ORIGINAL ARTICLE

Unnecessary dental procedures as a consequence of trigeminal neuralgia

Kajetan L. von Eckardstein · Moritz Keil · Veit Rohde

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Abstract Trigeminal neuralgia (TN) is a disorder characterized by repetitive lancinating pain along one or more branches of the trigeminal nerve and is commonly triggered by chewing and manipulation of the gums. The second and third divisions are most commonly affected. Due to these symptoms, patients are likely to consult their local dentist when symptoms first develop and may receive further dental evaluation and treatment before they are referred to a neurologist or neurosurgeon. We sought to answer questions regarding evaluation and possible dental treatment as well as referral patterns in TN patients. Using a surgical database, we obtained data of patients undergoing an intervention for trigeminal neuralgia. Telephone interviews were conducted, focusing on initial evaluation and possible dental treatment, on referral patterns, and on present status. Secondly, a written questionnaire was mailed to local dentists. Eighty-two percutaneous rhizotomies and 33 microvascular decompressions were performed in 99 trigeminal neuralgia patients. Of 92 patients contacted, 51 were alive and willing to participate. Two thirds reported being pain-free. Forty-one patients (82 %) initially consulted their dentist; of these, 27 patients received invasive dental treatment for the pain syndrome, including extractions, root canal treatments, and implants. Of 98 local dentists contacted, 51 responded, with three quarters feeling competent in evaluating trigeminal neuralgia. A high percentage of patients that are surgically treated for trigeminal neuralgia consult

Kajetan von Eckardstein and Moritz Keil have equally contributed to this study.

K. L. von Eckardstein (☒) · M. Keil · V. Rohde Department of Neurosurgery, Göttingen University Medical Center, Robert-Koch-Straße 40, 37070 Göttingen, Germany e-mail: kajetan.voneckardstein@med.uni-goettingen.de their dentist first and receive possibly unjustified dental treatment. Differential diagnoses include odontogenic pain syndromes as well as atypical orofacial pain. The present literature acknowledges difficulties in correctly diagnosing trigeminal neuralgia, but seems to underestimate the extent.

Keywords Trigeminal neuralgia · Odontogenic pain · Atypical orofacial pain · Differential diagnosis

Introduction

Idiopathic trigeminal neuralgia (TN) is a common facial pain syndrome affecting four patients in 100,000. Typically, patients suffer from unilateral, sharp, lancinating pain attacks, lasting for several seconds to a few minutes. Quite frequently, patients are able to identify pain triggers, such as laughing, chewing, shaving, or brushing their teeth. The pain usually involves the maxillary nerve, often in combination with the mandibular nerve. The supraorbital nerve is less frequently affected and almost never exclusively [7]. This symptomatology of perioral facial pain affected by chewing or manipulation of teeth and gums is likely to make patients think of a dental cause for their pain when TN first manifests; hence, these patients consult their dentist first.

Difficulties distinguishing between TN pain and odontogenic pain were already extensively described in 1896 by Fedor Krause (1857–1937), one of the founding fathers of German neurosurgery. In his monograph, *Die Neuralgie des Trigeminus* [8], Fedor Krause wrote: "It is very common that patients experiencing neuralgia of the second or third branch of the trigeminal nerve, in the beginning of their suffering, will have extracted all teeth of the affected region." He concluded: "Generally, on thorough examination, it will be



easily recognized, whether the pain originates from the teeth or not'.'1.

The diagnosis of TN is still a clinical diagnosis. Idiopathic TN is a differential diagnosis of atypical odontalgia or neuropathic trigeminal pain (i.e., deafferentation pain) and of primary odontogenic pain syndromes, such as pulpitis and cracked tooth syndrome. Other differential diagnoses include sinusitis and other forms of sinus pain, migraine, and other primary headaches and musculo-fascial and joint pain (Table 1) [1]. Depending on the predominant pain distribution, herpes zoster, orbital disease, temporal arteritis, and intracranial tumors also need to be taken into account. When first confronted with a patient with a clinical suspicion of TN, magnetic resonance image (MRI) of the head with and without contrast enhancement should be performed to rule out symptomatic disease. Treatment modalities for idiopathic TN include medical [20], surgical, and radiosurgical [23] options. In percutaneous ablative procedures, the Gasseric ganglion is temporally or permanently damaged either chemically [21] or thermically or by pressure [10]. In 1985, Peter Jannetta introduced the microvascular decompression procedure in which a compressing vessel is separated from the trigeminal nerve root [6].

Little is known about the rate of patients that initially consult their local dentist when symptoms of TN first develop. In the international literature, only one series from Zurich, Switzerland, from 1983 can be found, addressing this question among others [2]. The authors concluded that 73 % of patients reporting to neurosurgery for TN had a prior dental evaluation and 48 % had at least one tooth removed. One might assume that 30 years later, the knowledge among dentists and dental surgeons has increased, resulting in a lower percentage of dental operations prior to TN diagnosis. We therefore elected to perform systematic patient interviews to further elucidate today's role of dentists and dental surgeons in the evaluation of TN.

Methods

Using the electronic surgical database, we performed a query for patients treated surgically for idiopathic TN in our department between January 2003 and December 2008. Patients were contacted by mail and asked to give consent for a standardized telephone interview. The interviews were performed in February of 2010 and included questions on first evaluation, performed tests and treatments, and the time

Tes gehört zu den regelmässigen Vorkommnissen, dass Leute, welche von Neuralgie des zweiten oder dritten Trigeminusastes ergriffen sind, sich im Beginne des Leidens nach einander alle Zähne des betreffenden Kieferabschnittes ausziehen lassen. [...] Im allgemeinen wird sich bei genauer Untersuchung unschwer erkennen lassen, ob der Schmerz von den Zähnen herrührt oder nicht." F. Krause, p. 143 [8].



Table 1 Differential diagnosis of idiopathic TN

Affection of the trigeminal nerve along its course by

Tumors

Multiple sclerosis plaques

Herpes zoster

Deafferentation trigeminal pain syndromes

Atypical orofacial pain, neuropathic trigeminal pain

Dental disease

Musculo-fascial and joint disease

Temporomandibular joint disease

Vascular disease

Temporal arteritis

Migraine headaches

Others

Other headache types

Referred pain from orbits, sinuses

Psychogenic causes

intervals between first symptoms, establishment of the diagnosis, and treatment for TN (Table 2). Patients were not asked to identify their dentists. Using the local telephone registry, we furthermore mailed out a written questionnaire to all local dentists, asking them for a self-assessment in the diagnosis and management of TN (Table 3). Dentists were offered to reply anonymously since matching between patients and their dentists was not intended. Descriptive statistics were used for interpretation of data. The study was approved by the Institutional Review Board (Protocol Number 17/5/09).

Results

Patient interviews

We identified 118 TN procedures in 99 patients. Eighty-two percutaneous rhizotomies and 33 microvascular decompressions were performed; three procedures were other surgical lesioning procedures. Ninety-two patients were contacted by mail, asking for permission to call and conduct an interview; in seven patients, no valid address was available. Fifty-seven patients or their families replied; 51 patients were alive and willing to participate (study population, 55.4 % return rate). The median age of this group was 69.3 years, and 26 patients were female. Overall, 66 % of patients reported being currently free of any TN pain attacks; there was no outcome difference in the rate of patients undergoing dental treatment and those who did not. The right side was predominantly affected (29 cases); the first branch was involved in eight cases. No patient recalled an association of the onset of pain with a dental procedure beforehand.

Table 2 Standardized questions for patient telephone interview

- Did you see your dentist on a regular basis prior to evolution of symptoms of TN?
- 2. When experiencing the first symptoms of TN, did you initially see your dentist?
- 3. If so, did your dentist establish or have the suspicion of a diagnosis of TN?
- 4. If you initially saw your dentist and if the dentist did not suspect a diagnosis of TN, were invasive dental or orosurgical procedures offered and performed?
- 5. If so, how was the further therapeutic and diagnostic workup? Did the dentist write for a consultation to another health care provider? If so, to which kind of specialist?
- 6. If your initial visit was at a dentist's office, do you recall what time elapsed between (a) first symptoms, (b) your visit, and (c) the neurosurgical evaluation and therapy?
- 7. Are you currently free of any TN pain?

Of these 51 patients, 41 positively stated that the first health-care provider visit regarding initial symptoms of TN was with their dentist (82 %). Of these 41 patients, only two remembered that the dentist established the differential diagnosis of TN (4.7 %); the remaining 39 patients (76.5 %) felt that the dentist did not recognize the correct diagnosis. Twenty-seven patients (66 %) received an invasive dental treatment for their pain syndrome, including the extraction of a total of 54 teeth in 13 patients (median 2 teeth, maximum 20 teeth), 13 root canal treatments in 5 patients (median 2 teeth, maximum 6 teeth), 7 preparations for tooth replacement procedures, 4 implants, 4 injections, and 3 fillings (Figs. 1 and 2). Only one of the eight patients with involvement of the first branch underwent a dental procedure compared to 26 of 43 patients without involvement of the first branch (p=0.02, Fisher's exact test). Concerning symptom relief after the neurosurgical procedure for TN, there was no significant outcome difference in the rate of patients undergoing dental treatment (63.0 % with symptom relieve) to those who did not (70.8 %, p=0.77, Fisher's exact test).

Twenty-one patients reported that the dentist referred them to another health-care provider at some point in time, whether be it before or after treatment. Thirteen patients were referred

 Table 3
 Written questionnaire to local dentists

- 1. Within the last 6 years, how often did you approximately suspect TN in the evaluation of patients with orofacial pain?
- 2. Approximately how many patients with known TN did you treat for coexisting dental morbidity within the last 6 years?
- 3. Do you feel competent establishing the clinical diagnosis of TN?
- 4. Do you know that carbamazepine is a potent medication for TN?
- 5. Are you aware of any surgical treatment modalities of TN?
- 6. Do you believe that there is a need for further information and education in regard to TN as a differential diagnosis in orofacial pain syndromes?

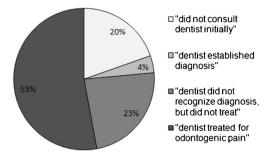


Fig. 1 Percentage of patients undergoing evaluation and possible dental treatment for trigeminal neuralgia

to other dentists, oral surgeons, or maxillofacial surgeons; six patients saw a neurologist or neurosurgeon next; and one patient was sent to a primary care physician and a physiotherapist. Of patients that reported to a dentist, 70.6 % did so within 4 weeks of the onset of symptoms; only 14.7 % remained without evaluation for over 1 year. In addition, 22.5 % of the these patients were finally seen by a neurologist or neurosurgeon within 6 months of the onset of symptoms, but more than 2 years elapsed prior to neurological or neurosurgical attention in 42.5 % of patients.

Dentist questionnaires

Ninety-eight local dentists were identified and received the questionnaire; 51 responded (52 %). When asked to estimate the number of patients with suspected TN as a first differential diagnosis in evaluation of orofacial pain within the last 6 years, 34.7 % said they had never identified a case suspicious of TN, while 8.2 % established this diagnosis at least five times and 4.1 % at least ten times. Thirty-five dentists (74.5 %) felt competent enough to identity TN as a cause of orofacial pain, and 25 dentists (50 %) stated that they knew that carbamaze-pine and surgical interventions were a treatment modality for TN. However, the majority (76.6 %) of dentists felt that there is a need for further information and ongoing medical education regarding TN.

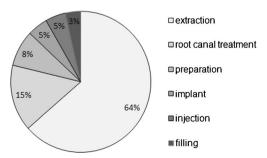


Fig. 2 Number of dental treatments per single tooth in 27 TN patients undergoing orosurgical therapy



Discussion

We have conducted retrospective interviews with patients that were surgically treated at our department for TN on initial evaluation and treatment for their pain syndrome before the initiation of a specialist treatment by neurologists or neurosurgeons. We learned that over 80 % of our patients, initially, saw a dentist for the emerging symptoms and that two thirds of them received various invasive, possibly unjustified dental treatments before they were evaluated by a neurologist or neurosurgeon.

Limitations of the study

Before discussing our findings in the light of the published literature, the authors would like to point out the main limitation of the study. The data were collected in patient interviews; original charts of local dentists were not reviewed. Dentist's assessments on clinical status and examinations, X-rays, and other imaging modalities were not evaluated. Hence, it cannot be excluded that the individual dental management and treatment was fully justified, be it due to concurring dental disease or due to prophylactic reasons. On the other hand, it seems possible that the neurosurgical diagnosis of TN is incorrect and that these patients were treated for a condition other than TN. However, for the sake of this retrospective review and in the light of the fact that two thirds of the patients remained pain-free after a TN-specific neurosurgical intervention, we assume that the rate of missed diagnosis is low. There was no difference in outcome between patients undergoing a dental procedure to those who did not.

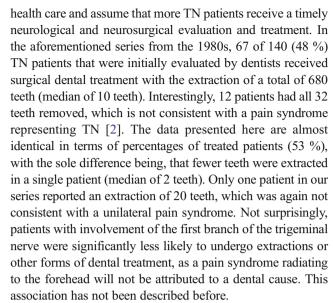
TN patients self-refer to the dentist and will see the specialist too late

We have found in our patients that over 80 % attributed the emerging pain syndrome to a dental condition and saw their dentist first. The Zurich study of Garvan and Siegfried assessed initial evaluation patterns of 140 patients, of which, 73 % received initial dental examination in the 1980s [2]. Surprisingly, this number has not changed throughout three decades, as we would expect growing public knowledge and readily accessible information in Western Europe of this uncommon condition.

Of note, time to referral to a specialist has improved but is still remarkably long. While 16 % of patients were referred to a specialist within 1 year after the onset of symptoms in 1983, 42.5 % of our patients saw a neurologist or a neurosurgeon within 1 year.

TN patients do receive dental treatment

Regarding medical evaluation and proposed treatment options, however, one would value improved dental and medical



Other high volume series regarding the relationship of TN in the differential diagnosis of orofacial pain syndromes and dental treatment are missing in the international literature. German medical dissertations have occasionally covered this subject over the decades, but rarely provided profound data and have not been published in medical journals [12, 13, 19, 4]. In a series from Halle-Wittenberg University from 1938, 39 out of 108 (36 %) patients had teeth extracted in the context of genuine TN [4], and a thesis from Cologne University from 1958 reported 22 extractions in 52 patients (42 %) [12]. The Zurich group mentioned a somewhat larger retrospective series on trigeminal neuralgia and tooth extractions in a Swiss dental magazine which has not been further elucidated [18]. Based upon smaller case series or single case reports, different authors—primarily from a dentist's perspective—underline the importance of considering TN a possible cause of orofacial pain [9, 5].

In a review and discussion of management issues published in 2004, the author retrospectively evaluated 50 consecutive dental patients with the concluding diagnosis of neuropathic trigeminal pain and pointed to the inconsistencies and difficulties in establishing this diagnosis [22]. More than one third of these patients received endodontic therapy. Idiopathic TN as a subgroup of trigeminal nerve-mediated pain is not mentioned as such, but a significant clinical overlap between neuropathic trigeminal pain, odontogenic pain, and temporomandibular disorders is described. The author very precisely demonstrated the wide range of signs and symptoms; some of them would fit well if seen in the proper combination and time course, with the diagnosis of idiopathic TN. More than likely, some of these patients would have received TN-specific neurosurgical therapies if evaluated by a neurologist or neurosurgeon.

In 2009, a similar paper from Los Angeles was published, retrospectively evaluating 64 dental patients with the



concluding diagnosis of atypical odontalgia [14]. In this series, 80 % had dental procedures done, and 26.6 % received endodontic therapy. Again, difficulties in establishing the diagnosis were listed and patient characteristics were evaluated. Interestingly, 64 % of the patients in this series reported no factor that could explain a deafferentation pain syndrome, which is thought to be a prerequisite for the diagnosis of atypical odontalgia. The paper does not make an assumption on how many patients were eventually diagnosed with idiopathic TN in the course of evaluation.

The aforementioned study [14] and a review published in 2008 [1] propose evaluation and treatment algorithms for dentists. Both papers acknowledge the difficulties in obtaining the correct diagnosis, the latter of which explicitly names idiopathic TN as one of the differential diagnoses. The diagnostic-therapeutic paradigm in the Los Angeles paper is more detailed and includes a path to a brain MRI and a consultation to the appropriate specialist if pain persists after dental measures.

Mistaking orofacial pain and atypical trigeminal pain for TN

In the telephone interviews with our patients, no case history was suggestive of an odontogenic, orofacial, or atypical pain syndrome that was inappropriately treated as an idiopathic TN. It should be mentioned, however, that there are reported cases of mistaking orofacial pain for TN. In 1978, Mumford cited individual cases of unerupted teeth and residual cysts leading to the misdiagnosis of TN [11]. Others have published case reports on similar findings [3, 15], also of osteonecrosis of the jaws that may produce a similar pain syndrome as TN [16]. Nevertheless, the majority of these reports date back 15 or more years, making it less likely nowadays that true odontogenic pain is missed with modern imaging modalities readily available [17].

Dentists feel the need for education on TN

Of the 51 local dentists that responded to our questionnaire, three quarters felt competent enough to identify TN in the diagnostic workup of orofacial pain. Nevertheless, the same percentage of local dentists saw the need for ongoing education regarding TN and only half of the dentists knew medical and/or surgical treatment modalities. It seems likely that there is a significant match between the treating dentists of our TN patients and the dentists returning their questionnaire, which, in combination, underlines a significant uncertainness of evaluation, diagnosis, and treatment of TN within the local dentist community. We have not found any similar assessment data or questionnaire projects in the literature.

Conclusion

We have shown that a high percentage of patients that was surgically treated for idiopathic TN in our department initially consulted their local dentist and received dental therapy first. This pattern has long been known, was first systematically assessed in Western Europe three decades ago, and surprisingly has not changed since. It is in the interest of neurologists and especially neurosurgeons to work closely together with local dentists to evaluate orofacial pain syndrome patients for possible idiopathic TN. Dental colleagues ask for support and ongoing education regarding TN, but neurosurgeons should also be familiar with the characteristics of differential diagnoses. Close cooperation and collaboration between neurosurgeons and dentists with a reliable pattern of mutual referrals will only be in the best interest of the patient.

Acknowledgments This study is in part based upon the doctoral thesis (*Dr. med. dent.*) of Moritz Keil.

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Comments

Francesco Acerbi, Milan, Italy

Trigeminal neuralgia (TN) is a syndrome whose patients suffer from episodes of excruciating facial pain in the territory of one or more divisions of the trigeminal nerve that can arise spontaneously or after a gentle tactile stimulation of a trigger point on the face or in the oral cavity or that can be triggered by natural activities, such as chewing, speaking, washing the face, or brushing the teeth.

When the pain involves the maxillary or mandibular division of the trigeminal nerve, primary odontogenic syndrome should be considered as differential diagnosis. However, usually, it should be relatively easy, after a throughout anamnesis and clinical examination, to identify if the pain originates from the teeth or not. Therefore, if from a patient perspective, it is understandable to ask for a dental evaluation after the appearance of the first symptoms; it is surprising from a neurological and neurosurgeon point of view that many patients with a clear history of idiopathic TN have their teeth extracted before a definitive diagnosis is made.

A paper appeared in 1983 by Garvan and Siegfried showed that 73 % of patients with trigeminal neuralgia had a dental assessment before diagnosis and that 65 % of them had a range from 1 to 32 teeth extracted (1). One should assume that 30 years after this study, the situation has changed with a higher recognition of the TN diagnosis and with less dental procedures administered.

Von Eckardstein et al. should be congratulated for their effort in trying to shed some light on this matter (2). They conducted a telephone interview on patients treated at their Institution for TN and found out

that, not surprisingly to me, in 82 % of them still, the first health-care provider was their dentist. However, only less then 5 % of the patients remembered that the dentist established the differential diagnosis of TN. Furthermore, 66 % of the patients received invasive dental procedures, even when the first branch of the trigeminal nerve was involved (1 case). Of the cases, firstly evaluated by a dentist, only 14 % was then referred to a neurologist or a neurosurgeon. More importantly, more than 2 years have passed before a neurological or neurosurgical evaluation in 42.5 % of the cases. An interesting part of the study regarded the questionnaire the authors sent to local dentist. Fifty-one percent of the dentist answered the questionnaire, stating that in 74.5 % of the cases, they felt competent enough to identify TN as a cause of orofacial pain, while only 50 % of them knew that carbamazepine and surgical intervention were treatment modalities for this disease. Furthermore, 76 % of the dentist stated that there is a need for further information and ongoing medical education on TN

The results of the paper by von Eckardstein et al. (2), even with the limitation related to the structure of the study and recognized by the authors themselves (telephone interview, no evaluation of dentist charts, etc.), are extremely important to capture the difficulties that still remains in 2014 on diagnosis and correct management of TN. There are two data that, in my opinion, should be particularly stressed: 1) almost one half of the patients waits 2 years before seeking for a neurologist or a neurosurgeon; 2) more than one half of the patients still undergoes invasive dental procedures before TN diagnosis is made. It is difficult to attribute responsibility for this situation. However, national and local health organizations should find ways to improve medical education on TN, particularly among general practitioners and dentist.

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Isao Date, Okayama, Japan

This is an interesting study showing that many patients of trigeminal neuralgia visit dentists first and many of them actually received unnecessary dental procedures. As the authors pointed out, it is a little bit surprising that the situation has not been changed so much for the past 30 or 40 years and very limited number of English publications could be found regarding this subject. Because the data are based upon telephone interviews and mails, this paper may not be scientifically high level in some sense. However, the data shown in this paper is quite practical for the daily neurosurgical practice and demonstrate the importance of communication between the medical doctors (especially neurologists and neurosurgeons) and dental doctors. Also, education of general public that there is a disease criterion of trigeminal neuralgia, which shows similar signs of dental diseases, should be promoted.

Ali Tayebi Meybodi, San Francisco, USA

The authors have touched a critical issue regarding the patients suffering from trigeminal neuralgia. As a neurosurgeon, I have seen many patients with trigeminal neuralgia who sought dental care at the beginning of their disease course. Surprisingly, a noticeable proportion of them received unnecessary dental treatment. This is in part because of the treacherous nature of the disease that mimics the more common dental conditions. However, the solution to this problem is not just providing stronger educational provision for our dentist colleagues. "Facial pain" should be discussed with more frequency and in more depth in joint meetings between all specialties involved in the field. This paper emphasizes the importance of what we recognize as "continuous medical education".

