A Bibliometric Approach to Research Into Fibromyalgia

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Objectives: To obtain a general and updated vision of research into fibromyalgia for its better understanding. A productivity analysis (by year and author) and a content analysis (by journal and topic) was carried out. Methodology: The computer databases of the Web of Knowledge: Science Citation Index Expanded (SCI-EXPANDED) and Social Sciences Citation Index (SSCI) of the Web of Science were used. For the exploration of the topics a content analysis was done on a sample of 356 articles, obtained using a simple randomized procedure based on a random number table (risk α =.05; z_{α} =1.96 \approx 2; p=q =0.5) with a sample error of 5%, with the purpose of reaching some general conclusions in the study of this topic area. Results and conclusions: Overall, 3201 article references have been obtained. In the years 1998 to 2003 the production of scientific articles increased considerably (43.5%) in comparison to the previous period, from 1992 to 1997 (27.4%). In the past decade (1996-2005) the production of articles tripled with respect to the first fifteen years (1980-1995). There is an overall 306 different journals that publish on this subject. The topic area that received most attention has been the study of the physiopathologic mechanisms (31.01%). There is an increment in the articles of psychiatry and psycology in the last few years.

Key words: Fybromyalgia. Scientific production. Bibliometric analysis.

Un acercamiento bibliométrico a la investigación en fibromialgia

Objetivos: Obtener una visión genérica actualizada de la investigación en fibromialgia para entenderla mejor. Se presenta un análisis de productividad (por años y autores) y un análisis de materias (por revistas y áreas temáticas). **Método:** Se ha utilizado las bases computarizadas de la Web of Knowledge: Science Citation Index Expanded (SCI-EXPANDED) y Social Sciences Citation Index (SSCI) de la Web of Science. Para la exploración de las áreas temáticas se ha realizado un análisis de contenido de una muestra de 356 artículos, obtenida por medio del procedimiento aleatorio simple, utilizando la tabla de números al azar (riesgo $\alpha = 0,05$; $z_{\alpha} = 1,96 \approx 2$; p = q = 0,5) con un error muestral del 5%, con el propósito de llegar a algunas conclusiones generalizables del estudio de este campo temático.

Resultados y conclusiones: Se ha obtenido un total de 3.201 referencias de artículos. En el período 1998-2003 aumenta de forma considerable la producción de artículos (43,5%) con respecto al período anterior 1992-1997 (27,4%). En la última década (1996-2005) se triplica la producción de artículos con respecto a los primeros 15 años (1980-1995). Hay un total de 306 revistas diferentes que publican sobre esta área. El área temática más trabajada ha sido el estudio de los potenciales mecanismos fisiopatológicos (31,01%). Hay un incremento de artículos de psiquiatría y psicología en los últimos años.

Palabras clave: Fibromialgia. Producción científica. Análisis bibliométrico.

Introducción

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The study of chronic illness has turned into a challenge for developed countries, especially due to the increment in their prevalence and the epidemiological findings concerning their multicausal origin that emphasize the relevance of a biopsicosocial perspective of health.¹

We have chosen to study the field of rheumatology because its diseases are among the most frequent human illnesses² and we have concretely centered on the fibromyalgia syndrome (FM) because it is a chronic illness, without a clear organic cause, common in the daily practice, manifested mainly by pain and fatigue; patients with this disease have a high index of health system usage and a deterioration in both labor and family relatinships.³ FM is a chronic rheumatic syndrome that is associated with a high degree of anxiety and an unknown etiology. Patients present with generalized osteomuscular pain and multiple pain triggering points with a characteristic localization. A degenerative or inflammatory process cannot explain pain and there is no treatment today that reduces all symptoms in all patients. With the exception of Hugh Smythe,⁴ who reviews the evolution of knowledge on the syndrome up until 1986, and more recently Goldenberg et al,⁵ there has not been an ample, comprehensive follow-up of how its study has evolved in the different fields through a bibliometric review, making it necessary to obtain a generic and up-to-date vision of this field to better understand it. Within the clinical psychology and chronic-pain related health research circle,⁶ our study tried, as a general objective of the bibliometric analysis done, to study the evolution of the research work done made on FM. This general objective was accomplished in two parts: a) productivity analysis: yearly production and the productivity of the authors was analyzed; in relation to this, their study topics were studied as well as the journals in which they published; and b topic analysis: centered both on the study of the most productive journals as well as the thematic analysis of a sample of the studies published during the past 26 years.

Methods

In order to cover our objectives we have done a bibliometric study during the 1980-2005 period. This review and analysis of the existing medical literature was done on the Web of Knowledge: Science Citation Index Expanded (SCI-EXPANDED) and the Social Sciences Citation Index (SSCI) of the Web of Science databases,⁷ allowing for an objective analysis of the literature. A review of the previous 26 years was carried out because the database does not extend beyond 1981 when the first publications began to be registered. The decision to look for the term *fibromyalgia* only in the title was taken, permitting more precision with respect to the content of the document. A total 3201 article references were obtained. If the search had been carried out in the fields marked as "key words" the search would have been more exhaustive, but when a larger number of terms are used, the possibility of background noise increases, as does the inclusion of "parasite" terms that would disorient the planned purposes of our study. For the analysis of each topic we selected a sample of 356 articles, through a simple randomized procedure, using a random number table (risk α =.05; z_{α} =1.96 ~2; p=q=0.5), with a sample error of 5% with the objective of insuring the representativity of the extracted sample as well as the feasibility of reaching some generic conclusions of the study of this topical field. From reading the sample abstracts we have been allowed to do the following classification according to topical areas:

- Articles with a psychological topic: in this category we included theoretical studies, review or empirical studies of psychological barriers that could be related to this disease.

1. Articles relating to intervention: in this category we included articles related with the application of some psychological treatment to a group of patients with the disease, comparing them to other rheumatic disease patients. We also included studies centered on intervention programs or the evaluation of the treatments employed.

2. Articles related to conceptual and methodological aspects: included in this category were publications unrelated to the aspects of intervention.

a) Articles that searched for explanations of a psychopathological type: in this category we included studies directed to the evaluation of psychopathological alterations related to fibromyalgia.

b) Articles related to psychosocial factors: in this category we included studies that were characterized for including cognitive, behavioral or social variables that play a relevant role in adapting to the disease.

c) Articles related to the quality of life: included in this category were studies that evaluated the alteration in the functional, psychological and social capacities of patients with the disease.

d) Instruments of evaluation: included in this category were the studies that approached aspects related to the validity and trustworthiness of the instruments employed for the evaluation and diagnosis of the patients with FM.

- Articles with a biomedical topic: included in this category were theoretical studies, review or empirical studies of the biomedical variables that could be related to the etiology, epidemiology, social and economic cost and the criteria for disease classification.

1. Articles related to intervention; in this category we included all of the studies characterized by the application of some form of pharmacological treatment and/or alternative therapies to a group of patients with the disease

or in comparison to the other patients with rheumatic disease.

2. Articles related to conceptual and methodological aspects: included in this category are studies unrelated to aspects of medical or psychological intervention.

a) Epidemiology. In this category we included studies that are centered on studying disease in the population, as well as the related conditions, causes and circumstances to which they are attributed in the distinct social groups.

b) Etiologic factors. In this category we included studies that are characterized by the analysis of the organic causes that could trigger the disease.

c) Concept, definition and diagnostic criteria. In this category we included studies that have as an objective the grouping and definition of the disease diagnostic criteria.

d) Social and economical costs. This category included studies analyzing the social and economic impact on health services that the majority of patients with the disease demand.

e) Unclassified articles. Including all articles that were not classified in the previous categories.

Results

Productivity analysis

In the past few years there has been a growing interest for the topic according to the number of published articles, in comparison to previous years (Figure).

Author Productivity

A total 6509 authors appear in the review, presenting important differences in their productivity. Thus, 75.31% of the authors have published only one article. Facing this proportion there is a small number of authors who have published 10 or more articles. This distribution is close to the distribution of Lotka in 1926⁸ and coincides with other scientific productivity studies9 that shows, in general, a small number of productive persons, opposed to a great mass that have few publications. Among the most productive there is one author (Clauw) that stands out for having published the largest amount of papers (102 studies) in a small period of time (1999-2004), original articles in most cases. Most of the articles are published in: Arthritis and Rheumatism and Journal of Rheumatology. His scientific interests are fibromyalgia, pain, Gulf-war syndrome and stress. Wolfe et al has published 84 papers since 1986. His interests are centered in questionnaires, pain, physiopathology, associated psychological

problems, rheumatic diseases and osteoarthritis, according to the descriptors most used in his publications. His most relevant contributions have been, in first place, the establishment of the official criteria for FM, developed by the American College of Rheumatology (ACR),¹⁰ and secondly, epidemiologic studies.¹¹ Since 2000, his contributions decreased in number. Goldenberg has published 64 articles. The journals in which he has published are: Arthritis and Rheumatism, with 21 articles; Journal of Rheumatology (12 articles). His line of investigation is centered on the diagnostic criteria and the pathogeny, concentrating his interest in the description of the symptoms that are superimposed on the diagnosis of FM, chronic fatigue syndrome and the myofacial syndrome. He dedicates 12 articles to reviewing the possible association between FM and psychological alterations. Of these we must point out the research that tries to explain the relationship between depression and illness.¹² When analyzing in a general form the contributions of the most productive authors, we realize that their interests have centered overall on physiopathology and the diagnostic criteria and disease classification, from a biopsychosocial perspective. All of them publish in rheumatology journals and most of the research is done in the United States, Canada, Germany, and Israel.

Topic Analysis

Analysis of the Most Productive Journals

In Table 1 we present the journals in relation to their productivity. The total umber of journals that publish in this area ascends to 306, with important differences among

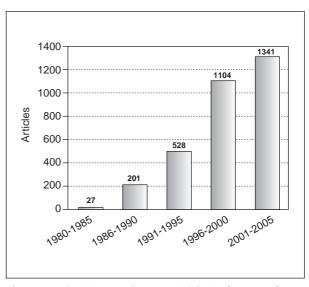


Figure 1. Productivity according to period during (1980-2005).

them. The total number of journals that publish research in this area is 306, with important differences between them. The most productive journals are *Arthritis and Rheumatism, Journal of Rheumatology,* and *Annals of the Rheumatic Diseases.*

These journals could be classified into 6 topical areas: rheumatology, psychiatry, internal and general medicine, clinical neurology, neuroscience and rehabilitation (Table 2). There is a notable increase in articles in the psychiatry category, especially in the following journals: *Sleep* (28 articles), *Journal of Psychosomatic Research* (27), *Psychosomatic Medicine* (17) and clinical neurology: in *Pain* (36 articles), *Sleep* (28), *European Journal of Pain* (12).

Analysis of Topical Areas

The qualitative analysis of the selected sample (n=356) revealed a clear division between two types of studies: on one hand those that are centered on a biomedical theme and on the other the ones that are dedicated to a psychological topic.

Etiologic Factors

This thematic category constitutes the most productive area. Several factors have been implicated in the genesis of FM, without a concrete etiology having been identified.¹³ Attention has been paid to the association with identifiable vital events such as a bacterial or viral infection, post-traumatic stress, the birth of a child and FM, though a triggering factor has not been identified in all of the patients. There are other studies that find evidence of a hereditary pattern, indicating that FM is transmitted through the females of the family. More recently, studies have centered on the abnormalities present in peripheral tissue, alterations in sleep, abnormal muscle metabolism, alterations in the immune system and abnormalities in the neurotransmitter serotonin. An increase in the nervous system pain producing substance has also been observed (substance P). Other research has pointed at FM as a "stress related syndrome."

Classification Factors

Between 1980 and 1990 several groups of criteria have been proposed for the diagnosis of FM.¹⁴⁻¹⁶ We found that Wolfe dedicates 21 articles to this topic and reviews the different contributions to the diagnostic criteria that characterize this group of patients.^{17,18} The ACR, for the definition of FM classification criteria,¹⁰ proposes to suppress the distinction between primary and secondary FM and different levels of diagnosis because they are not distinguishable in the studies that analyze these variables. The criteria proposed by the ACR and recognized worldwide are: generalized pain in combination with tenderness in 11 of 18 specific tender points. These criteria were designed for the classification of illness and not their diagnosis.

TABLE 1. Scientific Publications With the Most Productivity

| No. | Percentage of 2000 | |
|-----|---|--|
| 434 | 21.7 | |
| 204 | 10.2 | |
| 125 | 6.2 | |
| 87 | 4.3 | |
| 45 | 2.2 | |
| 38 | 1.9 | |
| 34 | 1.7 | |
| 33 | 1.7 | |
| 29 | 1.5 | |
| 28 | 1.4 | |
| | 434 204 125 87 45 38 34 33 29 | |

| TABLE 2. Number of Articles Published During the Period of | of 1980-2005 According to Topic* |
|--|----------------------------------|
|--|----------------------------------|

| | 1980-1985 | 1986-1991 | 1992-1997 | 1998-2003 | 2004-2005 | Total |
|-------------------------------|-----------|-----------|-----------|-----------|-----------|-------|
| Rheumatology | 10 | 112 | 339 | 303 | 304 | 956 |
| Psychiatry | | 10 | 19 | 44 | 62 | 125 |
| Internal and general medicine | 10 | 64 | 49 | 32 | 27 | 118 |
| Clinical neurology | | 17 | | 46 | 56 | 102 |
| Neuroscience | | 14 | 23 | 33 | 38 | 94 |
| Rehabilitation | | 14 | 42 | 34 | 14 | 90 |

*SCI-EXPANDED, SSCI databases.

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Epidemiology

The epidemiological perspective analyzes the estimations of prevalence of FM, and differs according to each population. Data, according to the international literature, comes from three different sources: communities (population based studies), reference centers (hospitals and clinics), and clinics specializing in rheumatology. The first references of community studies appear published by Jacobsson et al,¹⁹ and the most recent study published in our country is the EPISER study,²⁰ where FM prevalence in the adult Spanish population is estimated to be 2.4%, representing 700 000 affected persons older than 20 years. The sociodemographic and clinical characteristics of this type of patients is usually a female:male ratio of 21:1, and both the infant population as well as the adolescent population can be affected, though in them the female population is not as affected,²¹ with a peak in prevalence around 40 and 49 years, with a greater frequency in the rural areas, a low sociocultural level, chronic illnesses as comorbidities and affecting in a very significant manner the functional capacity and the quality of life, independent of gender, age, study level and the presence of comorbidities. Patients tend to have a mean duration of pain between 6 and 12 years and a history of therapeutic failure concerning their main symptom.²²

Socioeconomic Cost

Previous studies in other populations have calculated an annual cost for FM of \$2274 dollars per patient, with a mean number of yearly visits to the physician of 9 to 12 and 1 hospitalization every 3 years. Compared with other rheumatic disease, they undergo a larger number of surgical interventions (carpal tunnel syndrome, cervical spine, gynecologic, abdominal, etc), a larger number of allergies, anxiety, and depressive syndromes and hypertension.23 Regarding the pharmaceutical cost, it must be considered that these patients use an elevated number of drugs that have not always proven effective.²⁰ In different studies it is demonstrated that these patients consult different specialists and use a larger amount of alternative therapies for their problem, therefore generating larger work absenteeism costs per patient and demand more workers pensions due to dyscapacity.23,24

Articles Relating to Pharmacological Intervention

Pharmacologic treatments constitute another large topic of research. A great number of articles are dedicated to the use of antidepressants, centering on trycyclics (TCA) and the inhibitors of serotonin uptake or combinations of both (fluoxetin and amytriptiline), together with the therapeutic use of analgesics or corticotropin. Nonetheless, the studies point out that there is no current treatment that leads to a cure. O'Malley et al²⁵ examine the efficacy of antidepressants treating many of the symptoms of FM with or without depression. Patients informed of moderate reductions in individual symptoms, mainly pain, and mention that a future study topic would be to differentiate if this effect is independent of the effect on depression.

Relationship Between FM and Psychological Alterations

Different studies examine the relationship between FM and depression. The results indicate that a larger prevalence of affective disorders in the family and FM could be related.²⁶ Commonly it is depression which precedes symptoms of FM (in 50% to 70% of cases).^{27,28} The results of the psychological variables show elevated punctuations in the indexes relating to hypochondria, hysteria, and post-traumatic stress compared with other patients with chronic pain, depression and a psychosomatic profile in 80% of the patients and mental disease in 11%.²⁹ The identification of subgroups in the psychological analysis illustrates the heterogeneity of FM.³⁰ Psychological problems do not have to be an integral part of the syndrome nor do they occur always and in the same way in all patients.³¹

Articles Related to Psychosocial Factors

Regarding cognitive, social and behavioral variables that relate to adaptation to illness, topics such as belief in control,³² social support,³³ relationship to strategies for coping with pain,³⁴ or depression³⁵, in comparison to other rheumatic patients or the psychological factor analysis as well as the experience with pain of patients with FM, were analyzed with the intention of including them in treatment programs. With respect to these variables, we have proved their efficacy in the modulation of pain and their study in the evolution of knowledge of the disease started in the 1990s.³¹

Articles Relating to Health-Related Quality of Life

Studies on health-related quality of life thread on how FM modifies the perceived quality of life in the physical, psychological, and social spheres. The increase in the number of studies in this category has presented itself in the past 2 decades; nonetheless, in the analyzed sample they represent a lesser percentage of studies (0.56%). The results of the studies³⁶ show that FM has a negative impact on the quality of life of these patients. Other comparative studies reveal that quality of life in patients with FM in similar to that in rheumatoid arthritis.³⁷

Articles Relating to Evaluation Instruments

In this area we describe the instruments that have been used to obtain a psychological and psychosocial profile of the patients with FM. Topics such as attitudes, perception, and belief in control as well as coping strategies, pain conduct, functional limitation, emotional and psychopathological variables, quality of life, and multidimensional instruments were analyzed. In this last question, instruments such as the West Heaven-Yale (WHY-MPI) multidimensional pain inventory and the McGill-Melzack Pain Questionnaire (MPQ),³⁸ were used with the object of identifying patient profiles for chronic pain.

Articles Relating to Psychological Intervention

All of these studies give a great deal of importance to the psychological intervention on these patients. Excepting one article that deals with hypnosis,³⁹ the rest employ cognitive-behavioral therapies and physical exercise. Tendencies in this type of intervention are the use of multimodality treatment parting from a biopsychosocial approximation,⁴⁰ including health education,⁴¹ in which a good efficacy is demonstrated for this component in comparison with the cognitive intervention.

Discussion

A topical and productivity analysis has been used to obtain a general vision of the evolution of the work done on FM. This analysis can be useful to know the evolution of knowledge with respect to the disease in different fields, as well as the groups of investigators that produce and use the literature.

In the database used (Web Knowledge), in comparison with other bibliographic products (MEDLINE, PsycInfo, Scielo, Bireme, etc), the nucleus of the system is formed by bibliographic references and cited references (a citation index is a perfect topical index). This database does not contain full-text documents but facilitates access to the most important articles, book reviews and thematic reviews, database reviews, debates, editorials, meeting summaries, etc, of a scientific discipline through reliable sources of information, purveying the user with tools for analysis or evaluation and information management. Nonetheless, the majority of papers published are of Anglo-Saxon origin, making the title, abstract and key words available in English, excluding other publications in Spanish, so future analysis must complete this study. Other relevant databases such as The Cochrane Library are centered in systematic reviews of clinically controlled studies (CCS) on the effects of public health assistance, but our objective did not center on a particular work area but rather on the current tendencies in the different lines of work that can be generally identified (including CCS), such as the introduction to the content as an introduction to content that represents the term, and through this analysis, to compare the results with other databases. When carrying out the search in the Cochrane database with the same criteria, we found 335 articles; in MEDLINE, 2186; and in PsycInfo, 548. Therefore, the strategy used seems more extensive.

A limitation of this study is that the studies analyzed of the extracted sample are not homogeneous, but constitute important categories for research, making it possible for some of the strata were more or less represented than what is proportional due to the sample (for example, we found no publications on alternative therapies). In this case, sampling should continue until 356 patients are included into each group. This notwithstanding, the results (though they must be interpreted with caution due to the limitations that are part of databases, basically the extensiveness of the indexation) and the classification criteria of the sample employed are concordant with the previous literature,⁵ that shows the current tendency in the knowledge of the disease and is described in the following lines. The total number of published articles on FM during the 1980-2005 period in the Web of Knowledge is 3201 articles.

In the past decade (1996-2005) the production of articles has tripled with respect to the first 15 years (1980-1995). We have observed that, starting in 1989 the number of articles about this syndrome increased, in consonance with a larger knowledge on the disease and the inclusion of psychosocial factors in their maintenance. This data is related with the impact that the recognition of this rheumatic disease has had upon the scientific community and in the interest of attaining a larger comprehension of this disease from rheumatology's different perspectives.

There is a total 306 different journals that publish on this area. The journal with the most articles published on FM with respect to the total is the official ACR journal: *Arthritis and Rheumatism* (434 articles).

In the sample selected, the topics in which most articles have been published is the one of conceptual, aspects and methodology, mainly etiologic considerations, classification and diagnosis. Publications on medical interventions (13.41%) have centered on pharmacologic treatment. Most research has been carried out on trycyclic antidepressants and serotonin uptake inhibitors. Publications on psychological intervention programs (5.87%) have been scarce and complimentary, centered on cognitive-behavioral techniques and exercise.

We have found an increase in the number of articles in the category of psychiatry and psychology in the past years, associated to a shift in paradigms,⁴² that takes into account the psychosocial perspective^{43,44} as an approximation to the consideration of the main symptoms of the disease.

We do not interpret this as a psychiatric, social or neurological disease or is or not the part of the field of rheumatology. We believe that it is necessary to attain a group understanding of the different topical areas that contribute with knowledge and promotes collaboration in the treatment of pain by several specialties, to attain the integrative attention of patients, making it necessary to analyze it through an interdisciplinary approach. This concept is, at the same time, the basis of the modern strategy for the diagnosis and treatment of the chronic pain syndromes.

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