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Abstract

Introduction

The complex nature of facial pain conditions creates a diagnostic challenge and often necessitates specialist referral.

Aim

To identify the case mix presenting to a specialist tertiary care facial pain clinic.

Methods

A retrospective review of 112 patient records was undertaken. Trends in provisional diagnoses from referrers and the correlation to diagnoses made following specialist consultation were reviewed.

Results

The most common provisional diagnoses recorded in referral letters were painful temporomandibular disorders, trigeminal neuralgia and persistent idiopathic facial pain. Over a quarter of referrals did not include a provisional diagnosis. Following assessment, only one case was not given a definitive diagnosis and no patients were diagnosed with persistent idiopathic facial pain (PIFP). A causative factor was identified in all the initially queried PIFP cases, and post-traumatic trigeminal neuropathic pain was found in multiple patients.

Conclusions

Post-traumatic trigeminal neuropathic pain should be considered if pain onset coincides with dental treatment or other traumatic events. PIFP is a rare facial pain diagnosis and may be over-diagnosed by dental and medical practitioners. It is important to systematically exclude other causes before reaching this diagnosis. This will facilitate effective treatment, manage patient expectations and potentially reduce unnecessary referrals.

Introduction

Facial pain is complex and diagnosis is difficult[1,2]. Achieving optimal management is dependent on each patient having the correct diagnosis of their facial pain to inform decisions about appropriate treatment modalities.

The impact of these conditions on patients should not be underestimated; frequently affecting their daily life. Patients have reported that persistent orofacial pain can have significant effects on their psychological health, work performance and ability to participate in social activities[3]. Studies have also shown that there is a financial impact of persistent orofacial pain to both the patient and the heath service, particularly among those with high Graded Chronic Pain scores[4,5]. This may be linked to the increased number of consultations, with these patients attending multiple appointments across different specialties to obtain a diagnosis and find the root cause of their pain[3,4].

Methods

A retrospective cross-sectional analysis was carried out by two clinicians using case notes for patients attending a specialist facial pain clinic between January 2017 and May 2018 at the Charles Clifford Dental Hospital, Sheffield, UK. This clinic only accepts referrals for patients from 16 years and older and is led by a Consultant Oral Surgeon. Of the 210 new patients seen within this time frame, 120 cases were selected from the clinic list using convenience sampling based on clinical record availability. Data collected included patient demographics (age, gender and medical history), referrer details (specialist, general medical practitioner (GMP) or dental practitioner (GDP)), the referrer's provisional diagnosis and definitive diagnosis following assessment on the specialist clinic. Patients seen with an absence of facial pain symptoms were excluded from the review.

The principle aims of this review were to identify trends in conditions treated on a specialist facial pain clinic and to assess the correlation between referrer provisional diagnosis and definitive diagnosis following specialist consultation.

Results

Demographics

8 patients were excluded due to inappropriate booking on the facial pain clinic (i.e. patients not presenting with facial pain), resulting in 112 patients included in this study. 90 (80%) patients were female, which aligns with the known demographics of patients who experience facial pain[6]. Age of patients at assessment ranged from 16 to 98 years old, with a peak between 41-70 years and in under 21 years old (Figure 1). The majority of younger patients (26 of the 27 patients under 41 years old) were those diagnosed with painful temporomandibular disorders (TMD).

Most referrals came from general dental practitioners (Figure 2), but almost a third were from other dental and medical specialties including Oral and Maxillofacial Surgery, Neurology and Ear, Nose and Throat. The duration of pain symptoms for most patients varied between a few months to years (mode one year; median three years). However, for some this was as much as 20 years (Figure 3), indicating that their diagnosis may have not been clarified and/or their treatment not satisfactorily optimised for a significant length of time.

Although most patients did not report medical comorbidities, chronic pain conditions such as fibromyalgia, chronic back pain and irritable bowel syndrome were found in 14% of patients. All but one of these patients was subsequently given a diagnosis of TMD.

Diagnoses

The most common provisional diagnoses recorded in referral letters were TMD (31% of referrals), neuropathic pain conditions (19%) and persistent idiopathic facial pain (PIFP) (17%) (Figure 4). Of the neuropathic pain category, the most common provisional diagnosis (13% of all referrals) was trigeminal neuralgia (TN) (Figure 5). 43% of these TN diagnoses were confirmed as the classical subtype due to vascular conflict with the trigeminal nerve on Magnetic Resonance Imaging (MRI).

Six referrals provided multiple provisional diagnoses, however over a quarter (26%) of referrals did not include any provisional orofacial pain diagnosis (Figure 4). This could indicate that referrers may feel unsure of the diagnosis and/or unconfident in making a diagnosis and

highlights the challenge of diagnosing facial pain conditions. 12 patients were diagnosed in the department as having multiple diagnoses contributing to their facial pain.

Provisional or differential diagnoses proposed by the referrer matched the diagnosis made in the specialist clinic in 43% of cases (48/112). Of the 35 patients referred with a provisional diagnosis of TMD, 34 were confirmed to have a diagnosis of TMD on the specialist clinic.

The most common definitive diagnoses were TMD which was found in 74% of patients, followed by neuropathic pain conditions which were identified in almost a quarter (24%) of patients seen (Figure 4). 84% of patients diagnosed with TMD were female.

A dental cause for pain was identified in 9% of cases which was over double the number referred with this as the provisional diagnosis (Figure 4). In the case of the patients diagnosed with dental pain, the duration they had been experiencing pain ranged from one month to 10 years. Half of the patients with dental pain also had TMD. Interestingly, all 10 patients who were diagnosed with a dental cause of pain on the specialist clinic were referred from GDPs, of which only two considered dental pain as their provisional diagnosis.

All 19 cases where the referrer had provisionally diagnosed PIFP, or used the former descriptor of “atypical facial pain”, were given an alternative facial pain diagnosis following specialist consultation, with the majority found to have TMD. No patients were given a final diagnosis of PIFP, but one case did not have a definitive diagnosis following assessment on the clinic (Figure 4). One patient was diagnosed with myositis and painful dry eyes (“other” category, Figure 4). This was included in this study as this caused facial pain, however they were referred to dermatology as a more appropriate management centre for this condition.

TN was the most commonly diagnosed neuropathic pain (Figure 5). Three cases were found to have idiopathic painful trigeminal neuropathy and eight cases were identified to have painful post-traumatic trigeminal neuropathy (PPTTN) following specialist assessment (Figure 5). However, no patients were referred with either of these as a provisional diagnosis.

Few patients were referred or diagnosed with primary headache conditions or burning mouth syndrome (BMS) (Figures 4, 5). This had been anticipated as there are other more appropriate referral pathways for these conditions (such as Neurology, Ear Nose & Throat and Oral Medicine) within this hospital trust.

Discussion

TMD was the most frequent diagnosis

These results concur with the literature that TMD is a common facial pain diagnosis often affecting female and younger patients than other types of facial pain[7]. However, the frequency of TMD as a diagnosis on this clinic was a surprising finding as the dental hospital also has specific TMD tertiary referral clinics.

TMD represents a group of disorders where the temporomandibular joint itself and/or the surrounding musculature are the sources of the facial pain[8]. Common presentations include hypertrophy and tenderness of the muscles of mastication, clicking, locking and/or crepitus within the temporomandibular joint[9, 10] and the classification of TMD conditions reflects this (Table 1). Nevertheless, other presentations may include headaches, perceived altered sensation and/or dental pain[7,9,10,11]. It was noted that 4 cases had headache symptoms but were identified as having TMD, which are likely to represent 'headache attributed to TMD' subclassifications of TMD (Table 1). However, the retrospective nature of this study relied on comprehensive record keeping and in the case of TMD it was not always clearly documented as to the exact subclassification, so further analysis of this was not possible.

It should be noted that this condition may present concomitantly or secondary to other longstanding facial pain conditions[7]. In this study, TMD was by far the most common facial pain diagnosis in patients with other chronic pain conditions. Previous studies have highlighted that chronic facial pain conditions may be comorbid with other bodily chronic pain including fibromyalgia, neck pain, back pain and irritable bowel syndrome[12-15]. Comorbidity of chronic widespread pain conditions has been demonstrated in TMD specifically[14,16,17] and the presence of such conditions is a risk factor in the development of chronic TMD[18]. Chronic widespread pain has also been found to be negatively associated with treatment outcomes from splint therapy[19]. Practitioners should be mindful of the more refractory nature of TMD in patients with widespread pain and ensure appropriate management is implemented early[20].

The high numbers of TMD diagnoses highlights the importance of referrers considering this and being aware of the varying presentations of TMD conditions. Many patients were correctly referred with a provisional diagnosis of TMD suggesting that there was awareness of this condition among referrers. However, in this study TMD represented 31% of provisional diagnoses by referrers, but 74% of definitive diagnoses made on the specialist clinic suggesting that many TMDs seen were not being identified early. This may be because of the variable and complex nature of TMD presentation that can confuse diagnosis and account for the lower proportion of TMD cases identified by referrers compared to the specialist clinic. This highlights the importance of a comprehensive examination of the temporomandibular joint and muscles of mastication to exclude this as a possible diagnosis, prior to referral to the specialist pain clinic. Further education for referring practitioners may help them improve their diagnostic decision-making skills[10].

It should also be noted that this review looked at the primary diagnoses following initial assessment on the facial pain clinic. Given that TMD can be comorbid with other facial pain disorders, it may be that a proportion of these patients also had subsequent diagnoses that were further investigated and identified after treatment of their TMD. This may also contribute to the high proportion of primary definitive diagnoses of TMD.

Dental pain was the fourth most common diagnosis made on the clinic

Dental pain is a very common complaint, affecting 9% of dentate adults in the UK[21]. Nonetheless, these results demonstrate how this diagnosis can sometimes be hard to reach, even among the dental profession, and could be complicated by coexisting pain conditions. Four patients in this cohort were found to have both dental pain and TMD secondary to bruxism. One patient was found to have TMD following a dental procedure. This highlights the role of a thorough history, as well as clinical assessment including the facial musculature, is necessary. Signs and symptoms of tongue scalloping, linea alba and generalised bilateral dental pain and/or burning tongue may all indicate a potential bruxism parafunctional habit[22,23].

PIFP was not seen in this cohort of facial pain patients

All patients referred with PIFP were found to have other causes of their pain following acquisition of a detailed history and clinical assessment on the specialist clinic. Previous study of the prevalence of PIFP in Germany also found it to be a very rare condition (0.03% lifetime prevalence) and far less common than TN[24]. PIFP has been shown to present a particular diagnostic challenge for dentists[25]. The diagnosis of PIFP is reached by exclusion of all other known causes of facial pain[6,26] (Table 2).

Limited consultation time for many primary care practitioners may present a barrier to diagnosis of facial pain conditions, as thorough history-taking and examination assessment is required to facilitate accurate diagnosis[1,2,27]. Moreover, chronic facial pain conditions may have a large psychological component[1] and can cause hypervigilance in some patients. This could create confusion for the practitioner in making a confident definitive diagnosis, particularly if the patient presents a complex pain history. Over-diagnosis of PIFP may also be related to practitioner knowledge of facial pain diagnoses and the updated classification of these conditions.

There is potential for this cohort of patients to either not be treated appropriately or be over investigated given the challenge with reaching a diagnosis of PIFP[28], particularly given the absence of a known cause for this condition.

It should be noted that one patient was not diagnosed following assessment on the clinic and could be considered to fall into the diagnostic category of PIFP. However, from discussion with this patient, the specialist clinician did not feel that attaching the label of PIFP would be helpful in this case. Use of the diagnosis of PIFP should be with caution as this label may risk practitioners overlooking other, potentially treatable, causes of chronic pain. Moreover, for the patients involved this may result in them not receiving a clear explanation of causation of their pain if another cause has been overlooked, adding to the psychological burden of chronic facial pain.

Trigeminal Neuralgia was the most common neuropathic pain diagnosis

Similar to TMD, the diagnostic accuracy of the referrers with TN was generally good, with nine of the 14 referrals correctly offering this diagnosis. This may be because TN usually has very distinct features of unilateral severe electric-shock pain that lasts less than two minutes and is abrupt in onset and termination and is confined within the area innervated by one or more divisions of the trigeminal nerve[6]. However, whilst many patients have characteristic presentations, sometimes there are other features that may confuse the clinician such as a persistent background facial pain (defined in ICHD-3 as concomitant continuous pain[6,20]. This may account for the 5 TN patients provisionally diagnosed with either unknown pain or PIFP.

TN is a clinical diagnosis which can be subclassified based on MRI findings as *classical* (evidence of vascular conflict at the dorsal root entry zone of the trigeminal nerve), *secondary* (due to space occupying lesions or central demyelinating conditions such as multiple sclerosis) or *idiopathic* (exclusion of a vascular or secondary cause)[20]. Subclassification may assist with surgical treatment planning for patients where medical treatment is suboptimal. For example, microvascular decompression is the most successful surgical treatment option, but this is only effective for the classical TN subgroup[20].

Painful post-traumatic trigeminal neuropathy and idiopathic painful trigeminal neuropathy were not identified by referrers

Interestingly three patients were found to have idiopathic painful trigeminal neuropathy. Unlike TN, this is not associated with triggers and is a constant sensory deficit within the distribution of the trigeminal nerve indicating it has been damaged[6]. Unlike PIFP, in this condition there is clinical evidence of dysfunction in the nerve, which may include burning (dysaesthesia), reduced sensation (hypoesthesia), and/or tingling sensation (paraesthesia) (Table 2). It is therefore similar to PPTTN, although without the history of a traumatic event. These key differences in the history and examination findings help distinguish these similar conditions.

PPTTN was not included in any provisional diagnoses but represented 7% of definitive diagnoses. PPTTN is a condition of trigeminal dysfunction within months of a traumatic event. It occurs secondary to neural trauma in the facial region (Table 2)[6,29]. The term “trauma” can

relate to events such as tooth extraction, root canal treatment, dental abscesses, avulsion or radiotherapy[6,29]. Whilst most patients will recover uneventfully from such experiences, some patients continue to experience pain, even in the presence of clinically normal tissue healing[29]. Previous studies have suggested the prevalence may be as high as 3% of cases of trigeminal nerve injury[30] – a particularly alarming statistic given the number of dental procedures patients undergo. In these cases, patients can identify the onset of their pain correlating to the traumatic event[6,29,30]. However, it has been acknowledged that the signs of PPTTN may be subtle and may overlap with symptoms of other neuralgias or odontogenic pain[28].

The International Classification of Headache Disorders (ICHD-3) classify PIFP and PPTTN in completely separate categories (Table 2)[6], as does the recently published International Classification of Orofacial Pain (ICOP-1)[31]. One key difference in the ICOP-1 is that atypical odontalgia has been reclassified from a subgroup of PIFP to a separate category of persistent idiopathic dentoalveolar pain[31]. However it is acknowledged that atypical odontalgia symptoms, whilst generally categorised alongside PIFP, may be alternatively considered a subgroup of PPTTN when there is a history of trauma[6]. This similarity between these conditions highlights the need to establish a clear timeline of events during a chronic facial pain history to ensure that PPTTN is ruled out before concluding it is idiopathic. Patients may or may not have associated a traumatic cause to the onset of their pain. To elicit this level of detail from the pain history requires a systematic and thorough approach. In this study, we identified three cases that were provisionally diagnosed with PIFP but were found to have PPTTN following the specialist clinic assessment.

With persistent idiopathic trigeminal neuropathy, PPTTN and PIFP conditions, management is challenging, and recommended medicinal treatments are similar. Some practitioners may feel that such semantics will not affect the medical management of the pain, and therefore may not perceive a benefit in acknowledging the traumatic history in PPTTN over PIFP or persistent idiopathic trigeminal neuropathy. However, this classification may assist with informing patients and developing their understanding of their pain. In the sociological literature, it has been demonstrated that conditions with a perceived physical origin (for example within facial pain this may be a diagnosis of trigeminal neuralgia due to vascular compression of the trigeminal nerve) are considered to be less controllable and elicit a more

sympathetic and supportive response from others than those of mental-behavioural origin[32]. With the diagnosis of PIFP, the diagnostic features being a lack of identifiable cause would place this in the category of chronic pain conditions of medically unexplained symptoms. Patients may struggle with this diagnosis as they may be perceived as being psychologically-created by the patient and feel stigmatised[33]. Such labelling may attribute a level of blame and thus inhibit patient acceptance of pain. Pain acceptance is a well-recognised attribute to adjustment and functional improvement[34]. It is important that clinicians remove any sense of blame to empower patients to construct an illness narrative to aid their understanding and acceptance of pain[35].

In contrast, the cause and effect relationship of trauma with PPTTN is simple and logical to those who have experienced the pain. This may help to reassure patients and increase their ability to reach an acceptance with their pain. Giving patients a tangible mechanism of causation has been identified to improve their psychological assimilation of medically unexplained symptoms[35]. However further research into patients' perception of their diagnosis in the field of chronic facial pain is needed.

Limitations

The sample size was restricted due to the significant demands that service evaluation projects place on the clinical records team. Whilst a reasonable sample size was obtained, a larger sample of patients seen within a year may be more representative and increase reliability of the results. Including all patients seen, rather than using convenience sampling, may avoid any unintended selection bias. This area would therefore benefit from a more in depth, prospective study.

Furthermore, the retrospective nature of the data collection limited the depth of data capture in the study. For example, subclassifications of TMD were not analysed as there was variable recording of the category of TMD. Further work prospectively could utilise proformas to ensure that category of TMD (Table 1) is clearly recorded for every case where TMD is diagnosed. This could help identify whether there are trends in the type of TMD when this is comorbid with other conditions, such as dental pain.

Using a retrospective data collection technique also meant that no data was collected on the biopsychosocial aspects of the conditions seen. This data is not routinely collected on these clinics so was not available as part of this retrospective review. Previous research has identified the negative impact of facial pain on patients' quality of life[36], and existing validated questionnaires such as those used in the DC/TMD Axis-II[37,38] could shed further light on the patient experience in a future prospective assessment of this clinic.

This data only represents the experience of one centre in the UK. The varied referral pathways used in different tertiary hospitals may limit the generalisability of these findings.

As the review did not question referring practitioners, understanding of their provisional diagnosis and their knowledge of facial pain diagnoses was limited by the information included in the referral letters. No referrals mentioned PPTTN or idiopathic painful trigeminal neuropathy, but it is unclear whether this was because practitioners had actively excluded them or been unaware or unconfident of these diagnoses.

Moreover, as this review did not involve direct questioning of patients, or further analysis of patient records, it was not possible to ascertain whether any discussion or treatment had been provided successfully in primary or secondary care prior to referral. Further research is recommended to assess patients' perception of the impact of having a formal diagnosis and whether they found diagnosis of a causative factor (such as in TMD or PPTTN) was more helpful and more accepted than PIFP.

Conclusions

To our knowledge, this is the first review to specifically highlight the varied nature of facial pain presentations seen within a UK specialist facial pain clinic. It is clear from these findings that specialist facial pain clinics are an important referral service to support patients with diverse and complicated facial pain presentations. This review also highlights the need for further prospective research into the patients attending specialist facial pain clinics to enable concurrent evaluation and comparison of the biopsychosocial aspects of the conditions seen.

TMD is a common presentation and should be considered by practitioners within their differential diagnosis when assessing a patient with facial pain. Dental and medical practitioners should be aware of first-line management techniques for TMD; including educational advice, exercises and splint therapy[8]. These conservative treatments can be provided within a primary care setting and prevent unnecessary delays in pain management for patients. Clinicians should also consider the potential for concomitant pain disorders that may result in confusing pain histories.

In contrast, these results suggest possible over-diagnosis of PIFP, and that this may be a rarer facial pain diagnosis. Sometimes, despite investigating at great length, we are unable to diagnose facial pain, and may term this atypical or idiopathic facial pain. However, practitioners should be aware of the importance of identifying precipitating factors, such as dental treatment, that coincided with the pain development as this can help with PPTTN identification. PPTTN was an alternative diagnosis identified in several cases by the specialist clinic and it is important to exclude other causes before reaching a diagnosis of PIFP. This will facilitate more rapid delivery of effective treatment, manage patient expectations and potentially reduce unnecessary referrals.

Declaration of interests

The authors declared no potential conflicts of interest with respect to the research, funding, authorship, and/or publication of this article.

In brief

- Highlights the importance of excluding other causes before diagnosing persistent idiopathic facial pain
- Demonstrates the under-diagnosis of temporomandibular joint dysfunction among referrals
- Highlights the potential for dental surgery interventions to result in painful post-traumatic trigeminal neuralgia

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