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The specialist management of non-temporomandibular orofacial pain: maxillofacial surgery's known unknown?

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Abstract

The management of orofacial pain is considered to be within the remit of oral and maxillofacial surgery (OMFS). In this study we aimed to provide an overview of the healthcare “journey” of a group of patients referred to a specialist unit with “complex” non-temporomandibular orofacial pain. We retrospectively reviewed all those who were referred over a six-month period and followed for up to three years. A total of 133 were included, 69% were female, and the mean (SD) duration of symptoms before assessment was 66.4 (88.8) months. Patients were treated for seven different conditions by a mean (SD) of 2.6 (1.2) specialties, and 3.2 (2.5) medications had been tried before assessment at the unit. A mean (SD) of 3.9 (3.3) appointments were attended over the three years, and 80% of patients were prescribed at least one medication. Patients were under the care of the unit for a mean (SD) of 11.9 (14.0) months, and 38% were still being seen at three years. Those with trigeminal neuralgia were most likely to remain in care at three years ($p < 0.001$), and those with burning mouth syndrome ($p < 0.001$) or persistent idiopathic facial pain ($p = 0.005$) were most likely to be discharged. In the current NHS climate, the lack of resources to treat facial pain and the focus on a mix of skills, mean that OMF surgeons are likely to have an increasing role in the treatment of these patients. This paper provides an important insight into these conditions.

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Introduction

Orofacial pain affects up to 7% of the population,¹ and risk factors include age, anxiety about health, and the presence of chronic pain elsewhere.² In the UK its management is considered to be within the remit of oral and maxillofacial surgery (OMFS), as the service is available at most district general hospitals.

Odontogenic pain and disorders of the temporomandibular joint (TMJ) are well known and common,³ but several other orofacial pain syndromes are relevant to OMFS.^{1–4} We have

defined them as “complex” for the purpose of this paper. They can produce intense pain that may have unusual physical or pathological manifestations,^{5,6} and their management often requires a multidisciplinary approach.

Trigeminal neuralgia, which is characterised by a spontaneous, brief, unilateral, electric shock-like pain in the trigeminal distribution,⁴ and burning mouth syndrome, which is characterised by an intraoral burning or dysaesthetic sensation, are probably the best known. Trigeminal neuropathic pain affects the trigeminal region and varies in quality and intensity because of its inflammatory causes - for example, post-herpetic neuralgia, and trauma - most commonly from extractions, root canal work, and facial injuries.⁷

Trigeminal autonomic cephalalgias form a group of unilateral pain syndromes that affect the trigeminal region, and have various ipsilateral autonomic symptoms.⁴ Persistent idio-

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pathic facial pain (previously known as atypical facial pain) lacks any characteristic pattern of neuralgia and is a diagnosis of exclusion,⁸ while atypical odontalgia is characterised by dental pain in the absence of any physical or pathological change, and may be a form of neuropathic pain.⁴

Multidisciplinary treatment provides holistic care in these complex cases,^{1,3,9} but as local resources are often limited, management that is coordinated by OMFS may be the most feasible option.

The aim of this paper was to provide an overview for OMF surgeons of the healthcare “journey” of a group of patients who were referred to a specialist facial pain unit with complex orofacial pain, and the resources that were used.

Patients and methods

We retrospectively assessed all new referrals to a specialist facial pain unit at a London teaching hospital over a six-month period. The unit sees 700 new patients (approximately half of whom have disorders of the TMJ) each year from a wide catchment area.⁹ We included patients with complex primary orofacial pain who were aged over 17 years at the time of assessment. “Complex” was used to define pain that was not odontogenic or caused by a disorder of the TMJ, and “primary” was regarded as the condition that was chiefly responsible for the presenting symptoms. “Orofacial pain” was defined as any pain affecting the oral structures or the facial region below the orbitomeatal line, or both, above the neck and anterior to the ears.

Fig. 1 summarises the orofacial pain care pathway that is followed at our unit. It involves an initial one-hour assessment with multidisciplinary review if necessary. The core members of the team come from psychology, psychiatry, and physiotherapy, and there is access to restorative dentistry, oral surgery, and complementary and alternative medicine, if required. There are also joint clinics with neurosurgery and headache neurology.

The diagnostic process involves a meticulous pain history followed by a comprehensive examination of the head and neck.⁹ The use of questionnaires such as the Brief Pain Inventory or the McGill Pain Questionnaire can aid assessment,³ but laboratory investigations are not of great importance unless an autoimmune condition or cranial arteritis is suspected. Radiographs are useful for excluding odontogenic causes, while magnetic resonance imaging (MRI) can distinguish between primary and secondary trigeminal neuralgia. Computed tomography (CT) and ultrasound are used less often.

Generally, new patients are reviewed at around three months or referred to joint psychological-physiotherapy services, although the patients included in this study tended to be less typical. The psychology care package involves a group information session, individual psychological assessment, and individual or group treatment over six weeks with written information provided throughout. Psychometrically-tested questionnaires are completed at each visit, and patients reviewed annually.

We used clinical records to assess the healthcare “journey” of patients from their point of entry through to their assessment and subsequent management over the next three years. The information was recorded in Microsoft Excel. Independent *t* tests with one-tailed unequal variance were used for quantitative data such as the mean number of appointments or duration of symptoms. Probabilities of less than 0.05 were considered significant.

Results

Demographics and diagnosis

A total of 321 new patients with orofacial pain were seen over the six-month period. Of them, 186 were excluded as their primary cause of pain was odontogenic or related to

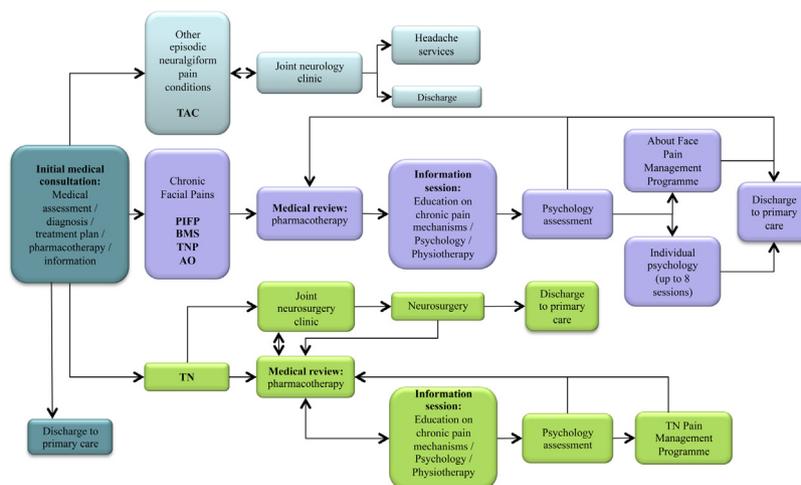


Fig. 1. Orofacial pain care pathway (TAC = trigeminal autonomic cephalgias; PIFP = persistent idiopathic facial pain; BMS = burning mouth syndrome; TNP = trigeminal neuropathic pain; AO = atypical odontalgia; TN = trigeminal neuralgia).

the TMJ. A further two were excluded because of age. A total of 133 were therefore included (mean (range) age 52.6 (18.2–82.6) years) and a 69% were female. According to the National Readership Survey (NRS) social grading system¹⁰ 53% were classed as category E (retired or unemployed), and only 5% as category A (professionals or higher managerial). Only 6% were from an NRS grade A or B postcode, and 31% were based in London.

Table 1 summarises the overall findings. The mean (SD) duration of symptoms before assessment was 66.4 (88.8) months, and patients with trigeminal autonomic cephalalgias ($t=3.3$, $df=30$, $p=0.001$) and trigeminal neuropathic pain ($t=2.3$, $df=157$, $p=0.01$) were referred earlier. A diagnosis of secondary orofacial pain was given to 20% of patients, and to 17% who had a psychiatric diagnosis (including anxiety, depression, and personality disorders). A further 21% were diagnosed with both. Only 23% had an unremarkable medical history.

Point of entry into healthcare

General dental practitioners made 31% of referrals, general medical practitioners 28%, and OMFS 11%, despite the fact that 41% of the patients had previously been treated by the specialty. Patients had seen a mean (SD) of 2.6 (1.2) specialties before referral (Table 2).

Patients were prescribed a mean (SD) of 3.2 (2.5) medications. These included eight types of antiepileptic drugs ($n=144$), 13 antidepressants ($n=112$), seven opioid ($n=48$) and nine non-opioid analgesics ($n=65$), and 31 other medications ($n=53$) that included benzodiazepines, antihistamines, triptans, and hypnotics. Most patients (55%) were given at least one antiepileptic drug (mean = 2.0), and half at least one antidepressant (mean = 1.7), while those with atypical odontalgia were prescribed fewer medications ($t=2.3$, $df=34$, $p=0.01$).

Assessment of pain to end of follow up

Patients attended a mean (SD) of 3.9 (3.3) appointments and 80% of patients were prescribed at least one medication. Most of those managed without drugs (17/27) had atypical odontalgia or persistent idiopathic facial pain. Table 3 summarises the prescribed medications. Antiepileptic drugs and antidepressants accounted for 45% ($n=86$) and 40% ($n=76$) of the total prescriptions, respectively, and analgesia accounted for only 9% ($n=18$).

Atypical odontalgia ($t=3.8$, $df=51$, $p<0.001$) and burning mouth syndrome ($t=2.2$, $df=10$, $p=0.02$) were managed with fewer medications, but trigeminal neuropathic pain required more ($t=2.0$, $df=49$, $p=0.03$). Patients with trigeminal neuralgia were almost exclusively managed with antiepileptic drugs ($t=4.5$, $df=39$, $p<0.001$).

After assessment, 53% of patients were given review appointments, 31% were given open appointments that allowed them to return as required within one year, and 11%

Table 1
Summary of results for each diagnosis of orofacial pain.

Primary diagnosis	No. of patients	Before assessment			After assessment				
		Mean duration of symptoms (months)	Mean number of specialties seen	Mean number of medications prescribed for pain	Mean number of medications prescribed		Mean time under care (months)		
					AED	AD		All	
Atypical odontalgia	25	52.1	2.4	1.9*	0.1	0.6	0.8*	4.1	13.4
Burning mouth syndrome	9	73.2	2.8	3.1	0.0	0.4	0.8*	2.1*	3.3*
Persistent idiopathic facial pain	27	90.0	3.0	4.1	0.2	0.8	1.1	2.7*	5.6*
Neuropathic pain	32	45.5*	2.5	3.5	0.8	0.8	1.9*	3.3	8.3
Other (headaches, migraines)	5	85.2	2.6	3.0	0.4	0.8	1.8	3.2	4.5*
Trigeminal autonomic cephalalgias	7	32.6*	2.6	4.7	1.4	0.4	2.9	6.9*	11.8
Trigeminal neuralgia	28	63.5	2.4	2.7	1.5*	0.1	1.6	5.4*	24.7
All primary diagnoses	133	88.8	2.6	3.2	0.6	0.6	1.4	3.9	11.9

AED: antiepileptic drugs.
AD: antidepressants.

* $p < 0.05$: each primary diagnosis was compared with the mean for the entire group.

Table 2
Specialties seen before assessment.

Specialty	No. (%) of patients
General dental practitioner	72 (54)
General medical practitioner	66 (50)
Oral and maxillofacial surgery	55 (41)
Neurology	34 (26)
Dental specialist	24 (18)
Neurosurgery	17 (13)
Ear, nose and throat	17 (13)
Complementary and alternative medicine	12 (9)
Pain management	10 (8)
Oral medicine	9 (7)
Psychiatry	7 (5)
Other	7 (5)
Accident and emergency	5 (4)
Physiotherapy*	5 (4)
Psychology*	4 (3)
Ophthalmology	2 (2)

* Requires a directive from a dental or medical practitioner.

Table 3
Prescribed medications used by the facial pain team over the three-year follow-up period.

Medication	No. (%) of prescriptions
Amitriptyline	8 (4)
Carbamazepine	17 (9)
Citalopram	5 (3)
Dosulepin	6 (3)
Duloxetine	4 (2)
Escitalopram	27 (14)
Gabapentin	11 (6)
Lamotrigine	13 (7)
Mirtazapine	1 (1)
Nortriptyline	24 (13)
Oxcarbazepine	17 (9)
Phenytoin	2 (1)
Pregabalin	24 (13)
Sertraline	1 (1)
Topiramate	2 (1)
Non-steroidal anti-inflammatory drugs	18 (9)
Other	11 (6)

were referred to other specialties for the coordination of care. Only 5% were discharged.

MRI was the most common investigation (32%, $n = 43$), followed by blood tests (28%, $n = 37$) and radiography (26%, $n = 34$). A minority of 15% ($n = 19$) had other investigations including CT, ultrasound, or neurophysiological tests. The condition investigated most extensively was trigeminal neuralgia ($t = 2.7$, $df = 36$, $p < 0.005$), while persistent idiopathic facial pain ($t = 1.8$, $df = 47$, $p = 0.04$) and burning mouth syndrome ($t = 2.5$, $df = 25$, $p = 0.01$) tended to be diagnosed and managed clinically.

Of the 515 follow-up appointments, 58% were reviews, 36% were with other members of the multidisciplinary team (Table 4), and 6% were emergencies. Patients with trigeminal neuralgia ($t = 2.0$, $df = 36$, $p = 0.03$) and trigeminal autonomic cephalalgias ($t = 3.1$, $df = 7$, $p = 0.008$) needed significantly more appointments, whereas those with persistent idiopathic facial pain ($t = 1.9$, $df = 43$, $p = 0.03$) and burning mouth syn-

Table 4
Other resources used by the facial pain multidisciplinary team.

Specialty	No. (%) of patients
Psychology	39 (29)
Neurology	16 (12)
Restorative dentistry	12 (9)
Complementary and alternative medicine	12 (9)
Neurosurgery	9 (7)
Psychiatry	7 (5)
Other (OMFS, oral medicine)	5 (4)

drome ($t = 3.3$, $df = 15$, $p = 0.002$) required fewer (11/27 of those with persistent idiopathic facial pain, and 4/9 of those with burning mouth syndrome attended the initial assessment only).

Patients were under the care of the multidisciplinary team for a mean (SD) of 11.9 (14.0) months, and 38% were still under its care at three years. The remaining 62% were discharged for various reasons including failure to make an open appointment by one year (40%), non-attendance (15%), and resolution of symptoms (8%). Patients with trigeminal neuralgia were most likely to remain under care at three years ($t = 4.0$, $df = 37$, $p < 0.001$), while those with burning mouth syndrome ($t = 4.9$, $df = 29$, $p < 0.001$) and persistent idiopathic facial pain ($t = 2.7$, $df = 49$, $p = 0.005$) were more likely to be discharged.

Discussion

To the best of our knowledge, this is the first reported UK study to assess the care pathway of patients with orofacial pain that is not odontogenic or caused by disorders of the TMJ. Beecroft et al¹¹ studied 101 referrals to OMFS for orofacial pain but as most of them had disorders of the TMJ (75%), their findings may not be comparable.

The demographics of our patients showed that orofacial pain was more prevalent in women, the middle-aged, and those of lower socioeconomic class.¹² These findings are similar to those in a large survey of over 50 000 people, the UK Biobank Study,¹³ which estimated a 1.9% prevalence of self-reported facial pain. Of this, 48% of patients had chronic pain, and the prevalence was highest in women and the middle-aged.¹³ The association that these characteristics have with the development of orofacial pain is poorly understood, but a complex interaction between physical and psychosocial factors is implicated.¹⁰ The fact that less than one-third of patients had a London postcode indicated geographical diversity and a lack of local resources.

The mean duration of symptoms before referral was over four years, but this was shorter in patients with trigeminal autonomic cephalalgias and trigeminal neuropathic pain, possibly because of a lower threshold for referral on account of the atypical autonomic features or possible malignant causes. The lack of adequate care pathways adds to the distress these patients experience.¹⁴

The UK Biobank Study found significant associations with psychological distress, and many patients in our group also had mental-health conditions.^{1,2,13} The complexity of our group was emphasised by the fact that a large proportion of the patients had diagnoses of secondary orofacial pain and chronic pain elsewhere, a finding that was also reported in the UK Biobank Study.¹³

As expected, most referrals were from primary-care practitioners,¹⁵ and evidence suggests that they generally have enough specialist knowledge to refer appropriately.¹⁶ The low referral rate from OMFS may indicate a lack of access to, or unawareness of, the resources available, though consultant-to-consultant referrals are often discouraged.

In their economic evaluation of orofacial pain, Durham et al showed that repeat referrals accounted for the highest cost of care.¹⁷ Ideally, more could be invested to develop specialist services to manage these patients who often require multidisciplinary care. The management of orofacial pain by OMFS is difficult because it is a surgical specialty with considerably shorter appointment times, and there is often limited access to specialist resources such as psychology and liaison psychiatry. Oral physicians with both medical and dental training may be more appropriate, but they are based only in dental schools. It may therefore be important to encourage OMF surgeons to seek professional development in this area.

The mean number of medications and specialties that were seen before assessment was comparable to those reported by Beecroft et al¹¹ and Zakrzewska et al.¹⁸ Patients with atypical odontalgia were prescribed fewer medications, but as the condition can mimic dental pain and there is a lack of guidelines, dental treatments may have been inappropriate.⁷ This has reinforced the need for research into the care pathways for orofacial pain so that they become more defined and efficient.¹⁷

Assessment of the management by facial-pain teams has shown that patients with trigeminal neuropathic pain and trigeminal neuralgia are the most likely to be treated with medication, as guidance is readily available for both.^{19,20} Atypical odontalgia is now considered a neuropathic pain so the relevant guidelines may also be appropriate.⁷

Over half of patients require a multidisciplinary review,^{1,3,14} and specialties other than that of facial pain provide one-third of the total appointments. While secondary care environments may lack access to resources such as complementary and alternative medicine, which are found in tertiary care, OMF surgeons in district general hospitals could develop closer links with pain medicine and headache neurology to increase their ability to provide more multidisciplinary care.

Patients with trigeminal neuralgia were generally kept under review because of the rarity of the condition and the severity of the pain, but also because of close links with the neurological team and the research interests of the clinicians. Patients with persistent idiopathic facial pain were most likely to be discharged after medical management that

had followed the care pathway of the unit, with or without a pain-management programme.²¹

Patients with trigeminal neuralgia and trigeminal autonomic cephalalgias used the greatest number of follow-up appointments, probably because of the severity of their symptoms. This again highlights the need to establish more effective care pathways, which could be based on those followed by the Danish headache service.²² Patients with burning mouth syndrome and persistent idiopathic facial pain required fewer appointments, possibly because their pain was less severe²³ and they had been given a careful explanation of their condition.⁹

Those with trigeminal neuralgia were most likely to have MRI to rule out symptomatic causes.²² In the other groups, investigations were done only in those with sensory changes that could not be explained from the clinical history.

In our group, patients remained under the care of the facial pain multidisciplinary team for a considerable amount of time, and over one-third remained under regular review at three years (particularly those with trigeminal neuralgia). Only a small minority was discharged with symptoms or with a treatment plan they could manage with their general practitioner. Most were discharged because they failed to make an appointment or did not attend, and we cannot assume that their condition had improved.

When we take into consideration the 60-minute slots, together with the number of appointments and the multidisciplinary services that are needed, a huge amount of resources are required. Nevertheless, some patients could be managed in secondary care if departments followed published care pathways and provided adequate time.

The main limitation of this study is its retrospective nature, which relied on the quality of the clinical records. Not all the data from the questionnaires were used.

In conclusion, the considerable variation in management incurs a substantial cost to the NHS and suggests a poor understanding of the conditions and the probable benefits of developing care pathways. The complexity of the symptoms, compounded by coexisting conditions, justifies the need for holistic, multidisciplinary care, and the large proportion of patients who remain under long-term care warrants the development of additional services. However, with the financial constraints of the NHS and the current focus on “skill-mix”, the role of OMFS will become increasingly important, and professional development with improvements in local services may provide the best solution.

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Ethics statement/confirmation of patients' permission

Ethics approval not required. Patients' permission/consent not required.

Conflict of interest

We have no conflicts of interest.

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References

- Zakrzewska JM. Multi-dimensionality of chronic pain of the oral cavity and face. *J Headache Pain* 2013;**14**:37.
- Aggarwal VR, Macfarlane GJ, Farragher TM, et al. Risk factors for onset of chronic oro-facial pain—results of the North Cheshire oro-facial pain prospective population study. *Pain* 2010;**149**:354–9.
- Zakrzewska JM. Differential diagnosis of facial pain and guidelines for management. *Br J Anaesth* 2013;**111**:95–104.
- Headache Classification Committee of the International Headache Society (IHS). The international classification of headache disorders, 3rd edition. *Cephalalgia* 2018;**38**:1–211.
- Aggarwal VR, McBeth J, Zakrzewska JM, et al. The epidemiology of chronic syndromes that are frequently unexplained: do they have common associated factors? *Int J Epidemiol* 2006;**35**:468–76.
- Aggarwal VR, McBeth J, Zakrzewska JM, et al. Are reports of mechanical dysfunction in chronic oro-facial pain related to somatisation? A population based study. *Eur J Pain* 2008;**12**:501–7.
- Baad-Hansen L, Benoliel R. Neuropathic orofacial pain: facts and fiction. *Cephalalgia* 2017;**37**:670–9.
- Mueller D, Obermann M, Yoon MS, et al. Prevalence of trigeminal neuralgia and persistent idiopathic facial pain: a population-based study. *Cephalalgia* 2011;**31**:1542–8.
- Napeñas JJ, Nussbaum ML, Eghtessad M, et al. Patients' satisfaction after a comprehensive assessment for complex chronic facial pain at a specialised unit: results from a prospective audit. *Br Dent J* 2011;**211**:E24.
- National Readership Survey. Social grade. Available from URL: <http://www.nrs.co.uk/nrs-print/lifestyle-and-classification-data/social-grade/> (last accessed 14 January 2019).
- Beecroft EV, Durham J, Thomson P. Retrospective examination of the healthcare “journey” of chronic orofacial pain patients referred to oral and maxillofacial surgery. *Br Dent J* 2013;**214**:E12.
- Adamo D, Celentano A, Ruoppo E, et al. The relationship between sociodemographic characteristics and clinical features in burning mouth syndrome. *Pain Med* 2015;**16**:2171–9.
- Macfarlane TV, Beasley M, Macfarlane GJ. Self-reported facial pain in UK Biobank Study: prevalence and associated factors. *J Oral Maxillofac Res* 2014;**5**:e2.
- Bonathan CJ, Zakrzewska JM, Love J, et al. Beliefs and distress about orofacial pain: patient journey through a specialist pain consultation. *J Oral Facial Pain Headache* 2014;**28**:223–32.
- Lang M, Selvadurai T, Zakrzewska JM. Referrals to a facial pain service. *Br Dent J* 2016;**220**:345–8.
- Aggarwal VR, Joughin A, Zakrzewska JM, et al. Dentists' and specialists' knowledge of chronic orofacial pain: results from a continuing professional development survey. *Prim Dent Care* 2011;**18**:41–4.
- Durham J, Shen J, Breckons M, et al. Healthcare cost and impact of persistent orofacial pain: The DEEP Study Cohort. *J Dent Res* 2016;**95**:1147–54.
- Zakrzewska JM, Wu J, Mon-Williams M, et al. Evaluating the impact of trigeminal neuralgia. *Pain* 2017;**158**:1166–74.
- Neuropathic pain in adults: pharmacological management in non-specialist settings. Clinical guidance 173, November 2013, updated April 2018. Available from URL: <https://www.nice.org.uk/guidance/cg173> (last accessed 14 January 2019).
- Crucchi G, Gronseth G, Alksne J, et al. AAN-EFNS guidelines on trigeminal neuralgia management. *Eur J Neurol* 2008;**15**:1013–28.
- Zakrzewska JM. Chronic/persistent idiopathic facial pain. *Neurosurg Clin N Am* 2016;**27**:345–51.
- Heinskou T, Maarbjerg S, RoCHAT P, et al. Trigeminal neuralgia—a coherent cross-specialty management program. *J Headache Pain* 2015;**16**:66.
- Komiyama O, Obara R, Uchida T, et al. Pain intensity and psychosocial characteristics of patients with burning mouth syndrome and trigeminal neuralgia. *J Oral Sci* 2012;**54**:321–7.