is not yet known to exist) spreads to the soft tissue surrounding the oral cavity most commonly from the lung, followed by the kidney.[12] Numerous other tissue sites have metastasized to this area on an infrequent basis.

The second case illustrates the spread of carcinoma from another location in the head and neck. While the patient's initial diagnosis and treatment was necessarily that of unknown primary carcinoma to the parotid gland, the later identification of a nasal vestibule squamous carcinoma makes it quite likely that this carcinoma did arise in the upper sinonasal tract. Head and neck tumors are known to cause symptoms of jaw dysfunction.[1,4,11-13] These tumors are frequently squamous cell carcinoma of the pharynx or posterior third of the tongue. Tumors growing along the path of the fifth and seventh cranial nerves may also mimic this syndrome, as can intracranial tumors, such as pituitary adenoma and tumors of the cerebellopontine angle.

The third case serves as a reminder not only that the skin overlying this area of the body may cause primary pathology, but also of the tendency of malignant melanoma to occur in an unusual location with swift growth and devastating malignant potential.

The actual incidence of malignancy occurring in the TMJ area cannot be readily calculated, since the occurrence of pain in this location is quite common and patients present to various practitioners. It is known that adenocarcinoma is the most frequent tissue type metastatic to the jaw, accounting for 70% of metastatic disease. The most common primary sites are the breast (30.4%), the kidneys (15.6%), and lungs (14.8%).[14] Even in a population with metastasis to the jaw, condylar involvement is rare (< 8%). Rubin and coworkers[15] reviewed theories accounting for patterns of mandibular metastasis. Poor blood supply to the condyle and the presence of red marrow in the area of the molars may account for the frequency of metastatic disease in this latter area.

The literature disclosed a total of 23 patients having malignancy involving the TMJ where sufficient information to categorize by history and examination findings was available.[1-7] Of these patients, 12 (51.6%) suffered symptoms consistent with TMJ disease, 12 (51.6%) experienced trismus, 13 (56%) had numbness, and 7 (30%) reported hearing loss. While these patients were evaluated by different investigators, separated in some cases by years, this serves to illustrate the frequency with which "concerning" symptoms occur in the presence of tumor. The presence of pain as a symptom is difficult to assess from a retrospective review. Most series noted patient reports of pain associated with the TMJ; however, in our experience, this discomfort was noticeably less than what is typical for a patient with TMJ disease.

Future descriptions may benefit from inclusion of the presence or absence of a trigger point or "jump sign" as described by Fricton and associates.[16] No trigger point of this severity was present in the cases presented here.

Conclusion

The history and physical examination are paramount in assembling a differential diagnosis. The routine use of extensive laboratory and imaging studies for CMD is neither cost effective nor intelligent health care. The cases presented here provide a glimpse into subtle, yet identifiable, differences in the presentation of neoplastic disease in comparison to the more common mechanical and inflammatory factors contributing to CMD/head and neck pain.

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Resumen

Lesiones Malignas que se Presentan como Síntomas de Disfunción Craneomandibular

Se reportan tres casos de carcinoma, observados en una consulta de manejo de dolor de cabeza y cuello, durante los dos últimos años y que se presentaron como síntomas de una disfunción craneomandibular. Los clínicos siempre deberían considerar el diagnóstico diferencial debido a la variedad de causas que pueden causar los síntomas presentados por el paciente. Estos síntomas actúan como pistas que ayudan a diferenciar entre un proceso rutinario o uno "maligno". La necesidad de realizar y de repetir los examenes de imágenes, cuando los síntomas clínicos no se correlacionan con el examen clínico, por medio de guías establecidas para la disfunción craneomandibular o para el dolor de cabeza y cuello, es de suma importancia.

Zusammenfassung

Maligne Neoplasien mit Symptomen einer Myoarthropathie des Kausystems

Drei Fälle von malignen Neoplasien werden präsentiert, bei denen einzelne Symptome der Myoarthropathie des Kausystems (MAP) vorliegen. Ausgedehnte Laboruntersuchungen und extensive Verwendung bildgebender Verfahren sollten jedoch nicht zum Standard für alle MAP-Patienten erhoben werden, weil diskrete, aber doch feststellbare Unterschiede zwischen Neoplasien und MAP — entzündlich oder degenerativ — auch in der klinischen Untersuchung auffallen.