Open Access Scientific Evidence of Cognitive Behavioral Therapy for Patients with Fibromyalgia

Evidencia científica de acceso abierto de la Terapia Cognitivo Conductual para pacientes con fibromialgia

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Abstract. Objective: To provide a summary of scientific open access resources Cognitive Behavioral Therapy interventions in patients with fibromyalgia. Method: Bibliographical search performed in Pubmed and PsycInfo, Latindex, Scielo and Redalyc online databases. No year of publication criterion was applied. Results: 377 citations were reduced to a list of 59 relevant publications, from 1993 to 2016. Most studies were performed in adult samples, in Spain or the United States, with pre-post quantitative design. Only 15 publications were open access. Conclusions: Empirical evidence is ample, yet mostly restricted. Open access material might promote interest in and encourage the design, implementation and evaluation of CBT interventions in the benefit of fibromyalgia patients, the health system and science.

Keywords. Fibromyalgia, Psychotherapy, Cognitive Behavioral Therapy, CBT, Open Access.

Resumen. Objetivo: Proveer un resumen de la evidencia científica de accesos abierto sobre intervenciones con Terapia Cognitivo Conductual en pacientes con fibromialgia. Método: Se realizó una búsqueda bibliográfica en las bases en línea de Pubmed y PsycInfo, Latindex, Scielo y Redalyc. No aplicó criterio por año de publicación. Resultados: Las primeras 377 citas obtenidas se redujeron a una lista de 59 publicaciones relevantes, publicadas entre 1993 y 2016. La mayoría de los estudios se realizaron en muestras de pacientes adultos, en España o Estados Unidos, con un diseño cuantitativo de tipo pre – post. Únicamente 15 publicaciones fueron de acceso abierto. Conclusiones: La evidencia empírica es amplia; sin embargo, el acceso a la misma es generalmente restringido. El material de acceso abierto puede promover el interés en las intervenciones con Terapia Cognitivo Conductual y estimular su diseño, implementación y evaluación, todo esto en beneficio de los pacientes con fibromialgia, los sistemas de salud y la ciencia.

Palabras clave. Fibromialgia, Psicoterapia, Terapia Cognitivo Conductural, TCC, Acceso Abierto.

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Introduction

Fibromyalgia syndrome (FS) is a term used for identifying a disorder mainly featured by chronic widespread pain perceived at musculoskeletal sites (Laroche, Sarzi-Puttini, & Perrot, 2015). Despite FS existence being recognized since long time, its definitive delineation as a distinctive clinical disorder emerged until 1990, when the American College of Rheumatology (ACR) delivered its first classification criteria (Wolfe et al., 1990), which were recently updated (Wolfe et al., 2010) and further modified (Wolfe et al., 2011).

Worldwide FS prevalence is relatively high, affecting between 1-10 % of the adult general population (Clauw, 2014; Laroche et al., 2015). In rheumatology care centers, FS can be diagnosed on 20% of referred subjects, and it is the second most common diagnosis in those sites (Buskila, 2001). Besides its relatively high prevalence and high demand of medical services, SF can be considered as an important medical and social burden because of its chronic and potentially disabling nature, in addition to the direct, indirect and untangible costs imposed to the society and the affected subjects (Ghavidel-Parsa, Bidari, Amir Maafi & Ghalebaghi, 2015; Skaer, 2014).

The typical FS patient is a middle aged woman (relation female:male from 3:1 in general population to 7:1 in hospital-based population) (Clauw, 2014; Laroche et al., 2015). Yet, estimates suggest that FS can onset early in life; they also suggest that it affects 2.1-6.1% of school children, mostly adolescent female patients, and patients with juvenile-onset fibromvalgia constitute a substantial proportion (7-15%) of referrals to pediatric rheumatology clinics (Kashikar-Zuck & Ting, 2013). The main complaint in patients is chronic diffuse musculoskeletal pain, particularly, but not exclusively, located at the axial muscles affecting symmetrically the upper and lower part of the body. Most of the affected people (80%) consistently refer fatigue, non-restoring sleep, depression, anxiety, and impaired cognition (a.k.a. fibrofog) (Clauw, 2014; Laroche et al., 2015). Interestingly, a large series of unspecific symptoms, such as acral paresthesia, nausea,

bloating, dysmenorrhea, or antecedent somatization disorders, such as non-infectious cystitis, irritable bowel syndrome, migraine, tension headache, and premenstrual tension syndrome can be elicited in a high proportion of patients (Clauw, 2015; Laroche et al., 2015).

FS diagnosis is usually reached by exploration of clinical history and the exclusion of other rheumatologic (e.g. rheumatoid arthritis, systemic lupus erythematosus, myositis, and polymyalgia rheumatic) and non-rheumatologic (mainly hypothyroidism) conditions as the cause of patient symptoms (Clauw, 2014; Rahman, Underwood & Carnes, 2014). In those patients, the ACR modified criteria can be a useful diagnostic tool (Wang et al., 2015; Wolfe, 2011).

Until the present, FS cause is unknown; however, the absence of inflammation, and the relative lack of response to non-anti-inflammatory drugs strongly suggest that FS pain is not mediated by inflammatory pathways. Most of present evidence points toward a multifactorial etiology in which central pain sensitization, sleep disturbance, disordered hypothalamic-pituitary-adrenal axis, and vegetative tone anomaly interplay as the FS pain triggers mainly by the result of a reduced pain threshold (Boomershine, 2015; Laroche et al., 2015).

Although FS almost never results on structural derangement, affected people experience a moderate to severe impact on their health related life quality and the physical function. Current FS treatment is unsuccessful in a relatively significant proportion of patients. It has been established that less than 50% of subjects find complete symptomatic relief, and less than 30% reach complete remission after 3 years of the diagnosis (Laroche et al., 2015; Skaer, 2014).

Despite the above, most of fibromyalgia patients can be successfully managed in the present time by a multidisciplinary team that must include, at least, a rheumatologist, a specialist in physiotherapy and rehabilitation, and a mental health professional (Laroche et al., 2015; Theadom, Cropley, Smith, Feigin & McPherson, 2015).

A large series of non-pharmacological, such as patient education, graded exercise, complementary modalities, and psychotherapy, specifically the cognitive behavioral approach (Clauw, 2014; Laroche et al., 2015; Lauche, Cramer, Häuser, Dobos & Langhorst, 2015; Theadom et al., 2015) and pharmacological interventions, such as analgesics (i.e. tramadol and paracetamol), antidepressants (such as amitryptiline, duloxetine, milnacipran, and venlafaxine), and agonist of the $\alpha 2\delta$ subunit of the neuronal voltage-calcium channels (pregabalin and gabapentin) (Calandre, Rico-Villademoros, & Slim, 2015; Clauw, 2014; Häuser, Walitt, Fitzcharles, & Sommer, 2014; Laroche et al., 2015) have been proved as efficacious by clinical trials and systematic reviews, and when are used with a multidisciplinary approach can positively modify the level of pain, the health related quality of life and the physical function in the vast majority of fibromyalgia patients.

From a psychological perspective, patients with FS experience high distress and are likely to overuse health services when that occurs. About 32.3% of FS patients have an anxiety disorder, and about 34.8% have a mood disorder, especially of a depressive type. Negative emotions exacerbate pain and impairment and might even be more difficult to manage than physical symptoms. Maladaptive coping strategies, catastrophic thoughts, external locus of control, low sense of self-efficacy, and feelings of helplessness, a tendency to internalize and suppress anger, and reduction of positive emotions are common in FS patients. The way a person with FS manages her/his emotions is relevant to her/his adjustment to pain. Thus, therapeutic programs may be particularly helpful to these patients (Amutio, Franco, Pérez-Fuentes, Gázquez & Mercader, 2015; Sephton et al., 2007).

The most used psychotherapeutic intervention modality is group treatment with a cognitive-behavioral approach (Lami, Martínez & Sánchez, 2013). Cognitive Behavioral Therapy (CBT) emphasizes changes in thought patterns and behaviors rather than providing 'deep insight'. Its beneficial effects can be achieved in short-term (e.g. 8 to 10 sessions) on a one-to-one basis and/or in a group therapeutic setting. Some key elements of CBT for FS patients are: education about the nature of FS, realistic goal setting for daily activities, relaxation training, pacing of activities, identification of dysfunctional thought patterns and practice of techniques to counteract them, assertive communication skills to release tension, strategies for relapse prevention and for facing pain episodes (Bennett & Nelson, 2006). Systematic reviews and/ or meta-analyses suggest that CBT provides a small incremental benefit over control interventions in reducing pain, negative mood and disability at the end of treatment and at long-term follow-up (Bernardy, Klose, Busch, Choy, & Häuser, 2013). Moreover, although CBT does not provide better results than other non-pharmacological treatments on outcomes of pain, fatigue, sleep disturbance and quality of life, at either a short or long-term, it is more effective on symptoms of depression for a short period and considerably improves the pain self-management and reduces the number of visits to the doctor (Minelli & Vaona, 2012).

Among the various techniques in CBT, mindfulness is one of the most promising. Mindfulness can be described as non-judgmental attention to experiences in the present moment implying self-regulation of attention and acceptance of one's own experiences in a non-evaluative way (non-reactive awareness). For FS patients, mindfulness has become a valuable tool for learning new non-reactive models of responding to the emotional discomfort and to relate to pain without suffering (Amutio et al., 2015). Systematic reviews on mindfulness on CBT interventions for FS patients revealed low short-term improvement of quality of life and pain when compared to usual care and to active control interventions (Lauche, Cramer, Dobos, Langhorst & Schmidt, 2013; Theadom et al., 2015).

Given the psychological distress that patients with FS are likely to face, it is especially important to propose and to implement psychotherapeutic interventions tailored for this population based on scientific evidence of their effectiveness. Research on CBT in patients with FS is ample, and (systematic) reviews have been previously published. Given that most resources are restricted from the general public, the main goal of this review was to provide its readers with a useful and practical summary of scientific information currently available to all through open access online resources, regarding the specific content of CBT interventions.

Methods

A bibliographical search was performed in Pubmed and PsycInfo introducing the following terms: "fibromyalgia" + "CBT", "fibromyalgia" + "cognitive behavioral therapy", "fibromyalgia" + "cognitive behavioural therapy", "fibromyalgia" + "mindfulness". Additionally, Latindex, Scielo and Redalyc databases were consulted introducing the terms "fibromyalgia" or "fibromialgia" (in Spanish) searching further for relevant material. No year of publication criterion was applied. Online consultations proceeded on the 16 and 17 of May, 2016.

Citations to works other than research articles were first withdrawn. Following, original research was filtered and (systematic) reviews and/or meta-analysis were omitted. Through the information provided in their abstracts, the publications that reported research performed in patients, not exclusively with FS, or focused on the evaluation of other features/ tools for intervention rather than on the effectiveness of CBT on treatment, not primarily of CBT content, were excluded.

Finally, the following features were recorded from each publication: year of publication, age stage of participants (juvenile, adults), location of research team, design, and open accessibility online. All steps of the procedure were performed by both authors working together; discrepancies were minimal.

Results

The initial search elicited 377 citations, of which 51 were duplicated. Through the review of available abstracts, they were reduced to a list of 59 relevant papers (figure 1). Year of publication ranged from 1993 to 2016, with juvenile (n=6, 10.2%) and adult (n=53, 89.8%) samples. Most reports are from Spain (n=22, 37.3%) and the United States (n=22, 37.3%). Most designs were quantitative clinical trials (n=48, 81.4%) of one (n=18, 30.5%) or two (n=26, 44.1%) groups; five (8.5%) were qualitative studies, with 4 reporting clinical cases and 1 semi-structured interviews. Descriptive data are summarized in table 1.

Regarding open online accessibility, 15 publications met this criterion. Links are provided in Table 2. Additionally, in Appendix 1, links to other open access material that did not specifically met the objective of the study might be relevant to readers interested in CBT for patients with FS.

Discussion

Chronic pain conditions often lead to disability, psychological distress and decremented quality of life, yet FS presents particular challenges. Patients with FS compared with others experiencing similar levels of pain, even show higher levels of anxiety and depression, lower levels of positive affect, and greater alienation from others in their social networks and greater social and emotional dysregulation. They need to develop effective means to cope with their condition; otherwise, pain can take over their lives, reducing interaction with positive affective resources, and ultimately triggering long-term physical and psychological disability (Davis & Zautra, 2013).

CBT has become the most widely used behavioral treatment for pain, targeting emotional regulation by minimizing the negative consequences of pain and other stressors and by sustaining and increasing positive engagement in social relations that are key sources of positive affect. CBT is based on the cognitive theory of emotional responses; pathological negative emotions are considered the result of a dysfunctional thinking shaped by the patient's belief system. The main CBT therapeutic techniques are twofold: the identification / modification of dysfunctional thought patterns and the engagement in new behaviors that break the vicious circle between symptoms and patterns of



Figure 1. Study flow diagram.

dysfunctional performance (Bennett & Nelson, 2006; Davis & Zautra, 2013).

Reviews and/or meta-analysis on the evidence of the effectiveness of CBT in patients with FS have come to diverse, though not necessary contradictory, conclusions. In most studies, CBT, alone or in combination with other treatment modalities, provides small incremental benefit over control interventions in coping strategies, negative mood (e.g. depression, anxiety, psychological distress), self-efficacy, painrelated behavior, and disability at the end of treatment and at long-term follow-up (Bennett & Nelson, 2006; Bernardy et al., 2013; Glombiewski et al., 2010; Gritzner, 2011). CBT seems to be more effective on symptoms of depression for a short period, whereas it considerably improves the pain self-management and reduces the number of visits to the doctor (Minelli & Vaona, 2012). Sustained improvements in pain are most evident when individualized CBT was used to treat patients with juvenile fibromyalgia (Bennett & Nelson, 2006). Moreover, other results suggest that

Table 1			
Distribution of publicatio	ns on CBT intervention ou	tcomes	
	Juvenile sample	Adult sample	Total
	n = 6	n = 53	n = 59
Year of publication			
1993 - 2000	0	5	5
2001 - 2005	1	6	7
2006 - 2010	1	16	17
2011 - 2015	4	24	28
2016	0	2	2
Location			
Spain	0	22	22
United States	6	16	22
Sweden	0	3	3
Germany	0	2	2
Norway	0	2	2
Canada	0	2	2
Italy	0	1	1
France	0	1	1
Netherlands	0	1	1
Turkey	0	1	1
Brazil	0	1	1
Mexico	0	1	1
Clinical trial	6	48	54
1 group	4	14	18
2 groups	2	24	26
3 groups	0	9	9
4 groups	0	1	1
Open access	1	14	15

CBT does provide better results than other nonpharmacological treatments on outcomes of pain, fatigue, sleep disturbance and quality of life, at either a short or long-term evaluation (Minelli & Vaona, 2012); nor does it, as a single treatment modality, offer any distinct advantage over well-planned group programs of education and/or exercise (Bennett & Nelson, 2006). Further research addressing cost-effectiveness (e.g. visits to the doctor, disability pension, weeks of unemployment and sickness) of CBT will help us to better estimate its overall benefits for patients and the public health system (Feliu-Soler et al., 2016; Fjorback et al., 2013; Luciano et al., 2014).

A growing interest on the application of CBT in patients with FS in the scientific community is evident, as multiple efforts have been made for the design and implementation of CBT programs and the objective assessment of their efficacy. Previous (systematic) Table 2

Electronic links to open access scientific evidence reporting on CBT intervention outcomes in patients with FS – Original research

(Alda et al., 2011) http://www.ncbi.nlm.nih.gov/pmc/articles/PMC3308108/

(Amutio et al., 2015) http://www.ncbi.nlm.nih.gov/pmc/articles/PMC4290530/

(Ang et al., 2013) http://www.ncbi.nlm.nih.gov/pmc/articles/PMC4185390/

¹(Castel, Cascón, Padrol, Sala, & Rull, 2012) https://www.researchgate.net/publication/221785188_Multicomponent_Cognitive-Behavioral_Group_Therapy_With_Hypnosis_for_the_Treatment_of_Fibromyalgia_Long-Term_Outcome

(Castel, Salvat, Sala, & Rull, 2009) http://docplayer.net/10277181-Cognitive-behavioural-group-treatment-with-hypnosis-a-randomized-pilot-trial-in-fibromyalgia.html

(Comeche-Moreno et al., 2010) http://www.copmadrid.org/webcopm/publicaciones/clinica/cl2010v21n2a1.pdf

^{1, 2} (Davis & Zautra, 2013) https://www.researchgate.net/publication/236740014_An_Online_Mindfulness_Intervention_Targeting_ Socioemotional_Regulation_in_Fibromyalgia_Results_of_a_Randomized_Controlled_Trial

³(Degotardi, 2005) http://jpepsy.oxfordjournals.org/content/31/7/714.full.pdf+html

¹(García, Simón, Durán, Canceller, & Aneiros, 2006) https://www.researchgate.net/publication/6668435_Differential_efficacy_of_a_ cognitive-behavioral_intervention_versus_pharmacological_treatment_in_the_management_of_fibromyalgic_syndrome

²(González-Ramírez & Landero-Hernández, 2010) http://rcientificas.uninorte.edu.co/index.php/psicologia/article/view/204/5376

^{1, 2, 4}(Jelin, Granum, & Eide, 2012) https://www.researchgate.net/publication/221838717_Experiences_of_a_Web-Based_Nursing_ Intervention_-_Interviews_with_Women_with_Chronic_Musculoskeletal_Pain

¹(Martínez et al., 2014) https://www.researchgate.net/publication/237070336_Cognitive-behavioral_therapy_for_insomnia_and_sleep_ hygiene_in_fibromyalgia_A_randomized_controlled_trial

²(Menga et al., 2014) http://www.ncbi.nlm.nih.gov/pmc/articles/PMC4171792/

⁵(Rodero, García-Campayo, Casanueva-Fernández, & Sobradiel, 2008) http://actaspsiquiatria.es/repositorio/9/52/ENG/9-52-ENG-223-226-765269.pdf

(Sánchez et al., 2012) http://www.redalyc.org/pdf/337/33723038003.pdf

¹Available through cost-free subscription to ResearchGate network (https://www.researchgate.net); ²Web-based CBT intervention; ³Juvenile FS patients; ⁴Qualitative outcome assessment; ⁵Content in Spanish. Links were last accessed the 26th of May, 2016

Note. Links were last accessed the 26th of May, 2016. ¹ Available through cost-free subscription to ResearchGate network (https://www. researchgate.net). ²Web-based CBT intervention. ³ Juvenile FS patients. ⁴ Qualitative outcome assessment. ⁵ Content in Spanish.

reviews and meta-analyses have been performed summarizing and comparing study designs, samples, measures, interventions and outcomes, (see Appendix 1)yet accessing such content is not always easy. This review aimed at providing readers with a useful and practical summary of open access scientific information regarding the specific content of CBT interventions for patients with FS. A total of 59 publications were found which reported on CBT interventions in groups including exclusively FS patients; only 15 were open access. The number of citations related to the topic has increased over the years, which can be attributed to a growing interest in CBT and FS but also to the interest of clinical and research professionals to team up to produce and share scientific evidence. It was found that most publications originated from Spain and the United States, though a couple was found from Latin America, specifically Mexico and Brazil. Although most publications are in English, having identified some working groups in Spain and Mexico

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might encourage the Spanish speaking public, scientific or lay, to search further for material available in their language. In a smaller number, research on juvenile samples was found; although, FS is more common in middle aged adults. Young FS patients can also benefit from tailored CBT programs. Quantitative assessments outnumber qualitative ones, yet both perspectives offer invaluable information on the experience of CBT in FS patients. Most studies rely on a pre/post design, with or without follow-up, and it is a common practice to perform a pilot study and report on its initials findings.

There is a considerable amount of information on CBT interventions for patients with FS, yet most of this information is restricted. Here, a list of open access publications is provided so that all those somehow related to the FS (e.g. physicians, psychologists, nurses, patients) can have a starting point to learn about how CBT is a very useful tool to connect the psychological and the physical aspects in patients. Content can also be useful to those interested on going a step further, pursuing the design and implementation of individual or group therapy programs for clinical practice and/ or research.

Conclusions

CBT's for patients with FS, adult or juvenile, are important interventions in the management of this condition. Empirical evidence is ample,; yet, mostly restricted. Here, we present a list of open access scientific sources in order to promote interest in the topic and to encourage the design, implementation and evaluation of CBT interventions. Further research must proceed in the benefit of patients, the health system and science.

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Appendix

Apendix 1

Electronic links to open access scientific evidence reporting on CBT intervention outcomes in patients with FS – Complementary

Reviews / Meta-analysis

(Bennett & Nelson, 2006) http://www.myalgia.com/PDF%20files/Bennett%20Nelson%20CBT%202006.pdf

(Bernardy, Fuber, Kollner, & Hauser, 2010) https://www.researchgate.net/publication/45494025_Efficacy_of_Cognitive-Behavioral_Therapies_in_Fibromyalgia_Syndrome_-_A_Systematic_Review_and_Metaanalysis_of_Randomized_Controlled_Trials

(Bernardy et al., 2013) http://onlinelibrary.wiley.com/doi/10.1002/14651858.CD009796.pub2/full#CD009796-sec1-0001

¹(Lauche et al., 2013) https://www.researchgate.net/publication/257890190_A_systematic_review_and_meta-analysis_ of_mindfulness-based_stress_reduction_for_the_fibromyalgia_syndrome

(Minelli & Vaona, 2012) http://reumatismo.org/index.php/reuma/article/viewFile/reumatismo.2012.151/pdf

(Theadom et al., 2015) http://onlinelibrary.wiley.com/doi/10.1002/14651858.CD001980.pub3/full

(Thieme, Flor, & Turk, 2006) http://arthritis-research.biomedcentral.com/articles/10.1186/ar2010

(Thieme & Turk, 2012) http://www.reumatismo.org/index.php/reuma/article/view/reumatismo.2012.275/pdf

(van Koulil et al., 2007) http://www.ncbi.nlm.nih.gov/pmc/articles/PMC1954607/pdf/571.pdf

Thesis/Dissertation

(Gritzner, 2011) http://commons.pacificu.edu/spp/233

(Langford, 2008) https://ecommons.usask.ca/bitstream/handle/10388/etd-07232008-113842/langford_m. pdf?sequence=1&isAllowed=y

Note. Links were last accessed the 26th of May, 2016. ¹Available through cost-free subscription to ResearchGate network (https://www.researchgate.net)