

A Systemic Review of Patients Appearing to Dental Professionals with Trigeminal Neuralgia Arising From Intracranial Tumours

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Abstract: A several number of patients seeking dentists with trigeminal neuralgia will have intracranial tumors as the primary cause. Before the right diagnosis is made, these patients may undergo unnecessary dental involvements. This is because there are difficulties of diagnosis as the symptoms sometimes lead to misdiagnosis. A search of the literature was done to find case reports of this incidence. Cases is to be analysed for common characteristics or presenting features that may assist dentists in recognizing patients with intracranial tumors.

Keywords: Intracranial tumour, epidermoid tumour, trigeminal neuralgia, orofacial pain

1. Introduction

Orofacial pain is an ever-present complaint, makes a patient to seek a dentist for management of the pain. Such disorders may have pain and related symptoms arising from a discrete cause, such pain associated with a malignancy or as postoperative pain, or may be syndromes in which pain as the primary problem, such as temporomandibular joint disorder pain, neuropathic pains or headaches. However, only in few rare cases the pain might arise from neurogenic sources. Intracranial tumors are one of the rare sources of orofacial pain. Out of 2000 patients with orofacial pain only about 20 patients has an intracranial tumor as an underlying cause.⁽¹⁾ Patients and dentists can easily get the wrong idea about this type of pain as pain arising from teeth or oral structures. Misdiagnosis might lead to unnecessary procedures and can delay definitive treatment. About 33% to 65% of patients with trigeminal neuralgia (TN) appearing to their dentist initially undergoes unnecessary dental treatments.⁽²⁾ I present cases of patients with orofacial pain arising from intracranial tumors that initially appeared to a dentist. We looked for common characteristics that may help distinguish orofacial pain arising from intracranial tumors. Although such an analysis will be subject to publication prejudice, given the infrequency of this occurrence the analysis is the best available data in place of retrospective cross-sectional studies.

2. Material and Methods

The electronic database was searched for all English-language case reports of orofacial pain that eventually arose from intracranial tumors. A wide-ranging search strategy was employed using any combination of keywords as such oral pain, orofacial pain, dental pain, intracranial tumor, brain tumor, intracranial malignancy, acoustic neuroma, cerebellopontine angle lesion, and meningioma. Search results were selected by title and abstract to choice for case reports of orofacial pain arising from intracranial tumors. Many papers were drawn from references made in other relevant case reports found using our search strategy. Papers that met the following criteria were included in our data: (1)

cases involving any complaint that would be of interest to the dental field, including but not limited to orofacial pain, temporomandibular disorders (TMD), and trigeminal neuralgia (TN); (2) initial visit to a dental specialist; (3) a well-described clinical course, including symptoms at visit and diagnostic work-up; and (4) involvement of a different intracranial tumor.

3. Results

The search queries produced 875 results with considerable overlap between the many combinations of search prompts.⁽³⁻²⁷⁾ Thirty-two case reports were found that met these criteria. Table I summarises these cases. Ages ranged from 14 to 74 years, with the mean being 43.3 years (SD=14.33). Sex distribution was slightly skewed toward females, with 62.5% female (20 of 32) and 37.5% male (12 of 32). Trigeminal neuralgia (TN), temporomandibular disorder (TMD), and persistent idiopathic facial pain (PIFP) were the three most common diagnoses made when patients were first seen by dental specialists. 37.5% were diagnosed with TN (12 of 32), 31.25% with symptoms associated with TMD (10 of 32) and 21.88% with PIFP (7 of 32). 2 cases did not include a clear report of the initial diagnosis and one patient had burning tongue syndrome. Of all the patients, about 62.5% (20 of 32) had additional symptoms that were not typical for the presumed initial diagnosis. These symptoms consisted of intraoral and facial sensory loss, cranial nerve palsies, hemiplegia, facial muscle weakness and more. From time of appearance to the dentist to diagnosis of an intracranial tumor, the average median delay in diagnosis was 1 year; however, there was a wide variation (0-40 months). A clear report of the delay of diagnosis was not included in majority of cases. In 18 cases medical therapy and pain control were attempted by dentists, of which most of it were either refractory or soon became refractory to pharmacotherapy. A various choice of medications was reported, including Dilantin, prednisone, nonsteroidal anti-inflammatory drugs, carbamazepine, dothiepin, gabapentin, and narcotics. Of all the cases, about 56.25% (18 of 32) underwent dental procedures prior to establishing the correct diagnosis. Procedures included tooth extractions, temporomandibular joint orthotic placement,

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selective nerve blocks, and endodontic procedures. Neurosurgical resection was performed in 25 cases; of these, 43.75% (14 of 32) reported complete resolution of pain, 6.25% (2 of 32) had partial resolution, 1 experienced no change, and 4 did not report response. Of the 4 deaths reported, 3 were the result of natural progression of glioblastoma multiforme and 1 was from pulmonary embolism in the instant postoperative course. Follow-up times ranged from 14 days to 36 months, with a mean average of 15.32 months (SD=9.02).

4. Discussion

Our analysis suggests that symptoms of trigeminal neuralgia, temporomandibular disorder pain and persistent idiopathic facial pain, are the three most common presentations of patients with intracranial tumors. This partly confirms the study conducted by Bullitt et al.⁽¹⁾, who found that of 13 patients with tumor-induced facial pain, 5 had atypical facial pain and 8 had trigeminal neuralgia; however, they did not report any cases of temporomandibular disorder pain.

Trigeminal neuralgia

Trigeminal neuralgia is characterized by episodic paroxysms of unilateral facial pain, described as electric shock-like or lancinating, most commonly in the second and third trigeminal divisions. Often, a “trigger zone” can be recognised; this is the point which low-intensity mechanical stimulation such as washing, speaking, light touch, or even air currents can aggravate the pain. Each attack typically lasts between ten seconds and two minutes, with pain-free periods lasting seconds to hours in between. Pathophysiologically, it is believed that trigeminal neuralgia arises from focal demyelination at the nerve root entry zone, which allows electrical spread of excitation between adjacent sensory axons.⁽³¹⁾ When any single neuron in a functionally linked neurons group discharges, the entire group can become activated and numerous nociceptors are activated by cross-excitation. The electric shock-like paroxysm of pain is caused by this rapid spread. The International Headache Society has established formal criteria for the diagnosis of trigeminal neuralgia within the International Classification of Headache Disorders II as follows^(5, 32) (1) Paroxysmal attacks of pain lasting from a fraction of a second to two minutes, involving one or more branches of the trigeminal nerve and accomplishing criteria B and C; (2) Pain has at least one of the following characteristics: (a) Sharp, intense, stabbing or superficial. (b) Precipitated by trigger factors or from trigger areas; (3) Occurrences are stereotyped in the individual patient. Trigeminal neuralgia can be further divided into “classical trigeminal neuralgia,” that is believed to be following of either micro neurovascular compression or otherwise idiopathic and includes up to 90% of cases, and “symptomatic trigeminal neuralgia,” is the type that arises from a demonstrable lesion such as a tumor or multiple sclerosis.⁽³¹⁾ Studies have shown that 5% to 16% of patients who have trigeminal neuralgia may have intracranial tumors.^(3, 33, 34) Various mechanisms have been hypothesized as to how intracranial tumors cause trigeminal neuralgia: (1) direct tumoral compression on the trigeminal nerve, (2) arterial displacement with resulting neurovascular

compression, and (3) chemical irritation of the nerve.⁽³⁵⁾ To aid practitioners in identifying this subclass of patients, numerous authors have tried to identify “red flag features” that may raise suspicion for intracranial lesions. In our review, we noted that 71.43% of patients with trigeminal neuralgia (10 of 14) also presented with orofacial neurologic deficits. This finding confirms larger studies such as that by Nomura et al. who presented that 50% of patients with trigeminal neuralgia secondary to intracranial tumors had associated sensory losses.⁽³⁾ Revilla reported in a review of 473 patients with trigeminal neuralgia that 24 had intracranial tumors; of these, 11 had atypical neurologic deficits.⁽³⁶⁾ Puca et al. reported in their series of 73 patients with tumor-associated trigeminal that symptoms can correlate with the extent of tumor involvement: tumors in contact with the trigeminal nerve were commonly associated with paresthesias, tumors causing compression caused facial pain, and infiltrating tumors caused dysesthesia.⁽³⁷⁾ Patients with symptomatic trigeminal neuralgia resulting from a cerebellopontine angle mass will often additionally show subtle facial weakness and hearing loss on the same side.⁽³¹⁾ Matsuka et al. list the following symptoms in patients with the most common trigeminal neuralgia causing tumor, acoustic neuromas: hearing deficits (60%-97%), tinnitus (50%-66%), vestibular disturbance (46%-59%), numbness or tingling in the face (33%), headache (19%-29%), dizziness (23%), facial paresis (17%), and trigeminal nerve disturbances (hyperesthesia, paresthesia, and neuralgia; 12%-45%).⁽²⁴⁾ McCormick et al.'s literature review of 120 trigeminal schwannomas revealed that neurological deficits other than trigeminal nerve abnormalities were present in over 75% of patients at the time of diagnosis.⁽³⁸⁾ They too report a wide spectrum of neurologic findings. Christiaans et al. report in their prospective study of 68 patients with already known cancer that the discriminative ability of history and physical examination was disappointingly low for differentiating headache from metastasis to the brain versus other etiologies.⁽³⁹⁾ However, they did identify three factors that were useful in identifying intracranial tumors: (a) emesis, (b) non-tension-type headache, and (c) headache duration of 10 weeks. In my review, 75% of patients (6 of 8) who presented with trigeminal neuralgia and were treated with pharmacotherapy were nonresponsive. In contrast, previous studies have demonstrated that symptomatic trigeminal neuralgia arising from intracranial tumors will readily respond to medical therapy, such as with carbamazepine.^(33, 40, 41) However, many of these cases soon relapse and become refractory. It is possible that continued tumor growth and nerve compression can worsen the underlying demyelination process of the nerve root.^(40, 41) Although our series does not corroborate this theory, we suspect that the previously reported findings are more likely to be valid. Our patient population is small in comparison and subject to publication bias. Therefore, we agree with the assertion that tumor-induced trigeminal neuralgia can respond readily to early pharmacotherapy. This observation helps dentists in the sense that it should prompt consideration for head imaging in trigeminal neuralgia cases that become refractory to medical therapy. However, it also confounds diagnosis because it implies that intracranial tumors cannot be ruled out in patients who respond favourably to medication.

Temporomandibular disorder

Temporomandibular disorder represent the most common overall cause of chronic orofacial pain, with a prevalence of 12% in the general population, 5% severe enough to warrant treatment.⁽⁴²⁾ The classification of temporomandibular disorder covers a broad variety of diagnoses; accordingly, symptoms of temporomandibular disorder can vary greatly. These symptoms can include facial pain that is aggravated by jaw function, tenderness upon joint or muscle palpation, limited mandibular range of motion, deviation or deflection of the mandible on mouth opening, and temporomandibular joint sounds.⁽⁴²⁾ Nine of the reviewed cases included patients diagnosed with temporomandibular disorder-like symptoms. Of these, only three presented with symptoms not characteristic of temporomandibular disorder. Three of the misdiagnosed temporomandibular disorder patients went on to develop drastic neurologic symptoms. It appears that this group of patients presented practitioners with the most difficult diagnostic challenge because there were no early signs of an alternative etiology. Matsuka et al. share this concern in their discussion when they suggest that many cases of orofacial pain that are not clearly understood are referred for temporomandibular joint evaluation, frequently delaying proper diagnosis and treatment.⁽²³⁾ Of note, Levitt et al. used a validated diagnostic tool, the temporomandibular joint scale, and identified brain tumors early on in patients with temporomandibular disorder in the absence of uncharacteristic symptoms.⁽²³⁾

Persistent idiopathic facial pain

Persistent idiopathic facial pain is previously known as "atypical facial pain". Persistent idiopathic facial pain is a difficult entity to recognize. It has been described as a continuous pain that is deep, diffuse, dull, and aching. It can be confined to a limited facial area, most commonly the maxillary region, but has also been reported to be bilateral. The pain is not associated with sensory loss and psychiatric comorbidities are common.⁽²⁾ An entity known as atypical odontalgia, characterized as severe throbbing tooth pain without major pathology, is often considered a subentity of persistent idiopathic facial pain.⁽²⁾ Symptoms in a small subset of persistent idiopathic facial pain patients may later evolve into those resembling trigeminal neuralgia; these cases are referred to as pre-trigeminal neuralgia, which is defined as dull, aching pain that precedes development of typical trigeminal neuralgia. Given the nebulous manifestations of persistent idiopathic facial pain, it is difficult to identify a specific pattern that is most readily associated with intracranial tumors. However, areas of facial anesthesia should be regarded with suspicion because sensory loss is not a described symptom of persistent idiopathic facial pain; 4 of 6 of the cases we found presented with such symptoms.

Imaging

It has been shown that magnetic resonance imaging of the brain is the most sensitive imaging modality for detecting intracranial tumors.^(43, 44) We posit that this test should be performed whenever a patient presents with warning signs suggestive of intracranial tumors. With respect to persistent idiopathic facial pain and temporomandibular disorder, imaging should be considered any time these entities present in conjunction with symptoms that extend beyond the

typical description of these syndromes. Areas of subjective or objective facial sensory loss or paresthesias should be of particular concern. In the case of trigeminal neuralgia, the current standard of care dictates that intracranial imaging should be performed for all newly diagnosed patients.⁽⁴⁵⁾ This practice should help rule out intracranial causes in this population. Based on our review, we additionally recommend that repeat imaging should be considered in patients who initially respond to and then subsequently fail medical therapy. It should also be noted that response to pharmacotherapy should not exclude the possibility of an intracranial tumor. Patients should be adequately followed for at least 1 year to monitor for incipient localizing signs of late intracranial tumors.⁽²⁷⁾ Finally, our data agree with previous authors that neurologic findings correlate to an extent with location of lesion within the cranium. For instance, Bullitt et al. found that peripherally placed tumors tend to cause persistent idiopathic facial pain associated with sensory loss; middle fossa tumors usually cause severe persistent idiopathic facial pain with a progressive neurological deficits; and posterior fossa tumors are more likely to cause trigeminal neuralgia accompanied by subtle neurological deficits.⁽¹⁾ Our data agree with these assertions: 78.57% of patients (11 of 14) with trigeminal neuralgia had posterior fossa tumors, whereas all but 1 persistent idiopathic facial pain case were caused by peripheral and middle fossa mass lesions.

5. Conclusion

Trigeminal neuralgia, persistent idiopathic facial pain, and temporomandibular disorder are three common misdiagnoses given to patients with orofacial pain arising from intracranial tumors. A detailed history and physical exam with particular attention to neurologic function, coupled with a comprehensive understanding of orofacial pain syndromes, are requisites for detecting abnormal signs and symptoms. Particular attention should be given to cranial nerve function, areas of unexplained sensory loss, concurrent atypical headaches, and distant neurologic symptoms. Well-designed cross-sectional or retrospective studies could further aid in identifying characteristics of patients with orofacial pain arising from intracranial tumors. The results of such studies would help dental practitioners better identify at-risk patients for occult intracranial lesions.

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