Abstract: This article reports on three patients who presented with pain following placement of dental implants in the anterior maxilla. It aims to document a rare complication of implant placement which arguably requires further investigation through characterization of cases. Three cases in total were identified and the literature review identified only one previous case series. All three cases were characterized by idiopathic pain which presented, in some cases, in a delayed fashion following fixture placement. All cases failed to resolve following treatment. Persistent chronic idiopathic pain is a recently described risk of dental implant placement. This case series highlights the need for careful case selection and informed consent, particularly when considering changing paradigms relating to consent procedures.

CPD/Clinical Relevance: This case series aims to raise awareness for this rare complication of implant placement in the anterior maxilla. It highlights the importance of thorough pre-surgical investigation, planning and consent.

Dental implants have progressively become more widely utilized in the rehabilitation of edentulous spans. Some studies suggest that 100,000–300,000 implants are placed per year. This progressive increase in use has been mirrored by an increasing awareness of the advantages, disadvantages and processes involved in placing and managing dental implants. An example of this includes the appropriate utilization of more advanced radiographic techniques to identify and avoid vital structures when planning and undertaking surgery. When correctly planned, placed and utilized, dental implants have the potential to improve oral health-related quality of life (OHR-QoL) significantly.

Common risks or complications of dental implant treatment include infection, failure of osseointegration, short-term post-surgical pain and the risks associated with trauma to vital structures (ie post-operative bleed or symptoms associated with nerve injury). Specific risks associated with placement in the mandible (risk to the inferior dental and lingual nerves) and posterior maxilla (risk to the maxillary sinus) are well covered in the literature. Potential risks associated with implant placement in the anterior maxilla are, however, less well investigated. There is a dearth of literature reporting on chronic idiopathic pain following implant placement. This condition is characterized by patient-reported experience of long-term persistent and chronic pain within the region of implant placement which begins after implant placement and for which no anatomic pathology can be found. It can involve a small or large region and responds unpredictably to implant removal and to pharmacological treatments. It may present with a wide gamut of pain characters.

Chronic idiopathic post-surgical neuropathic pain is associated with numerous medical procedures, including limb amputation. It is considered a rare complication of some dental procedures, including dental extractions and endodontic procedures (both orthograde and retrograde). Chronic idiopathic pain (CIP) after dental implant placement is poorly understood and uncommonly reported. It is poorly reported in the literature with only one other published case series. Furthermore, apparent pain without an identifiable cause can be difficult to predict and challenging to treat.
Despite being rare, the potential impact of chronic idiopathic pain on the OHR-QoL is significant. Further awareness and investigation of this is therefore pertinent to promote patient management.

This retrospective case series reports on three cases seen at the Leeds Dental Institute Restorative Dentistry and Oral Medicine clinics between 2009 and 2016. These cases highlight the variable history of presentation along with the possible late presentation of idiopathic facial pain following implant placement. Furthermore, these have included the restorative and prosthodontic management of CIP as well as management from an Oral Medicine perspective. The authors aim to document a rare complication of implant placement to raise awareness and support the evidence base through characterization of cases.

Materials and methods

Referrals relating to pain associated with dental implants presenting to the Leeds Dental Institute via the Restorative and Oral Medicine departments were identified. All cases with no identifiable cause have been recorded and followed up. Cases were collated by discussion with all individuals involved in the management of dental implants, along with discussion with all individuals involved in the management of idiopathic facial pain. This report has been constructed using Equator Network Care Guidelines for the reporting of case reports/case series.

All patients presented with CIP following implant placement have undergone thorough history, examination and investigation processes by both departments. This has included a full pain, medical, social and dental history, along with details of implant placement (including operator and surgical factors). Some patients may present with either pre-existing or associated psychological symptoms and appropriate referrals were made to other specialties, including psychiatry, if indicated. Furthermore, all patients underwent a thorough clinical examination, including assessment of cranial nerves, temporomandibular joints, muscles of mastication, lymph nodes and facial symmetry. Intra-oral hard and soft tissues were assessed in detail, including in-depth assessment of dental implants. In some cases, if appropriate, coronal restorations were removed to aid assessment. Furthermore, clinical examination was supported by radiographic assessment of plain film images and either computed tomography (CT) or cone-beam computed tomography (CBCT), based on clinical justification and the available technologies. Implants were evaluated by assessment of position and orientation of placement, bone levels and bone loss over time, peri-implant soft tissues, probing depths, and quality, contour, orientation, occlusion and aesthetics of coronal restorations.

Chronic idiopathic pain associated with dental implants in the maxilla was defined as pain presenting following implant placement (either immediate or delayed following surgery; cases with preceding pain were excluded) within the region of implant placement and for which no cause could be identified following thorough clinical and radiographic examination. Patients were selected based on placement of implants in the maxilla, however, all cases identified involved pain following placement in the anterior maxilla. Possible sources of pain include damage to the neurovascular bundle or accessory nerves, peri-implantitis or peri-implant mucositis, traumatic occlusion, pathology associated with the adjacent tissues (eg fractured alveolus). All of these were excluded in the described cases.

Relevant data were collected in a Microsoft® Excel 2016 Spreadsheet.

Results

Three cases in total were identified and are described below. A summary of the cases is presented in Table 1.

Case 1

A 45-year-old female presented following implant placement at UR1 in the practice setting. Previous dental history included UR1 having been traumatized 18-months previously, causing the tooth to fracture subgingivally. UR1 had been crown lengthened and was restored with a post-crown. This tooth was subsequently extracted after persistent pain. Following extraction, the pain resolved, and an implant was placed 8 weeks post-extraction and subsequently restored with a screw-retained single crown after a 2-stage protocol.

The patient began experiencing dysesthesia around the implant site and a dull pain from her buccal mucosa bilaterally. This was progressive in onset with no clinical changes being evident throughout the process.

Examination including periapical radiographs and CT imaging identified no pathology. These specifically assessed for the spatial orientation of the implant fixture to the nasopalatine canal or any relevant anatomy which may have indicated possible
involvement of the nasopalatine nerve; no such involvement was identified (Figure 1a).

Numerous treatment protocols involving paracetamol, NSAIDs, opioids, hypnotherapy and acupuncture failed to relieve symptoms. A stepwise process was used. The patient had self-prescribed paracetamol and NSAIDs. Opioids were utilized as the next step as per available guidance. The patient was keen to avoid further medication and, therefore, adjunctive therapies were utilized; these were not successful. The patient subsequently declined further treatment.

**Case 2**

A 31-year-old male presented following a road traffic accident with pain from the UL1 tooth. UR1 had already been extracted in primary care following the trauma. The patient had suffered from headaches following trauma which resolved following removal of glass fragments from soft tissue (forehead).

The patient complained of a persistent soreness from the gingivae and pain from UR1. This was relieved by applying pressure to the tooth (including biting). Examination identified tenderness labially from UL1, UL2 and UL3 sites with UL1 being tender to percussion. The UR1 edentate ridge was also tender to palpation. Assessment by both restorative and oral medicine departments identified UL1 as the source of pain as a result of an apical root fracture and acute apical periodontitis.

UL1 was extracted without complications and UR1/UL1 sites were restored with an acrylic partial denture. The patient’s symptoms subsided.

Implant work-up included a CT scan with a radiographic stent. A 13 mm OsseoSpeed Astra Tech implant was placed using a two-stage approach (second stage 3 months after fixture placement). A definitive screw-retained restoration was placed 4 months later.

The patient began experiencing pain 4 months following definitive restoration (as described in Table 1). This was progressive in nature but changed little throughout the course of further reviews and treatment. In this case, the patient’s symptoms presented spontaneously throughout the course of a day with no diurnal variation. Furthermore, there were two aspects to the patient’s symptoms: pain from UR1 palatal mucosa; and tingling to the left cheek and forehead. It was not possible to elucidate whether all the patient’s symptoms relate to implant-based treatment or prior injuries. The delay between implant treatment and presentation of symptoms makes the process of ascertaining a definitive cause more difficult, particularly given the lack of clinical and radiographic findings (Figure 1b).

Ongoing reviews identified no pathology. Paracetamol, NSAIDs and replacement restorations failed to resolve symptoms, however, a combination of treatment strategies,

<table>
<thead>
<tr>
<th>Case</th>
<th>Age</th>
<th>Gender</th>
<th>Implant Location</th>
<th>Site of Pain, Character and Radiation</th>
<th>Onset</th>
<th>Exacerbating &amp; Relieving Factors</th>
<th>Severity (VAS max 10)</th>
<th>Medical History</th>
<th>Management</th>
<th>Pain Relief Achieved</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>45 years</td>
<td>Female</td>
<td>UR1</td>
<td>Sharp stinging UR1 palatal mucosa, dull pain zygomas and mandible</td>
<td>18 months following definitive restoration of UR1 implant</td>
<td>Cold foods and resting on right side of face</td>
<td>6/10</td>
<td>Anxiety disorder managed with beta blocker No known allergies</td>
<td>Paracetamol/NSAID Opioids Hypnotherapy Acupuncture</td>
<td>No</td>
</tr>
<tr>
<td>2</td>
<td>31 years</td>
<td>Male</td>
<td>UR1, UL2</td>
<td>Spontaneous dull ache UR1 palatal mucosa, tingling left cheek and forehead</td>
<td>Gradual increase months following implant placement</td>
<td>Biting</td>
<td>10/10</td>
<td>Coeliac disease No known allergies</td>
<td>Paracetamol Hypnotherapy Acupuncture</td>
<td>Partially</td>
</tr>
<tr>
<td>3</td>
<td>27 years</td>
<td>Female</td>
<td>UR1, UL1</td>
<td>Persistent, spontaneous, migraine-like pain UL1, spreading to left eye</td>
<td>Sore following definitive restoration, progressed to severe pain 1 year later</td>
<td>Biting</td>
<td>Variable: 4/10–10/10</td>
<td>Fit and well History of trauma to face No known allergies</td>
<td>Paracetamol/NSAID Replacement crown</td>
<td>No</td>
</tr>
</tbody>
</table>

Table 1. A summary of the cases presented with chronic idiopathic facial pain following placement of dental implants in the anterior maxilla.
including hypnotherapy and acupuncture, were used. The patient experienced partial relief of symptoms using this treatment strategy.

**Case 3**

A 27-year-old female presented 10 years following implant placement at the central incisor sites with a complaint that teeth were ‘too high’. This was resulting in an unsatisfactory appearance and speech. The patient had an anterior open bite thought to be attributed to continued facial growth after fixture placement and proclined implant fixtures. Periodontal and peri-implant health was good. Adequate aesthetics could not be achieved given the fixture position and orientation, and therefore consent was obtained for implant fixture removal and replacement.

Implant work-up was completed including a CT scan with a radiographic stent. 13 mm and 15 mm OsseoSpeed Astra Tech implants were placed using a 2-stage approach (second stage 5 months after fixture placement). The fixtures were restored with provisional crowns 1 month following second stage surgery. Definitive screw-retained restorations were placed 4 months later.

The patient returned 18 months following definitive restoration complaining of a dull ache from the palatal mucosa in the UR1 region over the preceding 14 months (summarized in Table 1). Clinical examination and periapical radiographs identified no pathology (Figure 1c). Replacement restorations and analgesics failed to resolve the pain. A review by Oral Medicine identified tenderness of masseter muscles bilaterally at their superior attachment and of the left sternocleidomastoid muscle in the mastoid region. The right temporomandibular joint was tender on closing and on lateral movements.

The patient was offered opioid analgesics and tricyclic antidepressants but declined all further pharmacological treatments.

**Discussion**

This article reports on three cases of persistent idiopathic pain following implant placement. The number of cases remains small, however, all presented with persistent chronic idiopathic pain within the region of implant placement. No cases had been diagnosed with pre-existing pain disorders. All identified cases involved placement in the anterior maxilla. No cause was identified for any case and all patients continue to experience symptoms.

The presentation of pain is important to note. All cases presented with pain in a delayed fashion following implant placement and restoration. Classically, pain resulting from neurological trauma presents soon after the injury, or during healing. All cases presented with pain which appeared to, at least partially, mimic neuropathic pain. However, the described symptoms were vague in some cases. The findings presented here are suggestive of this presentation being distinct from pain which is conventionally attributed to neurological or soft tissue trauma.

The management of all cases has involved the use of pharmacological treatment by the Oral Medicine department. Case 2 involved removal and replacement of the overlying prosthesis. Neither of these interventions relieved symptoms.

Despite all these similarities, there are many differing factors: the character of presenting symptoms varied quite significantly. Nonetheless, the number of cases reported here is small and there is a need for awareness of this possible risk to be raised and for any further cases to be reported and assessed across dental units.

One of the cases reported included a history of pre-existing anxiety disorder; whilst it has been proposed that both psychological and anxiety disorders are associated with a higher prevalence of chronic pain, the mechanisms for this are unknown. Pain is reported to manifest through the interaction of numerous biological, psychological and psychosocial processes, resulting in a broad range of pain presentations that we can observe and describe. It has been suggested that a history of some diagnoses, such as an anxiety disorder, may increase the risk for developing a condition such as chronic idiopathic facial pain following a surgical intervention. Furthermore, such presentations may, in some cases, be the result of interactions relating to theories of central sensitization. In such cases, multiple pain disorders may present without any apparent anatomic pathology and with poorer responses to treatment. Whilst the relationship between dental trauma and chronic pain is less clear when considering the presentations described, all the patients identified in this series suffered tooth loss related to dental trauma at the site of surgery, and this may be important to consider. Both factors are essential in the initial assessment of patients presenting to the dental practitioner and are important to consider when planning any rehabilitation.

There was no identifiable cause for pain in any of the cases described. It has been suggested that neuropathic pain can present without identifiable injury to nerves. Such cases are important to identify as the literature suggests that fixture removal is unlikely to improve symptoms in such cases. This may result in further surgical trauma and a poorer prosthetic outcome, consequentially reducing quality of life.

Various sources report on the use of pharmacological and behaviour treatment modalities to alleviate symptoms. However, these may not always result in an outcome that is acceptable to the patient. Some studies suggest that fewer than half of patients may experience adequate or significant pain relief. It is this combination of difficulty in predicting cases where persistent chronic idiopathic pain following placement of dental implants might occur, and the difficulty in subsequently managing pain and quality of life, that makes this risk particularly important to investigate further.

In cases where persistent pain does occur, it is important to place patient quality of life at the centre of any treatment planning decisions, with careful consideration of the potential effectiveness, prognosis and impact of any viable interventions. One may suggest conforming to the World Health Organization Analgesic Ladder in the first instance. The WHO recommends a thorough pain assessment and a simple approach to providing pain...
relief. This should adopt a stepwise approach, recommending or prescribing the simplest regimen that relieves the patient’s symptoms. It is also important to highlight the intended purpose of self-prescribed analgesics. Some analgesics (NSAIDs in particular) should not be consumed for the management of chronic pain without referral to a medical practitioner (the patient’s GP in the first instance). Consumption of over-the-counter medications such as Ibuprofen, even for relatively short time periods, can result in significant side-effects, including an increased risk for gastrointestinal pathologies such as peptic ulcers.11

Patients suffering from the symptoms described here are, however, likely to need management in a specialist setting. In such settings, patients may be prescribed non-opioid analgesics, including tricyclic antidepressants (such as nortriptyline) or anticonvulsants (such as gabapentin).12 At this point in time there is a dearth of evidence on the most appropriate analgesic to provide for patients with this presentation and, as such, an appropriate regimen is provided based on the practitioner’s analysis of the patient’s symptoms and medical history, and may involve changes to regimen over a period of time. This is evident in the cases presented here where, in some cases, multiple treatment modalities were successively trialled. Nonetheless, it is recommended that this process is undertaken by a specialist working in a secondary care environment. Initial pain control should involve the utilization of the aforementioned analgesia ladder.7

Finally, one might argue that this is an important risk factor to consider in treatment planning discussions prior to implant placement, particularly in light of changes in UK case law9 and the consent process. This is something that must be considered through a patient-centred approach to ensure that informed consent is gained appropriately from patients prior to proceeding with treatment.

Conclusion

Persistent chronic idiopathic pain is a recently described risk of dental implant placement. Awareness and understanding of this potential risk factor is limited and therefore dissemination of identified cases, along with investigation of the aetiology, risk factors and treatment modalities for this are highlighted. Such reports highlight the need for careful case selection and informed consent, particularly considering changing paradigms relating to consent procedures. In the eventuality that a case with persistent pain following implant placement presents to practice, a referral to the secondary care environment is recommended.

Conflicts of interest

None.

References