Psychotherapy in the treatment of chronic refractory orofacial pain. Case reports

Psicoterapia no tratamento da dor orofacial crônica refratária. Relato de casos

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ABSTRACT

BACKGROUND AND OBJECTIVES: Psychological intervention can contribute to repairing the quality of life and altering behavior while dealing with chronic orofacial pain, especially those that can become refractory to treatments. Thus, the objective of this study was to present the narratives of three patients with refractory chronic neuropathic pain during the process of psychotherapeutic intervention and the follow-up results.

CASE REPORTS: Pain behavior, pain perception and depressive and anxious symptoms were assessed. Patients were under treatment at the Orofacial Pain Clinic and the instruments used in the beginning and end of psychotherapy were the visual analog scale, Beck Inventories (anxiety and depression), Wisconsin Pain Inventory, McGill Pain Questionnaire and Pain Catastrophizing Scale. They attended weekly 50-minute sessions. Each narrative was qualitatively analyzed and the comparison between the evaluations made before and after psychotherapy was included in the context of a phenomenological approach.

CONCLUSION: Emotional familiar conflicts and fear of pain crises were the most important aspects described by these patients. Case 1 presented the less cooperative profile and secondary gains. Despite of that, all cases presented improvement and psychotherapy helped to cope with their problems and pain. There was a remarkable impact on the life of patients as well as on their cooperation with pain treatments while helping the patients to build a proactive attitude and to understand their role in their condition.

Keywords: Chronic pain, Neuropathic orofacial pain. Pain behavior, Psychotherapy, Trigeminal neuralgia.

RESUMO

JUSTIFICATIVA E OBJETIVOS: A intervenção psicoterapêutica é uma ferramenta importante para melhorar a qualidade de vida de pacientes com dor orofacial crônica, especialmente aqueles que se tornam refratários aos tratamentos. Assim, o objetivo deste estudo foi apresentar, na forma de narrativa, três casos de pacientes com dor facial refratária ao longo do processo de intervenção psicoterapêutica e os resultados obtidos.

RELATO DOS CASOS: Observou-se o comportamento, a percepção da dor e sintomas ansiosos e depressivos. Os pacientes estavam sob tratamento na Equipe de Dor Orofacial e os instrumentos utilizados no início e no final da intervenção foram a Escala Visual Analógica, os Inventários de Beck para Ansiedade e Depressão, o Inventário de Dor de Wisconsin, o Questionário de Dor McGill e a Escala de Catastrofização de Dor. Os pacientes foram atendidos semanalmente em sessões de 50 minutos. Cada narrativa foi analisada qualitativamente e a comparação entre as avaliações (antes e depois do tratamento) foi incluída em um contexto de abordagem fenomenológica.

CONCLUSÃO: Conflitos familiares emocionais e medo das crises de dor foram os aspectos mais importantes descritos pelos pacientes. O caso 1 apresentou um perfil menos cooperador, embora todos tenham apresentado melhora em algum âmbito avaliado. A proposta psicoterapêutica os auxiliou no enfrentamento do sofrimento por conta de seus problemas e da dor. Houve um impacto notável na vida dos pacientes como em sua cooperação com os tratamentos, uma vez que aprender a lidar com seu sofrimento os conduziu a uma postura mais proativa através do entendimento de seus papéis no tratamento da condição dolorosa.

Descritores: Comportamento da dor, Dor crônica, Dor orofacial neuropática, Neuralgia do trigêmeo.

INTRODUCTION

According to the International Association for the Study of Pain, pain is characterized by “an unpleasant sensory and emotional experience associated with, or resembling that associated with, actual or potential tissue damage”. As a complex biopsychosocial phenomenon, it becomes chronic when its duration surpasses three to six months. Sensory and emotional experiences in suffering and triggers pain behaviors, which can also become chronic¹. These behaviors depend on personality, anxiety and depression traits or diagnoses and patients’ strategies for coping. Pain is subjective and individualized, and its duration and natural history can interfere with clinical features.
Chronic pain is prevalent, and 30% to 40% of individuals with pain go five times more to the emergency room than other patients\textsuperscript{1-4}. It is a highlighted reason for absenteeism, and it has a severe impact on life. Orofacial pain affects 10% to 30% of the population and its psychological impact is remarkable due to the psychological and social importance of the face, and vital functions of this body area such as eating, breathing and talking\textsuperscript{5,6}. Most patients present perceptual distortions of the face\textsuperscript{7}. Moreover, a subgroup of orofacial pain that is neuropathic is responsible for 4-18% of cases, and their treatment often results in partial alleviation of symptoms\textsuperscript{8}. Neuropathic pain is associated with dysfunction or lesion of the nervous system\textsuperscript{9}.

The most common types of neuropathic orofacial pain are trigeminal neuralgia, burning mouth syndrome and post-traumatic neuropathic pain; other chronic facial pain syndromes that have complex assessment are persistent idiopathic facial pain and atypical odontalgia\textsuperscript{8}. Trigeminal neuralgia is considered one of the worst pain conditions and the intense suffering can even lead to suicide. In general, it is idiopathic, however around 5% of the patients present a primary cause (eg. meningioma, schwannoma, multiple sclerosis)\textsuperscript{10,11}.

Burning mouth syndrome is characterized by continuous burning pain at the oral cavity with no primary cause. These patients often present emotional comorbidities and severe impairment in daily activities\textsuperscript{12}. Persistent idiopathic facial pain and atypical odontalgia consist in constant, dull, aching pain with unclear pathophysiology\textsuperscript{8}.

Neuropathic pain patients often suffer from sequelae of surgical procedures including complications from invasive procedures to treat pain. There are several psychiatric comorbidities in these patients, including depression, dystimia, social phobia, generalized anxiety, and worrying about the pain is a main associated factor in patients with orofacial pain\textsuperscript{13}. This complex clinical condition requires special medical and psychological assessment. Chronic pain can be associated with subjective bad self-description, reinforced by words the patients heard previously in their lives\textsuperscript{14,15}.

Psycho-educative tools are part of the strategy for pain coping, and philosophical precepts are the base of intervention techniques to alleviate or eliminate suffering. Treatment modalities include psychoanalysis, cognitive behavior therapy, biofeedback and hypnosis. Graduated exposure to pain and systematized desensitization helps the patient release from the fear of pain and return to normal life, both constituting positive coping mechanisms. Negative coping in chronic pain is associated with secondary gains and/or avoidance of several activities that were part of the normal daily life of the patient\textsuperscript{16}. Positive and negative reinforcements or punishments need to be identified and addressed.

There is evidence in the literature that chronic pain patients can benefit from psychotherapy\textsuperscript{17}; however, there is a lack of studies on orofacial or neuropathic pain. Thus, the objective of this research was to present the narratives of three patients with refractory chronic neuropathic pain during the process of psychotherapeutic intervention and the follow-up results.

**CASE REPORTS**

All patients were informed about the purposes of this study and signed the informed consent. The local ethics committee from University of São Paulo (1.172.960) approved this study. This manuscript was written based on CAsE RePort Guidelines (CARE)\textsuperscript{18}.

**Patient Information**

Primary concerns and patient symptoms.

The narratives of three patients at the Orofacial Pain Clinic of a general hospital are here recorded. They were a woman with burning mouth syndrome, a man with idiopathic trigeminal neuralgia and a woman with secondary trigeminal neuralgia and multiple sclerosis. All of them were literate and had had orofacial pain for more than 6 months.

There were no complaints of generalized pain or pain in other parts of the body. They were under pain treatment, which was not changed during the period of psychotherapy and evaluation of this study. It consisted of antidepressants and physical treatments (heat packs and exercises).

**Timeline**

Patients were individually evaluated and the weekly individual sessions lasted 50 minutes by a trained psychologist, with supervision of a psychologist with robust experience with orofacial pain. Duration of the treatment was individually determined, and the cases were discussed among the researchers after the sessions to establish the assessment. It included psychoanalysis, familiar and systemic psychological approaches.

**Diagnostic assessment**

In the beginning and in the end of psychotherapy, the patients were evaluated with:

1. Visual analog scale to determine the pain intensity. Consists of a scale from zero to 10: zero represents no pain and 10 represents the worst possible pain.
2. Beck Depression Inventory\textsuperscript{19} validated to the Portuguese language to determine depressive symptoms. Consists of 21 questions that can be scored from zero to 3. According to the final score, the patient can be classified as having absent or minimum (zero-13), mild (14-19), moderate (20-28) or severe (29-63) depression.
3. Beck Anxiety Inventory\textsuperscript{19} validated to the Portuguese language to determine anxious symptoms. Consists of 21 questions that can be scored from zero to 3. According to the final score, the patient can be classified as having absent or minimum (zero-13), mild (14-19), moderate (20-28) or severe (29-63) anxiety.
4. Wisconsin Pain Inventory\textsuperscript{20} validated to the Portuguese language to determine pain limitations.
5. McGill Pain Questionnaire (short form)\textsuperscript{21} validated to the Portuguese language to determine pain descriptors. Consists of 15 items divided into 3 domains (sensitive, evaluative, and affective).
6. Pain Catastrophizing Scale\textsuperscript{22} validated to the Portuguese language to determine thoughts, perceptions or feelings associated...
with the pain. Consists of 13 items with numeric scores from zero to 4 for each of them.

Clinical findings, therapeutic intervention, follow-up and outcomes
Clinical findings are presented individually for each case, including relevant past symptoms, as well as interventions and their outcomes. Similarly, types and administration of therapeutic intervention besides changes in therapeutic interventions with explanations. Additionally, clinician- and patient-assessed outcomes, important follow-up diagnostic and other test results are presented in this section.

Data analysis
Each narrative was qualitatively analyzed and the comparison between the evaluations made before and after psychotherapy was included in the context of a phenomenological approach. Each patient showed specific demands that are presented in this study. Parts of the exact narrative of the patients were transcribed.

Case 1
Woman, 55 years old, having had pain for more than 20 years, was diagnosed with burning mouth syndrome at the Orofacial Pain Clinic. Pain started after a surgery for dental implant. Burning was moderate during the day and got worse at night. Burning mouth syndrome is a neuropathic orofacial pain, more common in women after menopause, and causes intense suffering and emotional impairment. Although not having reported her beliefs about the origin of her pain, she has described fear of the invisible as something with high impact in her perception of life. At the pain onset, she already had depression and an episode of hospitalization because of a suicide attempt by ingesting an excessive quantity of drug (90 tablets of clonazepam). “I swallowed 90 clonazepam pills with no use, I do not think any more about killing myself because I am no worthy”. She attempted suicide to free herself from work because she could not stand the people there. It happened after 11 years of health leaves that kept her away from work sporadically, until finally she had to come back; then she retired on disability. Recently she has been diagnosed with bipolar disorder. She studied social sciences and worked for a little time in a hospital. “I stopped doing my job because I did not believe in what I was doing”. After that, she worked in the public sector in a position of trust. She had also been a secretary and police officer. She is single and lives with her parents. Her daily routine is to wake at 3h in the morning, keep lying down in bed until 6h am, walk the dog, go shopping at the market and the drugstore and come back. She has lunch at 11h am and often takes her parents to doctor appointments in the afternoon. She lays down to sleep at 6 pm. She has limited interests which include going out to eat at the bakery. She does not like physical activities because the pain gets worse.

Love relationships
The patient avoided talking about her affective relations, especially love relationships. “I do not cry anymore, I have cried a lot in my life, I have cried a lot for men”. Among the relationships, there was a disappointment with a boyfriend at college and an alcoholic man she lived with: “I wanted to be there because I did not want to come back to my home (parents’ house)”. After that, she was in love with a doctor she had worked with as a secretary but got disappointed. Currently, she is not in any love relationship because “after this burning I do not want to kiss, it makes me sick”.

From the last boyfriend she got herpes papilloma virus. Since diagnosed, she has not treated it. She thinks she should treat it now to avoid cancer.

Family and friends
This patient reported a terrible relationship with her father. “He is a religious fanatic, always saying that I am not a God’s person and that I am possessed”. She does not respond to these provocations because she fears she will be injured. She considers her mother a good person although the mother is too submissive to her father. “All he says, she accepts. She is on his side”. Her mother has cardiac disease, which worries her. She is afraid of her mother dying because she will miss her and will not be able to deal with that. She is worried about having to take care of her parents. “I will be the one who will have to take care, change diapers, take to the doctor, I don’t know if I can do that”. “I will do that because I will have to, if there was somebody else, I would like that”. She is the oldest daughter, and she cannot count on her sister or brother. “My sister lives in another state and my brother does not care”. They do not help her financially with the parents either. Other relatives are distant. There are no friends. She has a restricted social life. “I tried to go out and talk to people, I am always crying, it is useful. I like to stay in my room”. Although the psychologist has told her that activities help to manage pain, she does not want to do any. “I come here to treat my pain”. “I do not want to talk to others because my mouth burns, and I do not want to hear others complaining. I want to end this pain, only”.

She got a dog to help her lose her fear of being alone. “I am afraid of spirits”.

Self-description
She describes herself as a woman who hardly smiles or laughs. She talks loudly and has bad temper, feels bad about being overweight, but does not do anything to change it. She does not take care of herself due to laziness. She prefers to lie in bed. Her hair is white, and she does not want to spend money with that. She prefers to spend the money with medicine. Before being retired, she liked to go shopping and was compulsive with that. “I’ve bought many things, I loved to buy clothes”. She takes laxatives every day. “I need to go to the toilet every day, otherwise I get sick”. She does not like to take a shower every day. She feels that the best would be to live in the hospital. “You have to enjoy when you are young, I do not see the world through rose-colored glasses”.

Behavior repertoire
The behavior repertoire is limited with few reinforcements. The only positive ones are to go to the bakery and to come to therapy. However, to go to the bakery is not always possible because it is expensive.
for her. The main negative reinforcement is the father by whom she is afraid of being punished. The pain is the reason for why she does not go out, travel, or change the routine. She is very resistant to suggestions such as new activities, self-care, or social contact.

Psychotherapy results and pain alleviation
After six weeks of psychotherapy and severe pain (pain intensity = 10), her pain intensity became 9. She started to make jokes, to use makeup and to smile. The initial and final scores can be observed in table 1. The depression scores did not change but there was a remarkable difference in the anxiety and catastrophizing scores. An increase in sensitive and evaluative McGill indexes is possibly associated with the attention given to pain issues during therapy, and it was associated with the increase in catastrophizing score. Wisconsin scores for humor, walking ability, work, personal relationships, sleep and enjoy life improved.

Case 2
A 63-year-old man, retired, has had idiopathic trigeminal neuralgia for 23 years. He had treated it, and during the first 8 years, the pain was controlled, although there were several adverse effects. “You are and are not there, you get slow”. Trigeminal neuralgia is an excruciating paroxysmal shock-like pain, usually triggered by a light touch. He underwent four neurosurgeries. The last was 4 months ago and now the pain has been relieved. His routine is to wake up, do housekeeping, make woodcrafts and help the community he lives in. At the early stages of his pain, an intense crisis was associated with suicidal thoughts, of which he spoke about nervously. “I do not wish that pain to anyone; if I could I would have taken my life away”. He was in the bathroom and remained standing with his hands against the wall for hours. “I could not move; any movement caused pain”. He wanted to commit suicide but could not write a note to explain the reason. After the first surgery, he got depressed because his face became numb. “A person who has this type of pain loses 50% of their lives (...) I feel mutilated”.

Family, relationships, and self-description
Childhood was complicated and full of family problems, with the need to work since being a kid. He learned several jobs and now dedicates himself to his family and the community. He lives with his wife and has a good relationship with his sons. He described himself as being sensitive and full of hope. However, “people who live close to my house are negative”.

Coping
He has many plans and wants to start computer and chess courses. He wants to meet new people in these courses. His feelings about the pain possibly reoccurring are contradicting: although he hopes the pain does not come back, at the same time he is afraid that happens.

Psychotherapy results and pain alleviation
Listening about the crisis was the assessment tool in this case. To avoid the expectation of new crises is the challenge for trigeminal neuralgia patients. The scores for pain intensity, depression, anxiety, Wisconsin, McGill, and catastrophizing decreased (Table 1). Accepting the condition and developing an action plan in case the pain returns were the contents developed.

Case 3
A married woman aged 36 years old has had trigeminal neuralgia crises for 10 months. At the diagnosis, she also fulfilled the criteria for multiple sclerosis, which characterized secondary trigeminal neuralgia. Multiple sclerosis is a neurodegenerative autoimmune disease that can present demyelination at the entry zone of the trigeminal root, as it happens in this case25. Emotional stress and weather variations can trigger crises and this patient showed several familiar issues to be assessed. Her routine is to wake up early and work at a public service in the mornings, prepare lunch for three kids and her husband, and in the afternoon help her husband at work. He is a school bus driver. At night, she does some housekeeping.

Familiar relationships
Her oldest son is 15 years old. He is from a previous marriage. Her daughter is 10 years old, and the younger son is 6 years old. She complained about difficulties in the relationship between her current husband and the older boy “although they know each other for a long time”. “I got separated from my first husband when he was 1 year old, they are always arguing”. She thinks her husband is too much demanding, especially with the older one. “I always feel my face burning when they fight. It happens always when they are both at home”. This son had problems with drugs at school at the same time as the biological father started to have a relationship with him. After that, his father disappeared. He changed schools and failed a year. The current husband speaks too loudly, which is one of the reasons for arguing. She thinks her marriage is great and there are rare arguments about the relationship. The arguments are about the son. “He (husband) misses my attention, but I do not like to demonstrate my love. I have this same difficulty with my older son too”. She wanted a closer relationship with both, especially the husband. The older son is lazy with home tasks, and she does them for him to avoid the arguments. Once, one argument turned into a pain crisis. “I went to bed and cried; my husband does not believe in my symptoms”.

Self-description
At 14 years old, her father died, and she became independent. She worked since a young age and had her first son with only 21 years old. Disagreements were the cause of separation and divorce. “I was sensitive and accepted what the others told me to do”. She cried when her husband yelled at her and was very insecure and shy. “After this diagnosis, I am not so submissive anymore”.

Psychotherapy results and pain alleviation
Listening to complaints and working on family communication were the strategies in this case. A pain diary was also created to find out the relation between the crises and other issues24. She
clearly needed help to change the repertoire. Her behavior was proactive and responsive to the suggestions. Assertiveness was assessed and helped reducing pain crises and conflicts. There was an improvement in her relationships with the husband and the older son. “When they start to discuss I get out, when I come back, they have stopped it, it works”. She negotiates with the husband what tasks the older son should carry out for it to become less demanding on him.

The increase in scores shown in table 1 reflects the diagnosis of multiple sclerosis, which occurred after the initial evaluation. Multiple sclerosis is a progressive disease with several morbidities and severe impairment.

**DISCUSSION**

These narratives of pain suffering show the complex integration of multiple factors in pain expression and coping. These are common diagnoses at the pain clinic, but their expressions are singular. General behaviors and generalized psychological characteristics can be found in epidemiological studies, but only their investigation in individuals allows to determine nuances and slight differences which are crucial for the success in the treatment. The phenomenological investigation is necessary to understand psychological tendencies in behavior reported in sample studies in the literature.

There are different and unique life stories and different attitudes in front of the same symptom such as neuropathic facial pain. The singularity and specificity of everyone are essential in the choice of therapeutic intervention, although the common objective for all of them is pain alleviation. The approach includes management of family and interpersonal conflicts, reducing deficits in the repertoire for pain coping and problem solving. For some people, chronic pain seems to be more reliable than absent parents or fragile relationships. Pain is part of them.

Moreover, secondary gains, as observed in case 1, can be characterized by the resistance to cooperate with the therapeutic intervention and excessive valuing of the hospital care, and it can be associated with the increase in catastrophizing score. The persistence of the symptom seems to gain an operating function, which has also been observed in patients with chronic fatigue. The positive reinforcements would take the individual away from the intense suffering condition, which is not the case while secondary gains are at stake.

Those cases represent the challenge for therapy: to find alternatives of social reinforcement that would play a role for pain alleviation. However, the contexts of life stories can halt and complicate the assessment. Inertia from inactivity is another important issue because of the tendency to keep the pain alive in the routine. Psychoeducation and gradual suggestions are the way, but it must be individualized. On the other hand, it is natural to avoid anything that could potentially increase pain. This type of belief increases the resistance to find pain coping mechanisms and to come back to life, with a high cost for personal and family relationships.

Although suffering was present in all cases, these are three distinct people. Case 1 has a high level of resistance and secondary gains; case 2 has better strategies for pain coping and familiar and social support; and case 3 demonstrates a cooperative patient with several family problems, but the proactive attitude contributes to improvement. The resilient profile in cases 2 and 3 is, without a doubt, very positive for the psychotherapeutic approach. However, patient 1 needs creative strategies.

This qualitative study does not yield statistical analyses. However, epidemiological evidence testifying to the psychological aspects of chronic pain are abundant in literature. This study complements previous findings with the evidence that, despite the common issues, there are particularities that will make the final difference in patients’ rehabilitation for life.

**Table 1. Initial and final scores for pain intensity, depression, anxiety, Wisconsin sub-items, McGill indexes and catastrophizing**

<table>
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<tr>
<th></th>
<th>Case 1 Initial</th>
<th>Case 1 Final</th>
<th>Case 2 Initial</th>
<th>Case 2 Final</th>
<th>Case 3 Initial</th>
<th>Case 3 Final</th>
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<tbody>
<tr>
<td>Pain intensity (VAS)</td>
<td>10</td>
<td>10</td>
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<td>8</td>
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<td>Depression score (Beck)</td>
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<td>3</td>
<td>2</td>
<td>17</td>
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<tr>
<td>Anxiety score (Beck)</td>
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<td>13</td>
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<td>Limitations (Wisconsin)</td>
<td>General</td>
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<td>10</td>
<td>2</td>
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<td>6</td>
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<td></td>
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<td>3</td>
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<td></td>
<td>Walk ability</td>
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<td>7</td>
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<td>1</td>
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<td>Sleep</td>
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<td>0</td>
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<td></td>
<td>Enjoy life</td>
<td>9.5</td>
<td>9</td>
<td>3</td>
<td>2</td>
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<td>3</td>
<td>1</td>
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<td>3</td>
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<td>12</td>
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CONCLUSION

This study showed that psychotherapy had a remarkable impact in the life of patients as well as in the cooperation with pain treatments, even in a less resilient patient. While assessing individual profiles and particularities of each case, it is possible to help patients have a proactive attitude and to understand their role in their condition.

AUTHORS’ CONTRIBUTIONS

Mariana Barbosa Yamaguchi
Data Collection, Research

Maria de Fatima Vidotto Oliveira
Supervision

Silvia Regina DT de Siqueira
Project Management, Methodology, Writing - Preparation of the original, Writing - Review and Editing, Supervision

REFERENCES
