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ORIGINAL REPORT: QUALITATIVE RESEARCH

Care Pathways in Persistent Orofacial Pain: Qualitative Evidence from the DEEP Study

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Abstract: *Persistent orofacial pain is relatively common and known to have an adverse effect on quality of life. Previous studies suggest that the current care pathway may be problematic, but it is not well understood which health services patients access and what their experience is. The aim of this study was to explore care pathways and their impact from the perspective of patients. Qualitative interviews were conducted with a maximum variation sample of patients recruited from primary (community based) and secondary (specialist hospital based) care in the United Kingdom. Questions focused on the stages in their pathway and the impact of the care that they had received. Interviews were digitally recorded and transcribed verbatim, and analysis followed principles of the constant comparative method. NVivo 10 was used to help organize and analyze data. Twenty-two patients were interviewed at baseline, and 18 took part in a second interview at 12 mo. Three main themes emerged from the data: the "fluidity of the care pathway," in which patients described moving among health care providers in attempts to have their pain diagnosed and managed, occurring alongside a "failure to progress," where despite multiple appointments, patients described frustration at delays in obtaining a diagnosis and effective treatment for their pain. Throughout their care pathways, patients described the "effects of unmanaged pain," where the longer the pain went unmanaged, the greater its potential to negatively affect their lives. Findings of this study suggest that the current care pathway is inefficient and fails to meet patient needs. Future work needs to focus on working with stakeholder groups to redesign patient-centered care pathways.*

Knowledge Transfer Statement: *Data from qualitative interviews conducted with patients with persistent orofacial pain suggest significant problems with the existing care pathway, consisting of delays to diagnosis, treatment, and referral. Patients describing their struggle to progress through the current care pathway highlighted the difficulties occurring while living with orofacial pain. This study suggests a need for a revised care pathway, which better meets the needs of people with persistent orofacial pain.*

Keywords: chronic pain, facial pain, health services research, longitudinal studies, primary health care, secondary care

Introduction

Persistent orofacial pain refers to a group of chronic disorders affecting the face and mouth and includes temporomandibular disorder, burning mouth syndrome, trigeminal neuralgia, persistent dentoalveolar pain disorder, and persistent idiopathic facial pain (Benoliel and Sharav 2010). Persistent orofacial pain is relatively common, affecting 7% of the UK population (Aggarwal et al. 2010), and it is known to cause substantial impacts on quality of life (John et al. 2006; Shueb et al. 2015). Despite its prevalence, there is a limited understanding of patients' experiences of persistent orofacial pain and the impact that it has on their everyday lives. The small amount of qualitative data present in the literature—focusing on persistent

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orofacial pain collectively (Wolf et al. 2006, 2008; Mohr et al. 2011) and on singular specific persistent orofacial pain conditions (Hakeberg et al. 2003; Peters et al. 2005; Durham et al. 2010; Durham et al. 2012)—suggests multiple impacts on several areas of a person's life, including social, emotional, and financial aspects of everyday life. Some of the same research also highlights that patients experience difficulties in communicating their pain to doctors, potentially leading to problematic relationships (Wolf et al. 2008; Mohr et al. 2011; Durham et al. 2012). However, there has been no longitudinal concurrent quantitative and qualitative examination of patients' costs and experiences during their journey in a care pathway for persistent orofacial pain—that is, a concurrent "quan-qual" approach to understanding care pathways better (Morgan 1998; Durham et al. 2016).

Management of persistent orofacial pain spans both medicine and dentistry and multiple specialties and, in common with other chronic pain disorders, requires a multidisciplinary approach to management. Madland and Feinmann (2001) hypothesize that the location of orofacial pain may be a barrier to its management, suggesting that while the location of the pain implies the requirement of dental services, the pain may share qualities with other medical chronic illnesses, which need >1 clinical specialty or profession's input. This may represent a problem for patients, however, as medical and dental practitioners have expressed substantial uncertainty about how to manage persistent orofacial pain when it presents to them (Durham et al. 2007; Aggarwal, Joughin, Zakrzewska, Appelbe, and Tickle 2011; Durham et al. 2011; Peters et al. 2015) and there are few services available to provide specialist advice on management (Aggarwal, Joughin, Zakrzewska, Appelbe, and Tickle 2011).

Patients with persistent orofacial pain are known to use more health care services when compared with other groups of patients (White et al. 2001), which often includes multiple consultations and diagnostic tests (Glaros et al. 1995). Durham and colleagues' (2011) study of temporomandibular disorder care suggests that some of this increased health care resource utilization may be accounted for by inadequate care pathways resulting in cyclical referrals and unnecessary consultations. This finding is supported by recently published data (Peters et al. 2015) suggesting that patients and health care professionals find persistent orofacial pain difficult and frustrating to manage. Despite these data, there is limited understanding of how and why patients access health care services over the longer-term management of their condition and what their longitudinal experiences are of this care pathway. This article seeks to address this gap in knowledge by critically examining patients' use and experience of health care for persistent orofacial pain over a 12-mo period and link this to the recently published cost analysis (quantitative data from the concurrent quan-qual approach of the DEEP study) from the same cohort (Durham et al. 2016).

Methods

Research Question

The aim of this qualitative study was to critically examine patients' journeys through care, identify their experiences of the care pathway, and use these findings to help explain some of the findings in the cost analysis of the care pathways that ran concurrent to this substudy (Durham et al. 2016).

Study Design

The full protocol of the DEEP study (Developing Effective and Efficient Care Pathways for Patients with Chronic Pain; http://research.ncl.ac.uk/deepstudy) is available online in open-access format (Durham et al. 2014), so a full description of the study will not be given. Briefly, the aim of the DEEP study was to use mixed methods to describe and understand current care pathways for persistent orofacial pain patients and identify areas where the current pathways could be improved. Following ethical approval (NHS Research Ethics Committee NRES reference 12/YH/0338), serial quantitative data based on validated self-complete instruments were collected from patients with persistent orofacial pain in the northeast of England at 6 monthly intervals: quality of life (EuroQol-5 Dimensions; Herdman et al. 2011), multidimensional pain experience—West Haven–Yale Multidimensional Pain Inventory and Graded Chronic Pain Scale (Kerns et al. 1985; Von Korff et al. 1992), illness perceptions (Illness Perceptions Questionnaire; Moss-Morris et al. 2002), and anxiety/depression (Patient Health Questionnaire–4; Kroenke et al. 2009)—and use of services and productivity (Wordsworth and Thompson 2001). The first article from this study, examining costs and their predictors, was recently published (Durham et al. 2016), and this article refers to, and helps explain, some of those quantitative findings.

Mixed methods research has gained in popularity, recently helping to explain more complex problems in health care (Tariq and Woodman 2013), and in the context of this study, we anticipated that qualitative data would complement and give greater context to the quantitative data collected. This qualitative study is an example of longitudinal qualitative research (Calman et al. 2013), in which qualitative data were gathered at multiple time points, thereby allowing patients time to explain their experiences of care and elaborate on their patterns of service utilization and the impact of the current care pathway. A longitudinal approach to qualitative data collection was particularly appropriate in the context of this study's aim, as it allowed the collection of data over time, an examination of transitions in care, and examination of the stability of concepts or constructs patients explained at different points in time (Calman et al. 2013).

Prior to joining the DEEP study, all patients (n = 198) completed a screening questionnaire to identify the most likely origin of their pain (musculoskeletal, neurovascular/neuropathic, or combined origin) and provided written informed consent. Patients then provided a detailed structured history of their pain: duration, diagnosis received, health care professionals visited, number of appointments, and medications or treatments provided. Following this, for the purposes of the current article, a purposive maximum variation subsample of those participating in the DEEP study was taken according to sex, care environments experienced, time in care, and origin of persistent orofacial pain.

This subsample was recruited in the first 6 mo of patients enrolling in the DEEP study. Two experienced and trained interviewers (M.B. and S.M.B.) conducted these semistructured interviews by telephone (with the exception of 2, where at the patient's request, interviews took place face-to-face on university premises). While telephone interviews can be viewed less favorably to face-to-face interviews, it has also been suggested that the 2 approaches obtain similar results (Sturges and Hanrahan 2004), with some even suggesting that telephone interviews may offer some advantages in causing respondents to relax more and disclose sensitive information (Novick 2008).

The initial topic guide for the interviews was informed by previous research (Durham et al. 2010; Durham et al. 2012), and questions focused on the experience of using health care services since the interviewee's pain began. The topic guide focused on the nature and impact of pain, the use of health services, the information received, and whether care could have been improved. The topic guide was flexible, enabling new topics to be explored as they emerged, and it evolved as themes emerged from interviews. Changes to the topic guide were discussed regularly among the research team, and data collection continued until saturation. The longitudinal design of the qualitative study meant that interviews were repeated at 12 mo with the same topic guide but with additional questions designed to further explore themes arising at baseline from the qualitative and quantitative data.

Data Analysis

This research followed best practice for mixed methods research (Creswell et al. 2011). The analytic approach used iterative thematic analysis (Rapley and Silverman 2011). It follows the Standards for Reporting Qualitative Research (O'Brien et al. 2014). Interviews were digitally recorded and transcribed verbatim. Immersion in the data began with an initial reading of the transcripts while researchers listened to the recording; this served to familiarize them with the data and identify transcription errors or missing data. The 2 interviewers carried out coding of data, and regular discussion of emerging themes took place at project team meetings. Data from baseline and 12 mo were treated identically with respect to coding and identification of themes, but pairs of interviews were also examined together to examine experiences in the period between the 2 interviews. Care pathways for each patient were assembled according to interview data consistent with methodology previously reported by the research team (Durham et al. 2011). NVivo qualitative data analysis software (version 10; QSR International Pty Ltd.) was used to help organize and analyze the data.

Results

Twenty-two patients who had experienced persistent orofacial pain between 3 mo and 27 y were interviewed at baseline. Interviews were repeated 12 mo later with 18 of these participants. Reasons for dropout at 12 mo were nonresponse (n = 3) and health issues unrelated to persistent orofacial pain (n = 1). Participant characteristics are summarized in the Table along with those who dropped out. Presentation of data is structured according to the 3 major themes to emerge from the data: "fluidity of care pathway," "failure to progress," "impact of unmanaged pain." Patients' quotes are used throughout the results to illustrate findings, and the parentheses following the quote contain patients' qualitative study numbers and the time of contact (baseline or 12 mo). Although patients talked extensively about the experience of their pain, this article focuses on the care pathway itself.

Fluidity of Care Pathway

One of the major themes in these interviews related to the movement of patients among health care professionals and settings in an attempt to obtain a diagnosis and effective treatment for their pain.

Patients reported that their first contact with the health service was usually through their general dental practitioner or general medical practitioner (GP). Patients chose to seek help from either a general dental practitioner or GP on the basis of whether they perceived their pain to originate in their teeth/jaw or elsewhere. Formal referral pathways or processes between GPs and general dental practitioners were not evident in the data, but there were sporadic reports of GPs suggesting that patients discuss with their dentists and vice versa. This tended to occur when a health care professional suspected that the cause of pain was outside one's professional remit: "He [the general dental practitioner] went 'No, you've got trigeminal neuralgia you need to go and see your GP'" (Q13, baseline).

In some cases where the diagnosis or origin of pain remained unclear, patients continued to seek help from both dental and medical services: "I've been to see them all [dental and medical professionals]. The GPs don't seem to know what to do. I just don't know where to go next. I go to GPs and they just give me more tablets and that's it" (Q12, 12 mo).

The referral process from primary care to secondary care seemed to vary greatly among patients; although a minority of referrals happened quickly for some, other patients described a long delay or

Table.
Participant Characteristics.

	Patients, *n* (12 mo)[a]
Sex	
Male	5 (4)
Female	17 (14)
Reported diagnosis on entry to study[b]	
Temporomandibular disorder	8 (6)
Trigeminal neuralgia	3 (3)
Persistent idiopathic facial pain	1 (0)
Peripheral neuropathy	1 (1)
Migraine	2 (2)
No diagnosis	8 (7)
Age, y	
≤40	4 (4)
41 to 49	6 (5)
50 to 59	4 (2)
60 to 69	6 (5)
≥70	2 (2)
Time since pain started	
≤12 mo	3 (2)
12 mo to 5 y	9 (9)
5 to 10 y	2 (2)
≥10 y	8 (5)
Health care professionals seen, *n*	
1 to 4	11 (10)
5 to 8	7 (5)
9 to 12	4 (3)
Maximum appointments with a single health care professional, *n*	
1 to 20	15 (13)
21 to 40	2 (2)
41 to 60	4 (2)
61 to 80	1 (1)

[a] Patients following dropout at 12 mo.
[b] Individuals could have >1 diagnosis.

even a reluctance to refer to a specialist in secondary care. Some patients who reported that they had not received effective management in primary care felt that their case was not being prioritized or that their primary care practitioner did not appreciate the severity of their pain: "I think you know migraines clearly isn't [*sic*] life threatening is it so you're kind of back to the back of the queue in a way" (Q5, baseline).

> I mean you go to the doctor [GP], they don't know what pain you are in, they can't feel it, nobody can feel the pain you're in. As far as the doctor was concerned, he was giving me something, it was for fits [epilepsy] or something, Tegretol, I don't know what is for, it's not for neuralgia, but it's for facial pain. . . . He gave me them and he didn't send me to anyone, so that's how bad it was, he didn't send me anywhere [for further treatment] so you can work that one out yourself, you know, whether it's, whether they're bothering or not [to take the problem seriously] . . . the dentist bothered more than the doctor but he [the dentist] couldn't see nothing wrong with me. (Q14, baseline)

Following a number of appointments without effective treatment, many patients felt that the next step should be a referral to a specialist, and they became frustrated when this did not happen. Several reported the need to emphasize the seriousness of their pain or insist on a specific course of action, such as referral to a specialist: "I actually went to my GP. . . . I had the confidence to do that, a lot of people don't. . . . [I] said you know it's just getting . . ., you know I can't live with this at the moment, and could I be referred to a neurologist" (Q5, baseline).

While many people described a situation in primary care in which they struggled to receive a diagnosis, pain management, or a referral, a small group of patients described their experience more positively. This group of patients had several factors in common: they had received effective treatment relatively quickly from their GP, they described a positive relationship with their GP in which they trusted the GP's ability to manage their condition, and they could easily access their GP, "I have faith in them [the GPs] and, and they're good in that they listen and they act on what you say in that they've never kind of gone oh you're talking rubbish about anything" (Q13, baseline).

Patients who were referred to secondary care reported being sent to a range of departments in medical and dental hospitals—for example, neurology, pain clinics, oral surgery, ear nose and throat surgery, physiotherapy, ophthalmology, maxillofacial surgery, and hematology. For some, secondary care was the first time that they had received a diagnosis for their pain; others received confirmation of a suspected primary care diagnosis; and some reported contradictory diagnoses from secondary care health care professionals. Those who received treatment in secondary care reported a range of treatments: new or revised pain medication prescriptions, dental splints, exercises, and, in some cases, surgery. A number received further referrals to other specialists within secondary care, and others were prescribed pain medication that was subsequently managed longer-term within general practice.

Some patients received treatment in secondary care, which had a positive effect on the level of pain: "He [the consultant] made me a mould thing [splint] that I wear at night, and that's lessened the pain, it hasn't gone away but I can cope with it" (Q12, baseline). A small number of patients who continued to experience pain reported being referred to pain management services, whereas others found themselves back in primary care having attended multiple appointments without a diagnosis or effective pain management. Although criteria for pain management services were unclear, 1 patient reported being referred from secondary care and being refused (ie, turned down for funding for a specific item of care): "I would have loved to have gone to the pain management in [location's name] that [the dental consultant's name] suggested but the [name of health care commissioning body who pays for the treatment] turned me down for that" (Q12, 12 mo).

Following secondary care consultation, some described returning to primary care for a continuation of treatment suggested in secondary care (e.g., repeat prescriptions). Others explained that they were redirected within secondary care after their initial consultation—that is, to another, different speciality within secondary care: "The hospital [then] actually wrote to my GP and I think it was them [the hospital] that gave them [the GP] the push and they [the GP] actually sent me for a . . . referred me to a neurosurgeon" (Q15, baseline).

The extent to which patients' pain was managed varied and did not appear to be related to the length of time since it began. A minority of patients were pain free following treatment, and some reported a reduction in pain while others remained with severe or worsening pain. Over time, patients reported different behaviors with regard to accessing health care. For a minority of patients who remained with little pain, they reported no need to seek care again unless their level of pain increased or their medication became less effective:

> I am very close to actually going back now [to name of primary care practitioner], because it's [the pain] starting [to increase again]. . . . I am going to [through] a few bits at the moment with stress and it's [the pain] coming back how it was, and it's really bad. I can feel it coming back. It [the jaw] clicks out of joint and then it hurts, but it's [the pain] nowhere near as bad as it has been, but it's only getting worse. (Q18, 12 mo)

A group of patients with unmanaged, sometimes severe, pain reported continuing to make appointments seeking help for their pain. Some patients had specific objectives for these appointments: some attended to see if there were any new treatments on the market, while others wanted a specific course of treatment or referral: "Now [my] last GP visit that I went to, oh, some months ago, I can't remember when, so obviously I persuaded them to let us take some Voltarol [strong anti-inflammatory analgesia; generic name, diclofenac]" (Q20, baseline).

Following inconclusive investigations, some described feeling that they had reached the end of their medical care: "And then it [the pain] was sort of forgotten about because I'd had all that [polypectomy, GP appointment, referral, secondary care appointment, prescription of pain medication, magnetic resonance imaging] done and everybody just sort of let me drift back into the ether" (Q5, baseline).

Failure to Progress

Whereas patients reported multiple appointments, often with different health care professionals, a "failure to progress" was a strong theme within the data and related to achieving a diagnosis or management of their persistent orofacial pain.

Few respondents reported being given a diagnosis and/or any successful management at their initial appointments in their care pathway. Several respondents described misdiagnosis in primary care, resulting in unnecessary treatment and further delays in pain management: "I was told it [the pain] was [due to] sinus trouble when I first went so I got like a nasal spray and it actually made it [the pain] worse" (Q15, baseline).

Initial attempts at management of pain in primary care were reportedly often made without a firm diagnosis. Respondents reported a variety of initial management attempts, including 1) prescriptions of antibiotics or pain medications from GPs and 2) fitting of dental splints, replacement of fillings, extractions, and the prescription of jaw exercises from general dental practitioners. Sometimes patients reported several consecutive treatments in an attempt to find one that was effective with acceptable levels of side effects. This process could last for an extended period without resulting in any symptomatic relief:

> When I go [to the GP] it's just kind of a cycle, try this new medication and come back 6 weeks say when you can tell me if it's [the pain] improved and if it has improved we'll keep you on it for a bit longer, if it hasn't we'll try something else, that's what's been like for the past 2 or 3 years. (Q7, baseline)

A number of patients attributed the fact that they had not received effective management to professionals failing to take seriously or prioritize their pain:

> I think for me the frustration initially was a lot of appointments [with varying primary care practitioners] but I wasn't really getting anything from them because nobody was really—well I didn't feel like they [the health care professionals consulted in primary care] were taking the whole situation very seriously. (Q22, baseline)

When pain remained unmanaged, this sometimes led to patients questioning the ability of their primary care practitioner to provide management of their complaint: “I just feel sometime well the GP is really nice, but how many times did she come across this [the pain problem and its diagnosis]?” (Q2, baseline). For others, referral to secondary care functioned as a setting in which to conduct diagnostic tests, such as magnetic resonance images or computed tomography scans, but following negative results, patients were discharged. On several occasions, tests ruled out specific conditions but still left patients without a diagnosis. Some patients described communication of these results as further evidence of a failure to acknowledge their condition on the part of health care professionals and a failure to progress:

> He [GP, following referral for computed tomography scan] thought: “Well, there’s nothing wrong”; nothing—obviously you haven’t got a tumor; you haven’t got this or that; but I don’t think he understood that even though that showed that there wasn’t anything major in those respects; that there was still—I still had the same problem [persistent facial pain]. (Q19, baseline)

Negative diagnostic tests were not always a reassurance, and sometimes, rather than being relieved patients described disappointment accompanying uncertainty over the cause of their pain and a possible treatment: “I think I’d rather just see the worst, to be honest. Even if it had said it was cancerous or something, at least I would have known, rather than not known” (Q7, 12 mo). Another group, however, despite unmanaged pain, felt that it had little to gain from accessing further health care and described disengaging with the health service. Some attributed this to a failure on the part of the health care professionals and the existing care pathway:

> I think they [several general dental practitioners consulted previously] didn’t really believe . . . so partly that, partly ’cos it [different treatments attempted] sometimes made it worse and also ’cos you know it just wasn’t getting anywhere with it. I sort of still had the pain but just stopped going. (Q10, baseline)

Although a number of patients who stopped attending appointments felt that health care professionals were responsible for their lack of pain management, others believed that the main problem was the lack of effective treatments available:

> I don’t really talk about my pain at the doctors much unless I am asked about it because there is not a lot of point because what can they say to you, what can they say? They can’t say, “We got a cure next week, get in a queue and you are at the hospital next week.” (Q11, 12 mo)

While many people described frustration with health care professionals and the lack of progress throughout their care pathway, those who remained with unmanaged pain emphasized the importance of health care professionals being empathetic, even in the absence of being able to offer any effective treatment:

> That made such a difference, to think that somebody actually cared about you, as a person, and not just wanting to get you out of the surgery as quickly as possible. Even though she can’t do anything practical, to change the situation, it just made a difference to see that she really understood how bad the problem was, and was concerned. It does make a big difference as to how you feel. (Q19, 12 mo)

Although, in some cases, patients described a diagnosis leading to successful treatment, diagnosis did not always seem to equate to management of the pain, and some without a diagnosis described their pain as well managed. As time progressed, the relevance of a diagnosis appeared to change from that of the functional purpose (i.e., to have a label for their condition) to one of legitimization as they became aware of others, even themselves, questioning the physical basis of their pain: “I’ve got to admit, my wife suggested this to us [me] as well, and I have thought about it, could it be psychological?” (Q20, baseline).

Impact of Unmanaged Pain

Patients spoke extensively of the impact that unmanaged pain had on multiple areas of their lives. In addition to medical and dental treatment, ongoing pain management comprised an element of self-management for many patients. For some, this consisted of trying to identify and avoid triggers and make lifestyle changes, while others used home remedies, complementary therapies, or private medical care. Some felt that these strategies played an important role in helping them to manage their persistent orofacial pain, and over time they reported getting better at strategies that they felt led to a decrease in their pain: “I think I’m aware of that [trigger for pain], and so if I am very tired or stressed I’ll think, ‘No, I’ll not have any alcohol’ or something, just because I know that that might have an adverse effect” (Q17, 12 mo).

The impact of remaining with unmanaged pain varied among individuals and appeared to be influenced by their personal circumstances and the nature and severity of their pain. The sensation of pain and its accompanying symptoms were discussed in detail: “The pain is very very uncomfortable but it’s the feeling sick, the sort of not being able to coordinate your speech, not being able to do anything, not being able to smell anything you know the whole thing that surrounds the migraine” (Q1, 12 mo).

The impact of pain was not limited to the unpleasant sensation, and patients described the impact on multiple areas of their lives, including everyday tasks, psychological well-being, family life, social activities, and work and personal finances. Patients described difficulties working while experiencing pain: “I was tired because I wasn’t sleeping terribly well I do think it affects my performance at work” (Q16, baseline).

Fewer individuals reported experiencing pain that would leave them unable to work, and for others the severity and continuation of their pain and medication side effects were disabling:

> I don't do anything. I just haven't got the energy. The pain takes control over your life. You feel so worn out with it that all you want to do is just sleep if you can. Sometimes when I take all of the pills, I don't know what date it is. I forget things. I'm just not in control of my own life. If you take all of the pills, you're absolutely a walking zombie and it doesn't seem to make any difference. (Q12, 12 mo)

Although several patients had undergone diagnostic tests, which had ruled out specific conditions, the precise cause of their pain remained something that caused concern:

> Why a diagnosis would help me is because my mind, since 1987, has been, shall we say, in a bit of turmoil. I think, "What is happening inside my head? Have I got a tumor?" etc., etc. Do you know what I mean? I suppose this is the way your mind works, everybody's does work, but I've had to overcome this by myself, and with help from my wife as well. (Q20, 12 mo)

Even patients who had eventually made progress toward management of their pain could have continued negative consequences on their lives: "I just wish that all these things like the pain therapy that it was all offered sooner I would maybe not have lost my job if they could have got the pain under control. I could have kept my job" (Q3, baseline).

Summary

Patients generally recalled and described fluid and complex care pathways in which they visited medical and dental primary care practitioners and medical and dental specialists in a range of secondary care settings: neurology, pain clinics, oral surgery, ear nose and throat surgery, physiotherapy, ophthalmology, maxillofacial surgery, and hematology. Although patients were recruited to this study independently from primary and secondary care, the complex nature of their pathways meant that this position was often fluid with continual movement among care sectors. Several care pathways were evident in the data, but a consistently recurrent theme was a failure to progress, which spanned diagnosis, referral, and treatment in both primary and secondary care.

Discussion

These data highlight problems with the care pathway from the perspective of a group of patients with persistent orofacial pain in the United Kingdom. The major problem appeared to be the length of time that it took for patients to progress from presenting with pain to receiving a diagnosis and effective treatment, with some of this sample having received neither after an extended period.

Many patients described persistence in attending appointments in primary and secondary care, in an attempt to access effective treatment. From a patient's perspective, relationships with health care professionals were at times problematic, with continued failure to manage pain being attributed to health care professionals' lack of interest, empathy, or knowledge. Previous literature has highlighted similarly problematic relationships from a health care professional's perspective with patients with long-term, sometimes medically unexplained, conditions who attend frequently and remain dissatisfied, being labeled "heartsink" patients (O'Dowd 1988; Bligh 1999). In this sense, persistent orofacial pain may share some aspects with other painful and/or difficult-to-diagnosis conditions, such as fibromyalgia (Arnold et al. 2008) and chronic fatigue syndrome. Where persistent orofacial pain differs from many other chronic illnesses is that the location of the pain means that persistent orofacial pain's care pathways often involve appointments with medical and dental health care professionals.

The repeated appointments described in these qualitative data are reflected in the quantitative data from the same study (Durham et al. 2016), which highlight a large number—and, consequently, high cost—of consultations. These interview data provide some explanation for these appointments but also suggest that, despite a high number of appointments, patients' pain remains unmanaged for extended periods. Furthermore, the longer the pain remains unmanaged, the greater the potential for significant impacts on patients' lives. This is mirrored in the quantitative data from the DEEP study, with significant impacts on patients' quality of life at a level similar to those caused by other chronic illnesses, such as arthritis (Durham et al. 2015).

The referral process from primary care seemed to vary greatly among patients, with some being referred quickly and others waiting for a long period. Despite a previously reported tendency for general dental practitioners to refer to secondary care (Durham et al. 2007; Aggarwal, Joughin, Zakrzewska, Appelbe, and Tickle 2011; Peters et al. 2015), there is evidence that persistent orofacial pain does not always require specialist diagnosis. Aggarwal, Joughin, Zakrzewska, Crawford, and Tickle (2011) suggested that despite the knowledge gaps, general dental practitioners may have sufficient capability to provide a diagnosis, and Dworkin et al. (2001) concluded that nonspecialist management can be effective. Our data also suggest that primary care may be an appropriate place to manage some patients, either in isolation or in partnership with specialist secondary care, dependent on the presenting complaint and comorbidity. In other persistent pain conditions, there is suggestion that stratified care pathways may be more appropriate in terms of health outcomes and cost savings (Hill et al. 2011), and there may be lessons to be learned from other chronic conditions in the management of persistent orofacial pain.

The majority of those interviewed had taken a long time to receive a diagnosis, and some were still unable to recall being given a diagnosis despite having experienced pain for a number of years. Although patients initially described the need for a diagnosis to direct health care professionals to appropriate treatment, this may have taken on greater importance as their pain remained undiagnosed despite diagnostic tests and multiple appointments. For patients where a negative diagnostic test was communicated to them as "good news," there was a clear discord between how a negative test was perceived by the patient and the health care professional. For some patients, a negative test result appeared to mark an end point in health care professionals' efforts to diagnose and treat their pain; therefore, it seemed to those individuals that a diagnosis was necessary to qualify for further medical care.

Previous chronic pain research has highlighted the importance of diagnosis and treatment for patients to gain legitimacy; the labeling and treatment of their condition are proof of their illness (Glenton 2003). The presence of uncertainty from health care professionals, coupled with a failure of tests to provide a diagnosis, may serve to "disconfirm" patients' pain as well as prompting others, such as family, work colleagues, and employers, to question the legitimacy of an "invisible" complaint (Rhodes et al. 1999). Based on a study of people suffering from chronic back pain, it has been suggested that the inability of health care professionals to provide patients with a diagnosis, information, or treatment acts to prolong the dependency of patients on health care professionals (Glenton 2003). This may be particularly relevant in primary care, where appointments are initiated by the patient rather than accessed via referral, as in secondary care consultations. Additionally, Durham et al. (2010) emphasized the negative effects of the uncertainty that living with undiagnosed pain can have. In orofacial pain, Aggarwal et al. (2008) emphasized the importance of an early diagnosis in ensuring contact with the correct health care professionals to contain health care resources. Our data not only suggest that patients struggle to achieve an early diagnosis in the existing care pathway but also highlight a need for continued management of their pain, with or without a diagnostic label.

Reflections on Methodology

This research adds to a limited amount of qualitative research on care pathways in persistent orofacial pain and helps to contextualize previous and current cost data within the patient experience. Our sample comprised a heterogeneous group of patients with regard to sex, care environments experienced, time in care, origin of persistent orofacial pain, and diagnosis. In the wider context of the DEEP study, these data act in an explanatory role with regard to costs of care and outcomes. This study is based on a purposive sample within a specific geographic area; as such, caution needs to be exercised in generalizing our findings to other localities or other health care systems. The data do, however, triangulate with other data in the literature, pointing to similar difficulties for patients with persistent orofacial pain. The constructs that patients raised also seem to be stable over the time frame of the longitudinal interviews, and there were limited changes, as depicted in the data, in their perceptions of the care pathway over time.

As with other qualitative studies, we cannot assume that this sample is representative of the wider persistent orofacial pain population; however, through the use of careful purposive sampling, we sought to capture and illustrate a depth and breadth of experiences and perspectives. In addition, as data collection and analysis progressed, we deliberately sought out the views of those who may have had different experiences by virtue of their diagnoses, sex, age, or locations. Nearly half of our sample had pain that began >5 y prior to the study, which, though providing an important perspective on care pathways, may have led to recall bias. In conjunction with quantitative data from the DEEP study, these qualitative data help to highlight areas of the care pathway, which could be improved.

Conclusions

Current care pathways do not appear to be meeting patient needs despite indications of substantial health care resource use. Patients describe being without a diagnosis and effective pain management for extended periods. Changes to care pathways need to ensure that patients receive a timely diagnosis, treatment, and referral, if necessary, and one way of achieving this may be through a restructuring of care provision for persistent orofacial pain in primary and secondary care. A care pathway that contains a reduction in the number of consultations may have the potential to reduce the biopsychosocial effects of pain, in addition to limiting the number of ineffective health care appointments. There is also a need for a greater understanding of the role of primary and secondary medical and dental care in both short- and longer-term management of persistent orofacial pain. Future research should involve working with stakeholder groups to explore the design and feasibility of new care pathways.

Author Contributions

M. Breckons contributed to data collection and analysis and drafted and critically revised the manuscript. S.M. Bissett contributed to data collection and analysis and critically revised the manuscript. C. Exley contributed to study conception, design, interpretation of data and critically revised the manuscript. V. Araujo-Soares contributed to study conception, design, interpretation of data and critically revised the manuscript. J. Durham secured funding, led study conception and design, contributed to interpretation of data and drafted and critically revised the manuscript. All authors gave final approval and agree to be accountable for all aspects of the work.

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