Chronic idiopathic orofacial pain. A long-term follow-up study

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Patients suffering facial pain that does not fit with the traditional diagnostic criteria and which does not respond to dental treatment constitute a clinical problem. These patients lack a proper diagnosis and are frequently exposed to excessive and inadequate invasive treatment. The aim of this investigation was to study the long-term development of pain and the result of treatment in a cohort of patients suffering chronic idiopathic facial pain. The 74 patients referred to the Facial Pain Diagnostic Group at the Karolinska Institute School of Dentistry between 1981 and 1992 were invited to take part in a follow-up study. As 16 subjects were unwilling or unable to take part in the study and 13 had died, the remaining 45 were interviewed either in accordance with a standard protocol or by filling out a questionnaire mailed to them. The interview revealed that 10 were free of orofacial pain, but only 2 were totally free of pain. Over the 9–19 years' follow-up period the relationship between facial pain and pain in the rest of the body had changed substantially. Of 14 patients and more than 100 extractions, permanent pain relief was felt by only 3 patients. It is concluded that a diagnosis was given in only 2 cases. The distribution of the pain has changed dramatically. The low success rate of invasive treatments suggests that such therapeutic methods are to be considered contraindicated in patients suffering from idiopathic orofacial pain. □ *Facial pain; intractable pain; long-lasting orofacial pain; tooth extraction*

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Orofacial pain without any pathophysiological explanation has been described in a series of papers (1–8). Typically, the symptoms and clinical findings are not consistent with the present dental classification systems, and standard treatment has no permanent effect on the pain (9–11).

A group of patients suffering from orofacial pain without typical odontological pathology were investigated at the Karolinska Institute School of Dentistry, Huddinge, Sweden in the period 1981 to 1992. The clinical findings collected during this period have been presented in a series of papers (12–14). The patients were called to a further investigation 9–19 years later.

The aim of this investigation was to study the long-term development of pain and the result of treatment in this cohort of patients. The following questions are addressed: 1) After 9–19 years, does the pain have any etiological explanation? 2) In what way has the relationship between facial pain and pain in the rest of the body changed? 3) How have the patients responded to treatment during the observation period?

Materials and methods

Patients with facial pain, but without recognizable odontological pathology, seen at the Karolinska Institute School of Dentistry, Huddinge, Sweden between 1981 and 1992 were referred to the Facial Pain Diagnostic Group (FPDG). Seventy-four patients were received and investigated in this period ('first admittance'). As duration of pain was more than 6 months in all cases, and no causes of the pain were found, the pain was labeled as chronic and idiopathic. The data obtained during these investigations have been reported earlier (12–14).

Subjects

The subjects investigated in this research project are unique in that they constitute a strictly selected group of chronic pain patients: the patients of the original cohort were first treated in primary dental care, where no obvious cause of the pain could be detected. After referral to the Karolinska Institute School of Dentistry, some were given an odontological diagnosis and were treated successfully. A small fraction of the original cohort were then referred to the FPDG (the most senior experts on pain and related topics at the Karolinska Institute School of Dentistry). This fraction of the original cohort constituted the group investigated at the first admittance (12–14) and hereafter referred to as the 'cohort'. Nine to 19 years (mean 14 years) later, 45 members (42 F and 3 M) of this group participated in the present research project. Out of the original 74 patients addressed, 13 had died and 16 were either unwilling or unable to participate in the study (one of these was visited in hospital but was not conscious, 3 did not return the questionnaires despite repeated reminders, 4 were impossible to reach, and 8 refused to cooperate).

The mean age in the group of participants (67.5 years,

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range 46–89) was similar to that in the dropout group (65.5 years, range 37–92). However, the age of the patients in the deceased group was higher (81.4 years, range 56–99). The sex distribution in the group of participants was 6.7% men and the rest women. When those unwilling or unable to participate and the deceased patients were included, the proportion of men was 12.1%.

Procedure

The patients were informed by telephone about the research program and were invited to attend an interview at the Karolinska Institute School of Dentistry, Huddinge Campus. Those who were willing to participate but unable to move to Huddinge Campus were visited in their homes ($n = 12$; 27%) or received a letter ($n = 8$; 18%) consisting of written information and a questionnaire. The remaining ($n = 25$; 56%) patients were received at the School of Dentistry for interview. All the interviews were carried out according to the same interview form as that sent by mail and carried out by the same person (M.A.).

The questionnaire consisted of: 1) a figure of the whole body and face where the pain distribution was to be outlined, 2) 3 tables of expressions describing pain intensity, emotional distress, and pain character, 3) 52 general questions about pain and circumstances related to the pain, 4) the 52-item Illness Behavior Questionnaire (15), and 5) a table where different treatments were to be listed, and the patient's judgment of the therapeutic result.

All 45 patients received written information and were informed that they could withdraw from participation at any time during the investigation. The investigation was approved by the Ethics Committee of the Huddinge University Hospital.

Results

Relevant diagnosis

Ten patients (22%) reported no orofacial pain at the second admittance. A proper diagnosis resulting in successful therapy was produced in only two cases; in one of these there was a medical diagnosis (*polymyalgia rheumatica*) and a treatment with steroids for 10 years. The orofacial pain as well as the pain in other parts of the body had disappeared. Another patient had been given an odontological diagnosis (root fracture) and the orofacial pain was successfully treated by extraction of the affected tooth.

A diagnosis based on the result of treatment (*diagnosis ex juvantibus*) can be suggested in 8 cases: according to one patient the interruption of taking all sorts of analgesics resulted in disappearance of the facial pain. Three patients had their amalgam fillings replaced by other material and one of these had all teeth in the upper jaw extracted. Another patient reported that extraction of 2 teeth combined with being treated by 'the right dentist' resulted in disappearance of the pain in the face. In 2 cases the orofacial pain disappeared after stomatognathic treatment combined with endodontic and prosthetic treatment. Finally, the eighth patient reported acupuncture treatment to be the reason for the disappearance of pain.

The diagnosis chronic idiopathic orofacial pain must be considered relevant in those 35 patients with persisting facial pain with or without other pain problems.

Distribution between facial pain and the rest of the body

All the patients in this cohort had chronic facial pain when they were admitted to the FDPG. Only 2 were totally pain-free at the time of the second admittance 14 and 16 years later, respectively (Group 1). Another 2 patients suffered no pain other than facial pain at the first admittance but had developed such pain by the time of the second admittance (18 and 19 years later, Group 2). The 6 patients in a third group reported both facial pain and bodily pain at the first contact with the FDPG. Twelve to 18 years later the facial pain had disappeared but the bodily pain persisted. The main group (no. 4), consisting of 22 patients, had both facial and bodily pain at the first admittance and at the second admittance 6 to 19 years later. The same pattern was seen in 3 patients when they arrived for the first time (Group 5). When they were seen again 10 to 12 years later, facial pain persisted but bodily pain had disappeared. The sixth group consisted of 5 patients who had no pain other than facial pain at the first admittance but had developed such pain at the second admittance 12 to 19 years later. Finally, the remaining 5 patients (Group 7) reported facial pain but not bodily pain at the first admittance and the pattern was the same 9 to 12 years later. The pain duration (mean and range) in the different groups is presented in Table 1.

Response to invasive odontological treatment

Thirty-three patients underwent some sort of invasive treatment before the first admittance. Twenty-two of them were exposed to more than 75 endodontic treatments and more than 25 apicoectomies were performed in 17 patients. Between the first and second admittance, 18 patients were exposed to invasive treatments. Nine of these 18 patients underwent 12 apicoectomies. In this period, only 2 endodontic treatments were performed. None of these interventions resulted in any permanent pain relief.

The most popular invasive treatment was extraction of one or more teeth. At the first admittance it was found that more than 50 teeth had been extracted in 17 patients. In one of these cases all molar teeth had been removed on one occasion. In another patient all teeth had been extracted on one occasion at an unknown clinic abroad. The exact number of invasive treatments before the first admittance was not known.

Between the first and second admittance more than 100 teeth were extracted in 14 patients. Eight of these patients had more than 8 teeth extracted. In only 3 patients did

extractions result in permanent pain relief. The majority of extractions therefore did not produce any permanent pain relief.

Response to other dental treatments

Replacing amalgam fillings with other materials was performed in 5 cases in connection with the first admittance and in 7 cases between the first and second admittances. These treatment procedures resulted in permanent pain relief in 3 patients. Stomatognathic treatment was frequently used. The 26 patients treated prior to the first admittance did not report any relief. However, out of 11 patients subjected to this therapy in the period between the two admittances, one (18%) reported 'some' and another 'good' permanent pain relief. A similar pattern was observed concerning prosthetic rehabilitation. At the first admittance, 6 patients were treated but with no success. In the period between the first and second admittances 9 patients were subjected to prosthetic rehabilitation and 1 (11%) reported permanent pain relief. One patient had implant therapy, which did not produce permanent pain relief. Nerve block with local anesthetics in 5 cases and flour therapy in 1 patient did not result in permanent relief.

Response to other treatments

Acupuncture treatment was administered to 10 patients (4 reported at the first admittance and another 6 at the second). Three patients reported 'good' and 'some' permanent pain relief, and another was completely pain-free. No effect was reported by the 8 patients who received transcutaneous electrical nerve stimulation (TENS) treatment. One (9%) out of 11 patients treated with physiotherapy reported some permanent pain relief. One patient (in group 3) reported at the second admittance that just being referred to 'the right dentist' resulted in good permanent pain relief. However, the interview revealed that this patient also responded with good permanent pain relief to acupuncture and extraction of 2 teeth.

Discussion

In the literature, patients with facial pain have frequently been studied without the pain in the rest of the body being taken into account. From such a perspective 10 patients in this study had been considered pain-free. In our study, the whole patient was investigated, both facial pain and pain in other parts of the body, and a complicated pattern was revealed. Based on the change in pain distribution, the cohort could be divided into 7 groups: During the 9 to 19 years between the first and second admittance, 10 patients with facial pain remained free of pain otherwise (groups 1, 5, and 7). The case of polymyalgia rheumatica, however, is a contradiction, as this disease frequently causes pain throughout the body. The connection between polymyalgia rheumatica and temporal arteritis (16–18) may explain the facial pain. One patient reported that stopping taking medication was the reason for the disappearance of the facial pain. It is known from the literature that headache can be caused by overuse of migraine medications (19). We are unaware of any report of orofacial pain induced or maintained by analgesic overuse. Seven patients reported a permanent response to traditional odontological treatment and one responded to acupuncture.

Twelve patients (groups 2, 6, and 7) reported no pain other than facial pain when they were originally referred to the FPDG. Seven of these (58%) developed pain other than facial pain during the 9 to 18 years between the first and second admittance (groups 2 and 6). The 28 patients of groups 3 and 4 had suffered persistent pain other than facial pain for the whole follow-up period. These findings point to a general mechanism behind the atypical facial pain. It has been suggested (3, 9) that atypical facial pain may have much in common with unexplained medical syndromes that also affect other parts of the body. The complex etiological background to such syndromes can involve the central nervous system and psychological factors (10). Based on the assumption that there is a general mechanism behind idiopathic orofacial pain, treatment with acupuncture can be expected to be successful (20). However, only 1 of 10 patients treated with acupuncture reported permanent relief. The fact that

Table I. The cohort ($n = 45$) divided into 7 groups according to development and disappearance of both facial pain and bodily pain

	1st admittance		2nd admittance			Time between the 1st and 2nd admittance (years)		Pain duration (years)	
Group	Face	Body	Face	Body	No.	Mean	Range	Mean	Range
1	Yes*	Yes	No†	No	2	15.0	14–16	1.5	1–2
2	Yes	No	No	Yes	2	18.5	18–19	6.0	2–10
3	Yes	Yes	No	Yes	6	15.2	12–18	7.2	3–12
4	Yes	Yes	Yes	Yes	22	13.5	6–19	22.3	13–50
5	Yes	Yes	Yes	No	3	11.3	10–12	14.7	13–17
6	Yes	No	Yes	Yes	5	16.0	12–19	20.0	13–23
7	Yes	No	Yes	No	5	10.4	9–12	16.4	13–21

* Pain reported (Yes). †No pain reported (No).

the majority of patients suffering from chronic idiopathic orofacial pain are female (4) indicates that female hormones may be related to perceived pain (5, 6).

Although still suffering facial pain during the time between the first and second admittance, 3 patients (group 5) had recovered completely from the bodily pain they reported originally. One of these patients reported some permanent pain relief from odontological therapy and that the facial pain had shrunk from more than half the face to a small area during the period between the two admittances. This may indicate that the pain is progressively decreasing in this group.

The response to invasive dental treatments was meager. Up to the first admittance 33 patients had been exposed to more than 150 endodontic or surgical treatments. Of these, 17 underwent more than 50 extractions, and in 22 patients more than 75 endodontic treatments were performed. None of these treatments resulted in any permanent pain relief. In the period between the first and second admittance, invasive treatments were performed in 18 patients. Fourteen of these were exposed to more than 100 extractions, which resulted in good permanent pain relief in 3 patients. The very low success rate suggests that extraction should be considered contraindicated in patients with idiopathic chronic orofacial pain.

Out of 45 patients suffering from idiopathic orofacial pain on referral to the FDPG, 22% were free of orofacial pain and 4% were totally free of pain 9–19 years later. One of the pain-free patients had been given a medical diagnosis (*polymyalgia rheumatica*) and another an odontological diagnosis (root fracture), and 8 had responded to treatment (*diagnosis ex juvantibus*). In 35 cases the orofacial pain was still considered idiopathic. The relationship between facial pain and pain other than in the face had changed over the 9 to 19 years' follow-up period and is reflected by the division of the cohort into 7 groups. Concerning response to treatment; >75 endodontic treatments, ≥ 25 apicoectomies, and >50 extractions were performed in connection with the first admittance, which resulted in no cases of permanent pain relief. Between the first and second admittance, 18 patients were exposed to 2 endodontic treatments, 12 apicoectomies and more than 100 extractions resulted in good permanent pain relief in 3 patients. The low success rate suggests that such invasive methods are to be considered contraindicated in patients suffering from idiopathic orofacial pain.

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