

FREQUENCY OF RHEUMATIC DISEASES IN PORTUGAL: A SYSTEMATIC REVIEW

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Abstract

Objectives: To describe the frequency of rheumatic diseases in Portugal through a systematic review of published literature, critically appraising available information and identifying data collection gaps.

Methods: We systematically reviewed the literature to retrieve data on the occurrence of rheumatic diseases in Portugal through MEDLINE and *Índex das Revistas Médicas Portuguesas* searches, PhD theses, and national health surveys reports. Original articles in English or Portuguese published between 1 January 2000 and 31 December 2010 were included.

Results: We retrieved information for the prevalence of rheumatic diseases, osteoarthritis, back pain, work-related musculoskeletal disorders (WRMDs), osteoporosis, fibromyalgia, rheumatoid arthritis, spondyloarthritis and other systemic rheumatic diseases and for the incidence of back pain, osteoporotic fracture and other systemic rheumatic diseases.

The prevalence of rheumatic diseases ranged from 16.0% to 24.0% and the prevalence of osteoarthritis was 11.1% (95% confidence intervals (95%CI): 9.4-13.1) in the knee and 5.5% (95%CI: 4.3-7.0) in the hip. Regarding back pain, period prevalence ranged from 8.0% (95%CI: 6.1-10.1) to 29.5% (95%CI: 23.4-36.2) in children and from 12.3% (95%CI: 10.5-14.3) to 51.3% (95%CI: 48.6-53.9) in adults. The prevalence of WRMDs ranged from 5.9% to 84.2% (95%CI: 80.8-87.3). The yearly incidence of osteoporotic fracture (per 100 000) ranged from 93.3 to 481 (95%CI: 407-564) in women and from 31.9 to 154 (95%CI: 106-218) in men. The prevalence of osteoporosis in women ranged from 11.0% to 15.4% (95%CI: 13.4-17.6) and in men from 1.1% to 16.8% (95%CI: 12.2-22.3). The prevalence of

fibromyalgia ranged from 3.6% (95%CI: 2.0-5.2) to 3.7% (95%CI: 2.0-5.4). The prevalence estimates of ankylosing spondylitis and of spondyloarthritis were 0.6% and 1.6% (95%CI: 0.8-2.7), respectively. The prevalence of systemic lupus erythematosus was estimated in 0.2% (95%CI: 0.1-0.8).

Conclusions: There is a broad spectrum of information available that indicates an important burden of rheumatic diseases in the general Portuguese population. Gaps were identified particularly regarding inflammatory arthropathies and other systemic rheumatic diseases.

Keywords: Rheumatic Diseases; Burden of Disease; Frequency; Prevalence; Incidence; Portugal.

Introduction

Rheumatic diseases are conditions and functional disorders of the musculoskeletal system of non-traumatic causes. These diseases are a diverse group of conditions with regard to pathophysiology which are linked not only by the musculoskeletal structures affected but by their association with pain and consequent reduction in the range of motion and function^{1,2}. They encompass a wide spectrum of conditions, from those of acute onset and short duration to chronic progressive course disorders including osteoarthritis, rheumatoid arthritis, osteoporosis and low back pain³.

Rheumatic conditions are a major cause of morbidity throughout the world^{3,4}. They represent an important social and economic problem inflicting enormous costs on health and social care systems^{4,5}. They cause more pain and disability than any other group of conditions with consequent organ dysfunction, limitation in activity and restriction in social participation⁴. In Europe, nearly one quarter of adults are affected by longstanding musculoskeletal problems that limit daily activities⁶. Although affecting all ages, rheumatic diseases become increasingly common with ageing. Consider-

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ring the increasing number of old people throughout the world and the observed trends in lifestyles, which promote the occurrence of musculoskeletal disorders, the burden of these conditions on public health tends to rise^{4,6}.

Individual and population impact of rheumatic diseases is certainly high in Portugal according to estimates obtained in other developed societies. However, it is important to systematically collect indicators of the current burden of musculoskeletal conditions in our population, in order to monitor trends and guide health policies⁷. Although in Portugal, narrative reviews on the frequency of rheumatic diseases, individually or as a group, have been published^{5,8-10}, we believe that the knowledge about these conditions will benefit from a systematic approach.

We were interested in original studies estimating the prevalence and/or incidence of rheumatic diseases targeted by the *National Program against Rheumatic Diseases* because of their predicted impact at population level, either related to their high frequency or to their strong association with adverse health outcomes^{2,11}. These conditions are osteoarthritis, back pain, periarticular rheumatic diseases, including work-related musculoskeletal disorders, osteoporosis and osteoporotic fracture, fibromyalgia, microcrystal-related arthropathies, rheumatoid arthritis, spondyloarthritis, other systemic rheumatic diseases and pediatric rheumatic diseases.

Objectives

We aimed to systematically review published scientific literature on the frequency of rheumatic diseases in Portugal, to critically appraise the available information and to identify the gaps in data collection.

Methods

Following the 2009 Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines¹², data on the occurrence of rheumatic diseases in Portugal were obtained through MEDLINE and *Índex das Revistas Médicas Portuguesas* (Index RMP) searches using the expressions presented in Table I (Date of last search: 30th August 2011). We also reviewed PhD theses in the Index

RMP bibliographic database, periodic general health surveys such as the *4th National Health Survey* and publications including information on rheumatic diseases. In addition to this strategy, we also performed a cross references search and a full manual search in the journals *Acta Reumatológica Portuguesa*, *Acta Médica Portuguesa*, *Revista Portuguesa de Saúde Pública*, *Revista Portuguesa de Clínica Geral* and *Arquivos de Medicina*. Since a review on the studies before 2000 estimating the prevalence of rheumatic diseases in Portugal was published in 2002⁵ and to optimize the relation between the sensitivity desired and the resources available, original articles in English or Portuguese published between 1 January 2000 and 31 December 2010 were included.

For rheumatic diseases, 1 049 articles were retrieved by MEDLINE and Index RMP bibliographic databases. We identified 170 articles for osteoarthritis, 261 articles for back pain, 86 articles for periarticular rheumatic diseases, 260 articles for work-related musculoskeletal disorders, 345 articles for osteoporosis and osteoporotic fracture, 32 articles for fibromyalgia, 66 articles for microcrystal-related arthropathies, 200 articles for rheumatoid arthritis, 158 articles for spondyloarthritis, 853 articles for other systemic rheumatic diseases and 52 articles for pediatric rheumatic diseases.

Two reviewers (TM and RL) screened all the references. After reviewing the titles and abstracts and/or the full articles, we excluded those that were clearly unrelated to the study subject, (e.g. studies not estimating prevalence or incidence of the condition of interest) or that had a sampling strategy which did not include a random or consecutive sample of Portuguese subjects).

For rheumatic diseases that almost inevitably lead to hospitalization or medical appointments, such as osteoporotic fracture and juvenile idiopathic arthritis, studies estimating prevalence and/or incidence based on hospital records were also included.

Through the MEDLINE and Index RMP searches, and after exclusions were made, two studies were included for rheumatic diseases^{13,14}, one study for osteoarthritis¹³, five studies for back pain^{13,15-18}, two studies for work-related musculoskeletal disorders^{19,20}, seven studies for osteoporosis and osteoporotic fracture^{13,21-26}, one study for fibromyalgia²⁷, two studies for rheumatoid arthritis^{13,28}, two studies for spondyloarthritis^{13,29}, three studies for other systemic rheumatic diseases^{13,28,30} and one

Table I. Search expressions and systematic review results about rheumatic diseases occurrence

Diseases	Bibliographic database	Search expression	Number of results after duplicates removed	Excluded articles by title and abstract	Number of included articles and references
Rheumatic diseases	Medline	Publication Date - from 2000/1/01 to 2010/12/31 : (musculoskeletal disease OR musculoskeletal disorder OR rheumatic diseases OR skin and connective tissue diseases OR calcium metabolism disorder) AND (Portugal OR Portuguese) AND (prevalence OR incidence OR occurrence OR frequency)			
	Index RMP	Articles and PhD theses from 2000 to 2010: doença AND reumática AND prevalência doença AND reumática AND incidência patologia AND reumática AND prevalência patologia AND reumática AND incidência patologia AND músculo AND prevalência patologia AND músculo AND incidência patologia AND osteoarticular AND prevalência patologia AND osteoarticular AND incidência	1 049	Not incidence/prevalence studies of rheumatic diseases: n=1 041 Not Portuguese general population based studies: n=6	2 ^{13,14}
Osteoarthritis	Medline	Publication Date - from 2000/1/01 to 2010/12/31 : (arthritis OR osteoarthritis OR arthrosis OR osteoarthritis) AND (Portugal OR Portuguese) AND (prevalence OR incidence OR occurrence OR frequency)			
	Index RMP	Articles and PhD theses from 2000 to 2010: osteoarrose AND prevalência osteoarrose AND incidência arrose AND prevalência arrose AND incidência	170	Not incidence/prevalence studies of osteoarthritis: n=167	1 ¹³
Back pain	Medline	Publication Date - from 2000/1/01 to 2010/12/31 : (back pain OR back ache) AND (Portugal OR Portuguese) AND (prevalence OR incidence OR occurrence OR frequency)			
	Index RMP	Articles and PhD theses from 2000 to 2010: raquialgia AND prevalência	261	Not incidence/prevalence studies of back pain: n=251 Not Portuguese general population based studies: n=5	5 ^{13,15-18}

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Table I. (continuation)

Diseases	Bibliographic database	Search expression	Number of results after duplicates removed	Excluded articles by title and abstract	Number of included articles and references
	Medline	raquialgia AND incidência cervicalgia AND prevalência cervicalgia AND incidência dor AND cervical AND prevalência dor AND cervical AND incidência lombalgia AND prevalência lombalgia AND incidência dor AND lombar AND prevalência dor AND lombar AND incidência			0
Periarticular rheumatic diseases	Index RMP	Publication Date - from 2000/1/01 to 2010/12/31: (soft tissue injury OR bursitis OR tendinitis OR tenosynovitis OR enthesopathy OR capsulitis) AND (Portugal OR Portuguese) AND (prevalence OR incidence OR occurrence OR frequency) Articles and PhD theses from 2000 to 2010: patologia AND periarticular AND incidência tendinite AND prevalence tendinite AND incidência tenossinovite AND prevalence entesite AND incidence entesite AND incidence bursite AND prevalence bursite AND incidence capsulite AND prevalence capsulite AND incidence ligamentite AND prevalence ligamentite AND incidence	86	Not incidence/prevalence studies of periarticular rheumatic diseases: n=85 Not Portuguese general population based studies: n=1	

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Table I. (continuation)

Diseases	Bibliographic database	Search expression	Number of results after duplicates removed	Excluded articles by title and abstract	Number of included articles and references
Work-related musculoskeletal disorders	Medline	Publication Date - from 2000/1/01 to 2010/12/31: (occupational OR work) AND musculoskeletal disease AND (Portugal OR Portuguese) AND (prevalence OR incidence OR occurrence OR frequency)		Not incidence/prevalence studies of work-related musculoskeletal disorders: n=248	
	Index RMP	Articles and PhD theses from 2000 to 2010: reumatologia AND ocupacional AND prevalência reumatologia AND ocupacional AND incidência les* AND musculo AND prevalência les* AND musculo AND incidência LMERT AND prevalência LMERT AND incidência LMELT AND prevalência LMELT AND incidência	260	Not Portuguese general population based studies: n=10	2 ^{19,20}
Osteoporosis and osteoporotic fracture	Medline	Publication Date - from 2000/1/01 to 2010/12/31: (osteoporosis OR osteoporotic fracture) AND (Portugal OR Portuguese) AND (prevalence OR incidence OR occurrence OR frequency)		Not incidence/prevalence studies of osteoporosis and osteoporotic fracture: n=330	7 ^{13,21-26}
	Index RMP	Articles and PhD theses from 2000 to 2010: osteoporose AND prevalência osteoporose AND incidência fractura AND prevalência fractura AND incidência	345	Not Portuguese general population based studies: n=8	
Fibromyalgia	Medline	Publication Date - from 2000/1/01 to 2010/12/31: (fibromyalgia OR fibromyositis OR chronic fatigue syndrome) AND (Portugal OR Portuguese) AND (prevalence OR incidence OR occurrence OR frequency)		Not incidence/prevalence studies of fibromyalgia: n=30	
	Index RMP	Articles and PhD theses from 2000 to 2010: fibromialgia AND prevalência fibromialgia AND incidência síndrome fibromialgico AND prevalência síndrome fibromialgico AND incidência «fadiga crónica» AND prevalência «fadiga crónica» AND incidência	32	Not Portuguese general population based studies: n=1	1 ²⁷

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Table I. (continuation)

Diseases	Bibliographic database	Search expression	Number of results after duplicates removed	Excluded articles by title and abstract	Number of included articles and references
Microcrystal-related arthropathies	Medline Index RMP	Publication Date - from 2000/1/01 to 2010/12/31: (gout OR chondrocalcinosis OR crystal arthropath*) AND (Portugal OR Portuguese) AND (prevalence OR incidence OR occurrence OR frequency) Articles and PhD theses from 2000 to 2010: artrropatia AND prevalência artrropatia AND incidência gota AND prevalência gota AND incidência pirofosfato AND cálcio AND prevalência pirofosfato AND cálcio AND incidência	0	Not incidence/prevalence studies of microcrystal-related arthropathies; n=66	66
Rheumatoid arthritis	Medline Index RMP	Publication Date - from 2000/1/01 to 2010/12/31: rheumatoid arthritis AND (Portugal OR Portuguese) AND (prevalence OR incidence OR occurrence OR frequency) Articles and PhD theses from 2000 to 2010: artrrite AND prevalência artrrite AND incidência	200	Not incidence/prevalence studies of rheumatoid arthritis; n=198	2 ^{13,28}
Spondyloarthritis	Medline Index RMP	Publication Date - from 2000/1/01 to 2010/12/31: (spondyloarthropathies OR spondylarthropath*) OR psoriatic arthritis OR arthritic psoriasis OR ankylosing spondylitis OR bechterew disease OR reiter OR reactive arthritis OR postinfectious arthritis) AND (Portugal OR Portuguese) AND (prevalence OR incidence OR occurrence OR frequency) Articles and PhD theses from 2000 to 2010: espondil* AND prevalência espondil* AND incidência psoria* AND prevalência psoria* AND incidência sindrom* AND Reiter AND prevalência sindrom* AND Reiter AND incidência SAPHO AND prevalência SAPHO AND incidência	158	Not incidence/prevalence studies of spondyloarthritis; n=155	2 ^{13,29}

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Table I. (continuation)

Diseases	Bibliographic database	Search expression	Number of results after duplicates removed	Excluded articles by title and abstract	Number of included articles and references
Medline		Publication Date - from 2000/1/01 to 2010/12/31: (systemic rheumatic disease OR connective tissue diseases OR autoimmune diseases OR myositis OR vasculitis OR skin diseases, vascular) AND (Portugal OR Portuguese) AND (prevalence OR incidence OR occurrence OR frequency)			
Other systemic rheumatic diseases	Index RMP	Articles and PhD theses from 2000 to 2010: doença AND reumática AND sistêmica AND prevalência doença AND reumática AND sistêmica AND incidência doença AND tecido AND conjuntivo AND prevalência doença AND tecido AND conjuntivo AND incidência conectivite AND prevalence connectivite AND incidência doença AND autoimune AND prevalência doença AND autoimune AND incidência esclerose AND sistêmica AND prevalence esclerose AND sistêmica AND incidência polimiosite AND prevalence polimiosite AND incidence dermatomiosite AND prevalence dermatomiosite AND incidence vasculite AND prevalence vasculite AND incidence lúpus AND eritematoso AND sistêmico AND prevalence lúpus AND eritematoso AND sistêmico AND incidence Sjögren AND prevalence Sjögren AND incidence	853	Not incidence/prevalence studies of other systemic rheumatic diseases: n=850	3 ^{13,28,30}
Pediatric rheumatic diseases		Publication Date - from 2000/1/01 to 2010/12/31: (pediatric rheumatic diseases OR juvenile arthritis) AND (Portugal OR Portuguese) AND (prevalence OR incidence OR occurrence OR frequency)	52	Not incidence/prevalence studies of pediatric rheumatic diseases: n=51	1 ³¹

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Table I. (continuation)

Diseases	Bibliographic database	Search expression	Number of results after duplicates removed	Number of included articles and references
			Excluded articles by title and abstract	
	Index RMP	Articles and PhD theses from 2000 to 2010: artrite AND idiopática AND prevalência artrite AND idiopática AND incidência reumatismo AND juveni* AND prevalência reumatismo AND juveni* AND incidência		

study for pediatric rheumatic diseases³¹.

For their relevance in population-based measures and indicators we obtained also data from the *4th National Health Survey*, carried out by the National Health Institute Doutor Ricardo Jorge (INSA, IP) and Statistics Portugal (INE, IP)³², from the *ECOS project (Em Casa Observamos Saúde)*, an observation tool created by INSA, IP^{14,33}, and from the *Network of Sentinel Doctors*, a primary care surveillance system from the National Institute of Health based on a network of general practitioners^{18,34,35}.

Through the cross references search and full manual search we additionally identified two studies for back pain^{36,37} and seven studies for work-related musculoskeletal disorders³⁸⁻⁴⁴.

For each study, we extracted information on first author, year of publication, period of data collection, data collection method, case definition used, study population addressed, characteristics of the subjects evaluated (gender and age of the participants), sample size, participation/response rate and a summary of the estimates. Researchers were contact directly when studies did not present results in a format that could be used in the incidence and prevalence tables. A summary of the characteristics of the studies included in this systematic review and results on the frequency of rheumatic diseases are presented in Tables II to XI.

We conducted a thorough assessment of the methodological quality of the included studies using the checklist for incidence and prevalence studies of chronic diseases (Methodological Evaluation of Observational Research)⁴⁵. For each study, the report firstly lists flaws and issues not reported related to external validity followed by flaws and issues not reported related to internal validity. The authors of this checklist did not propose numerical rating of quality, quantitative values for criteria or numerical weighting of flaws. We did not appraise the methodological quality of two articles estimating the frequency of acute low back pain^{18,35} since it is not a chronic disease and of one article providing prevalence of several rheumatic diseases²⁸, since it was based on secondary data combining information from patients followed by specialists and the prevalence of these diseases in other countries.

Results

We identified 32 studies reporting the incidence or the prevalence of rheumatic diseases in the Por-

Table II. Frequency of rheumatic diseases (studies characteristics and results)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition	Study population	% males	Sample size	% participation	Age distribution	Estimates	
									Lifetime prevalence: Both sexes: 16.0% M: 11.1% F: 20.5%	Lifetime prevalence (medical or nursing diagnosis): Both sexes: 14.6% Prevalence in the previous year: Both sexes: 14.5%
INSA, IP/INE, IP, 2009 ³²	2005-2006	Face to face interview	To have or have had any rheumatic disease/Previous medical or nursing diagnosis of a rheumatic disease/To have or have had any rheumatic disease in the previous year (all self-reported)	Residents in Portugal familiar dwellings	48.4%	41 193 individuals (15 239 dwellings)	76%	Age groups (years): <15: 15.6% 15-24: 12.4% 25-34: 15.6% 35-44: 14.8% 45-54: 13.3% 55-64: 11.2% 65-74: 9.6% 75-84: 5.9% ≥85: 1.5%	Lifetime prevalence: Both sexes: 16.0% M: 11.1% F: 20.5%	Lifetime prevalence (medical or nursing diagnosis): Both sexes: 14.6% Prevalence in the previous year: Both sexes: 14.5%
Branco M, 2005 ¹⁴	2004	Telephone interview	To have a rheumatic disease (self-reported)	Residents in Portugal mainland familiar dwellings with landline (Portugal Telecom)	47.4%	2 820	82.1%	Age groups (years): 0-4: 2.8% 5-9: 4.3% 10-14: 4.8% 15-24: 12.6% 25-34: 12.6% 35-44: 12.8% 45-54: 14.4% 55-64: 14.8% 65-74: 12.6% ≥75: 8.2%	Point prevalence: Both sexes: 24.0% (95%CI:22.4-25.7)	Point prevalence: Both sexes: 24.0% (95%CI:22.4-25.7)
Costa L, 2004 ¹³	1999-2003	Face to face interview	History of medical diagnosed of, at least, one of these diseases: rheumatoid arthritis, systemic lupus erythematosus, ankylosing spondylitis, psoriatic arthritis, hip osteoarthritis, knee osteoarthritis and chronic low back pain (self-reported)	Adult population of the city of Porto	36.4%	1 238	70.0%	Age groups (years): ≤40: 33.9% 41-59: 36.2% ≥59: 29.9%	Lifetime prevalence: Both sexes: 23.0% M: 13.1% (95%CI: 10.2-16.6) F: 28.7% (95%CI: 25.6-32.0)	Lifetime prevalence: Both sexes: 23.0% M: 13.1% (95%CI: 10.2-16.6) F: 28.7% (95%CI: 25.6-32.0)

INSA, IP/INE, IP – National Institute of Health/Statistics-Portugal; M – males; F – females; 95%CI - 95% confidence intervals.

Table III. Frequency of osteoarthritis (studies characteristics and results)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition	Study population	% participation			Age distribution	Estimates
					% males	Sample size	% participation		
Costa L, 2004 ¹³	1999-2003	Face to face interview	History of medical diagnosed hip osteoarthritis or knee osteoarthritis (self-reported)	Adult population of the city of Porto	36.4%	1 238	70.0%	Age groups (years): ≤40: 33.9% 41-59: 36.2% ≥59: 29.9%	Lifetime prevalence (hip osteoarthritis): Both sexes: 5.5% (95%CI:4.3-7.0) M: 2.2% (95%CI: 1.4-2) F: 7.4% (95%CI:5.7-9.5) Lifetime prevalence (knee osteoarthritis): Both sexes: 11.1% (95%CI:9.4-13.1) M: 5.9% (95%CI:3.9-8.6) F: 14.2% (95%CI:11.8-16.9)

M – males; F – females; 95%CI - 95% confidence intervals.

tuguese population and published during the last decade.

Rheumatic diseases

Two studies meeting our inclusion criteria describing the overall prevalence of rheumatic diseases were identified from the bibliographic database search^{13,14}. Additionally we identified the *4th National Health Survey* from which we calculated the lifetime prevalence of rheumatic diseases³² (Table II).

The number of subjects evaluated ranged from 1 238, 36.4% males, in Costa *et al* (2004)¹³ to 41 193, 48.4% males, in the *4th National Health Survey*³². Branco *et al* (2005)¹⁴ and the *4th National Health Survey* described samples with wide age ranges (from childhood to over 75) while Costa *et al* (2004) enrolled participants over 18 years. In these three studies the cases of rheumatic disease were defined according the participant's self-reported history of any medically-diagnosed rheumatic disease. Overall, the prevalence of self-reported rheumatic disease ranged from 16.0%³² to 24.0%¹⁴. In women estimates ranged from 20.5%³² to 29.1% (95% confidence intervals (95%CI): 26.7-31.7)¹⁴ and in men these ranged from 11.1%³² to 18.3% (95%CI: 16.2-20.6)¹⁴.

Osteoarthritis

The prevalence of osteoarthritis was obtained from the aforementioned study by Costa et al (2004)¹³ (Table III). Cases were defined according to self-reported history of clinically diagnosed hip and/or knee osteoarthritis. Overall the prevalence of self-reported osteoarthritis was higher in the knee [11.1% (95%CI: 9.4-13.1)] than in the hip [5.5% (95%CI: 4.3-7.0)].

Back pain

We identified two studies estimating the incidence^{18,35} and seven studies estimating the prevalence of back pain^{13,15-17,33,36,37} (Table IV). Both studies estimating incidence were performed by the *Network of Sentinel Doctors* through registration of new episodes of acute low back pain in 2003¹⁸ and in 2004³⁵. In 2004, in 124 709 patients (47.8% males), the global yearly incidence was 1 817.8 per 100 000 population, higher in women (2 157.4) than in men (1 446.7)³⁵. Prevalence studies described samples that ranged from 215 [Vital *et al* (2006)³⁷] to 4 817 participants [Matos *et al* (2006)³⁶] and a male proportion varying from 35.4% [Rabaias *et al* (2003)³³] to 55.8% [Vital *et al* (2006)]. These

Table IV. Frequency of back pain (studies characteristics and results)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition	Study population	% males	Sample size	% participation	Age distribution	Estimates
Network of Sentinel Doctors, 2006 ³⁵	2004	New cases of disease notified by the physicians network	Clinical criteria for the diagnosis of acute low back pain	Clients of mainland Portugal health care centers	47.8%	124 709	2.3% of general practitioners in Portugal	Age groups (years): 0-4: 5.8% 5-9: 5.2% 10-14: 5.4% 15-24: 12.9% 25-34: 15.9% 35-44: 14.6% 45-54: 12.5% 55-64: 10.2% 65-74: 9.9% ≥75: 7.6%	Yearly incidence rate: Both sexes: 1 817.8 per 100 000 M: 1 446.7 per 100 000 F: 2 157.4 per 100 000
Rabiai S, 2003 ³³	2002	Telephone interview	Low back pain in the previous week (at least one day)	Adult residents in Portugal mainland familiar dwellings with landline (Portugal Telecom)	35.4%	1 414	84.6%	Age groups (years): 18-24: 5.8% 25-34: 8.3% 35-44: 18.0% 45-54: 19.9% 55-64: 20.8% 65-74: 17.4% ≥75: 9.7%	Prevalence in the previous week: Both sexes: 51.3% (95%CI:48.6-53.9) M: 36.5% (95%CI:32.2-40.9) F: 59.4% (95%CI:56.2-62.6)
Costa L, 2004 ¹³	1999-2003	Face to face interview	History of medical diagnosed chronic low back pain (lasting more than 3 months) (self-reported)	Adult population of the city of Porto	36.4%	1 238	70.0%	Age groups (years): ≤40: 33.9% 41-59: 36.2% >59: 29.9%	Lifetime prevalence: Both sexes: 12.3% (95%CI:10.5-14.3) M: 6.4% (95%CI:4.4-9.2) F: 15.6% (95%CI:13.2-18.4)
Ponte C, 2005 ¹⁵	2004	Telephone interview	Low back pain in the previous 6 months	Adult users of a health center of Porto	52.0%	296	98.6%	Age groups (years): 18-29: 19.6% 30-39: 24.3% 40-49: 22.0% 50-65: 34.1%	Prevalence in the previous 6 months: Both sexes: 49.0% (95%CI: 43.3-54.7) M: 44.2% (95%CI:36.2-52.4) F: 54.2% (95%CI:45.7-62.6)

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Table IV. (continuation)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition	Study population		% males	Sample size	% participation	Age distribution	Estimates
				% males	Study population					
Macos M, 2006 ³⁶	2006	Self administered questionnaire	Back pain in the previous 6 months (almost every day pain or more than once a week)	6th, 8th and 10th grades Portuguese students	49.6% 4 817	1,6% of the population of students enrolled in academic year 2005/2006	n/av	Mean age: 14 years	Prevalence in the previous 6 months:	
Costa M, 2002 ¹⁶	1999-2000	Face to face interview by a rheumatologist	Back pain in the previous 3 months	Public school students from the metropolitan region of Lisbon	45.6% 767			Mean age (standard deviation); [range]: 10.6 (2.4) years; [6-17]	Both sexes: 8.0% (95%CI: 6.1-10.1)	M: 4.3% (95%CI: 2.4-7.0) F: 11.0% (95%CI: 8.2-14.4)
Coelho L, 2009 ¹⁷	2005-2006	Self administered questionnaire	All and any kind of painful complaint in the lumbar region which has lasted at least 24 hours	Young people resident in the Greater Lisbon area and attending educational establishments there (both state-run and private)	47.6% 439	n/av		Mean age (standard deviation); [range]: 13.0 (1.1) years; [11-15]	Annual prevalence: Both sexes 20.5% (95%CI: 16.8-24.6) M: 11.0% (95%CI: 7.1-16.0) F: 29.2% (95%CI: 23.3-35.5)	
Vital E, 2006 ³⁷	2003-2004	Self administered questionnaire	Back pain in the previous 6 months (almost every day pain or more than once a week)	5th grade students from two Portugal Center region municipalities	55.8% 215	97.3%		Point prevalence: Both sexes: 16.3% (95%CI: 5.2-10.4) Lifetime prevalence: Both sexes: 25.5% (95%CI: 21.5-29.9)	Prevalence in the previous 6 months: 29.5% (95%CI: 23.4-36.2) Prevalence in the previous 6 months (first episodes): Both sexes: 16.3 (95%CI: 11.6-22.1) ¹⁸	

n/av – Not available; M – males; F – females; 95%CI – 95% confidence intervals; I, II – Estimates calculated from the results presented in the study

Table V. Frequency of work-related musculoskeletal disorders (studies characteristics and results)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition	Study population	% males	Sample size	% participation	Age distribution	Estimates	
									Point prevalence:	Point prevalence:
Cunha-Miranda L, 2010 ¹⁹	2006	Questionnaire sent to the occupational physician	Clinically relevant VRMDs	Large dimension companies in Portugal (over 250 employees)	48.4%	410 496 workers (515 companies)	62.3%	Mean age (standard deviation): M: 39.5 (5.7) years F: 37.9 (5.1) years	5.9% (total WRMDs clinically relevant)	5.9% (total WRMDs clinically relevant)
Parent-Thirion A, 2007 ³⁸	2005	Face to face interview	Backache or muscular pains in shoulders, neck and/or upper/lower limbs related to occupational activity	Residents in Portugal in paid employment (employees and self-employed), during the field-work period (over 15 years old)	n/av	1 000	67%	n/av	Point prevalence of backache: 30.7%	Point prevalence of muscular pains in shoulders, neck and/or upper/lower limbs: 28.8%
Carnide F, 2006 ²⁰	2004	Interview and a clinical exam conducted by an occupational medical doctor	WRMSs (presence of pain or paraesthesia during the last year, with episodes lasting for at least 1 week or occurring at least once a month, with no previous acute trauma)/ VRMDs evaluated by an occupational medical doctor	Automobile industry workers (paint area production)	n/av	29	n/av	Mean age (standard deviation): 37.7 (8.2) years	Point prevalence: Neck: WRMSs: 10.3% (95%CI:2.2-27.4) VRMDs: 58.6% (95%CI:38.9-76.5) Right shoulder: WRMSs: 24.1% (95%CI:10.3-43.5) VRMDs: 20.7% (95%CI:8.0-39.7) Left shoulder: WRMSs: 6.9% (95%CI:0.8-22.8)	Point prevalence: Neck: WRMSs: 10.3% (95%CI:2.2-27.4) VRMDs: 58.6% (95%CI:38.9-76.5) Right shoulder: WRMSs: 24.1% (95%CI:10.3-43.5) VRMDs: 20.7% (95%CI:8.0-39.7) Left shoulder: WRMSs: 6.9% (95%CI:0.8-22.8)

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Table V. (continuation)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition	Study population	% males	Sample size	% participation	Age distribution	Estimates
Serranheira F, 2001 2003 ³⁹		Self-administered questionnaire	WRMSS in the previous year/week (adaptation of the Standardised Nordic questionnaire for the analysis of musculoskeletal symptoms)	Automobile components industry workers (industry in Setubal peninsula)	16.1%	574	63.2%	Modal class [range]: 26-33 years [18-65]	Prevalence in the previous year: Cervical region: 83.0% Shoulders: 57.5% Elbows: 21.4% Wrists/hands: 66.7% Upper back region: 50.3% Low back region: 55.4% Hips/thigh: 31.1% Legs/knees: 71.2% Ankles/feet: 63.7% Prevalence in the previous week: Cervical region: 29.1%
									Continues on the next page

Table V. (continuation)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition	Study population	% males	Sample size	% participation	Age distribution	Estimates
Uva A, 2005 ⁴⁰	2002	Self-administered questionnaire	Back pain related to occupational activity (physicians, nurses, health care allied professionals and clerks working in health facilities, in government agencies and in other sectors not directly related to the provision of care)	Ministry of Health workers	25%	1 754	41%	Age groups (years): <20: 1.1% 20-29: 20.9% 30-39: 30.7% 40-49: 25.7% 50-59: 18.6% ≥60: 2.3% Not specified: 0.7%	Point prevalence: Both sexes: 47%
Carneiro P, 2005 ⁴¹	2003	Self-administered questionnaire	Pain or discomfort related to occupational activity	Portuguese dentists	44%	697	16.6%	Mean age (standard deviation): M: 36.1 (8.3) years F: 32.1 (6.2) years	Prevalence in the previous year: M: 75.5% (95%CI:69.8-80.5) F: 83.3% (95%CI:79.0-87.0)
Vilão S, 2005 ⁴²	2000	Self-administered questionnaire	Have suffered at least one WRMDs (self-reported)	Physiotherapists working in Portugal Center region hospitals	13%	41	n/av	Age groups (years): 21-30: 50% 31-40: 42% 41-60: 8%	Lifetime prevalence: Both sexes: 58.5% (95%CI:42.1-73.7)
Fonseca R, 2006 ⁴³	2004	Self-administered questionnaire	WRMSS (Standardised Nordic questionnaire for the	Nurses working in Porto hospitals	16%	507	56%	Median [range]: 31 years [22-63]	Prevalence in the previous year: Both sexes: 84.2% (95%CI: 80.8-87.3)

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Table V. (continuation)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition analysis of musculoskeletal symptoms)	Study population	% males	Sample size	% participation	Age distribution	Estimates
Martins J, 2008 ⁴⁴	2007	Self-administered questionnaire	WRMSs (Standardised Nordic questionnaire for the analysis of musculoskeletal symptoms)	Nurses working in one hospital (medicine, surgery and orthopedics services)	18.2%	176	80%	Mean age (standard deviation); [range]; 33.4 (7.5) years; [23-66]	Prevalence in the previous week: Cervical region: 53% Shoulders: 6 % Elbows: 33% Wrist/hands: 45% Dorsal region: 62% Low back region: 58% Thighs: 54% Knees: 56% Ankles/feet: 71%

n/av – Not available; WRMDs – work-related musculoskeletal disorders; WRMSs – work-related musculoskeletal symptoms; M – males; F – females; 95%CI – 95% confidence intervals

studies used distinct definitions of back pain and different prevalence estimates (point prevalence¹⁷, period prevalence in previous specified days / months/years^{15-17,33,36} or lifetime prevalence^{13,17}). In four studies samples of children and adolescents (6 to 17 years) were evaluated^{16,17,36,37} and three studies enrolled adults^{13,15,33}. Among children and adolescents, the prevalence of back pain ranged from 8.0% (95%CI: 6.1-10.1), if classified as pain in the previous three months¹⁶, to 29.5% (95%CI: 23.4-36.2), if classified as pain in the previous six months³⁷. In adults, low back pain prevalence ranged from 12.3% (95%CI: 10.5-14.3), when evaluated as lifetime low back pain lasting more than three months¹³, to 51.3% (95%CI: 48.6-53.9), when classified as at least one day of pain in the previous seven days³³.

Work-related musculoskeletal disorders

We identified nine studies estimating the prevalence of work-related musculoskeletal disorders (WRMDs)^{19,20,38-44} (Table V). To define cases of WRMDs Cunha-Miranda *et al* (2010)¹⁹ and Carnide *et al* (2006)²⁰ considered only clinically relevant disorders, with medical diagnosis, while other studies considered work-related musculoskeletal symptoms (WRMSs) as reported by workers³⁸⁻⁴⁴. The sample sizes ranged from 29 [Carnide *et al* (2006)] to 1 000 (Parent-Thirion *et al* 2007³⁸) when workers were inquired directly while, in the study by Cunha-Miranda *et al* (2010) addressing 515 occupational health doctors, a coverage of 410 496 workers was achieved¹⁹. The proportion of males ranged from 13.0% [Vilão *et al* (2005)⁴² to 48.4% (Cunha-Miranda *et al* (2010)], (the gender distribution was not available for two studies^{20,38}), and Parent-Thirion *et al* (2007) included workers from the age of 15. Cunha-Miranda *et al* (2010), Parent-Thirion *et al* (2007) and Uva *et al* (2005)⁴⁰ presented data on the prevalence on WRMDs for several professional categories whereas other studies focused on particular occupational activities (e.g. industrial workers^{20,39}, health care providers⁴¹⁻⁴⁴). The prevalence of WRMDs ranged from 5.9%, considering clinically relevant disorders in several occupational activities¹⁹, to 84.2% (95%CI: 80.8-87.3) regarding symptoms reported by hospital nurses⁴³.

Table VI. Frequency of osteoporosis or osteoporotic fracture (studies characteristics and results)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition	Study population	% males	Sample size	% participation	Age distribution	Estimates	
									Yearly incidence rate: M: 129.4 per 100 000 F: 351.9 per 100 000	Yearly incidence rate: Both sexes: 63.9 per 100 000 M: 31.9 per 100 000 F: 93.3 per 100 000
Pina M, 2008 ³³	2000-2002	Selection of admissions from the National Hospital Discharge Register, (Health Informatics and Financial Management Institute)	Hip fracture diagnosis according the ICD9-CM classification (codes ICD9-CM 820.x) caused by a low or moderate fall	Residents in Portugal mainland (over 50 years old)	n/ap	n/ap	n/ap	n/ap		
Network of Sentinel Doctors, 2009 ³⁴	2007	New cases of fracture reported to the physicians network	Medical diagnosis of hip fracture	Clients of mainland Portugal health care centers	47.9%	111 129 patients (141 general practitioners)	2.3% of general practitioners in Portugal	Age groups (years): 0-4: 5.3% 5-9: 5.3% 10-14: 5.1% 15-24: 11.7% 25-34: 15.7% 35-44: 14.8% 45-54: 13.2% 55-64: 10.8% 65-74: 9.6% ≥ 75: 8.4%	Yearly incidence rate: Both sexes: 63.9 per 100 000 M: 31.9 per 100 000 F: 93.3 per 100 000	Yearly incidence rate: Both sexes: 351 per 100 000 (95%CI:302-406) M: 154 per 100 000 (95%CI: 106-218) F: 481 per 100 000 (95%CI:407-564)
Costa J, 2009 ³⁵	2007	Clinical report consultation of patients hospitalized	Hip fracture occurring spontaneously or from a fall no greater than standing height	Residents in Viana do Castelo district (over 65 years old)	n/ap	n/ap	n/ap	n/ap		
Cruz M, 2009 ²¹	2004-2006	Clinical report consultation of patients hospitalized	Proximal femur fracture after a low impact fall	Residents in the catchment area of Caldas da Rainha Hospital (over 50 years old)	n/ap	n/ap	n/ap	n/ap	Yearly incidence rate: 2004: 80 per 100 000 2005: 100 per 100 000 2006: 80 per 100 000	Continues on the next page

Table VI. (continuation)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition	Study population	% males	Sample size	% participation	Age distribution	Estimates
INSA, IP/INE, IP, 2009 ³²	2005-2006	Face to face interview	To have or have had osteoporosis/Previous medical or nursing diagnosis of osteoporosis/To have or have had osteoporosis in the previous year (all self-reported)	Residents in Portugal familiar dwellings	48.4%	41 193 individuals (15 239 dwellings)	76%	Age groups (years): <15: 15.6% 15-24: 2.4% 25-34: 5.6% 35-44: 4.8% 45-54: 13.3% 55-64: 11.2% 65-74: 9.6% 75-84: 5.9% ≥85: 1.5%	Lifetime prevalence: Both sexes: 6.2% M: 1.1% F: 11.0% Lifetime prevalence (medical or nursing diagnosis): Both sexes: 6.1% Prevalence in the previous year: Both sexes: 5.6%
Marantes I, 2004 ²⁶	2001-2002	Telephone interview	Self-reported osteoporosis	Adult women residents in Portugal mainland	0%	1 800	n/av	Age groups (years): 18-29: 3.8% 30-39: 6.0% 40-49: 9.6% 50-59: 16.3% 60-69: 17.6% 70-79: 12.9% ≥80: 3.8%	Point prevalence: 15.4% (95%CI:13.4-17.6) of which 96.8% (95%CI:93.1-98.8) had diagnostic exams that confirm the disease
Bernardo A, 2003 ²⁴	2001-2002	Bone density measurement	WHO operational definition of osteoporosis based on the BMD values measured by DXA (lower spine and hip)	Males clients of one health care center of the city of Porto (over 20 years old)	100%	226	n/av	Mean age (standard deviation); [range]: 57.2 (16.0) years; [21-87]	Point prevalence (osteoporosis): 16.8% (95%CI:12.2-22.3) Point prevalence (osteopenia): 50.9% (95%CI:44.2-57.6)
Lucas R, 2008 ²²	1994-1995	Bone density measurement	WHO operational definition of osteoporosis based on the BMD values measured by DXA (non-dominant distal forearm)	Caucasian adult males residents in Portugal mainland	100%	739	n/av	Age groups (years): 20-39: 12.2% 40-49: 28.3% 50-59: 33.7% 60-69: 17.9% ≥70: 8.0%	Point prevalence: >39 years: 4.3% (95%CI:2.9-6.2) 40-49 years: 1.9% (95%CI:0.5-4.8) ≥70 years: 18.6% (95%CI:9.7-30.9)

n/aP – Not applicable; M – males; F – females; 95%CI – 95% confidence intervals; INSA, IP/INE, IP – National Institute of Health/Statistics-Portugal; n/av – Not available; WHO – World Health Organization; BMD – bone mineral density; DXA – dual energy X-ray absorptiometry.

Osteoporosis and osteoporotic fractures

Four studies estimated the incidence of osteoporotic fracture^{21,23,25,34} (Table VI). Data collection methods included the search of hospital admissions records with hip fracture^{21,23,25} or cases of fracture reported by primary care patients to the *Network of Sentinel Doctors*³⁴. All studies included both males and females and the yearly incidence of fracture (per 100 000 inhabitants) in women ranged from 93.3³⁴ to 481 (95%CI: 407-564)²⁵ and in men from 31.9³⁴ to 154 (95%CI: 106-218)²⁵.

Data on osteoporosis prevalence were obtained from three studies^{22,24,26} and calculated using the *4th National Health Survey*³² (Table VI). The number of subjects evaluated ranged from 226 [Bernardo *et al* (2003)²⁴] to 41 193 (*4th National Health Survey*). The *4th National Health Survey* included both sexes (48.4% males), Marantes *et al* (2004)²⁶ included only females while Bernardo *et al* (2003)²⁴ and Lucas *et al* (2008)²² included only males. Most studies evaluated samples of adults but the *4th National Health Survey* included participants with a wide age range (from childhood to over 85). Case definition was based on actual bone density measurement^{22,24} or self-reported history of osteoporosis^{26,32}. Among women the prevalence of self-reported osteoporosis ranged from 11.0%³² to 15.4% (95%CI: 13.4-17.6)²⁶. Among men it ranged from 1.1% when self-reported³², to 16.8% (95%CI: 12.2--22.3) based on actual bone mineral density measurements²⁴.

Fibromyalgia

One study by Branco *et al* (2010) estimated the prevalence of fibromyalgia²⁷ (Table VII). A positive screen case was considered if meeting the 4-pain criteria of the *London Fibromyalgia Epidemiological Study Screening Questionnaire* (LFESSQ-4), or meeting both the 4-pain and the 2-fatigue criteria (LFESSQ-6). This study included 4 517 participants over 15 years (48% males) and the prevalence of fibromyalgia in the general population was estimated by applying the positive-predictive values obtained in a rheumatology clinic-based sample to eligible community subjects (i.e., positive screens). In the Portuguese general population the estimated overall prevalence was 3.7% (95%CI: 2.0-5.4) based on positive screens for LFESSQ-4, and 3.6% (95%CI: 2.0-5.2) based on positive screens for LFESSQ-6²⁷.

Rheumatoid arthritis

Two studies described the prevalence of rheuma-

toid arthritis^{13,28} (Table VIII). In the aforementioned study by Costa *et al* (2004) the prevalence of self-reported rheumatoid arthritis in women was 2.5% (95%CI: 1.5-3.9) and in men was 0.0% (95%CI: 0.0-1.1)¹³. Abreu *et al* (2006) estimated the prevalence of rheumatoid arthritis based on secondary data combining information from patients followed by Internal Medicine specialists and the prevalence of other countries. The proposed prevalence for Portugal was 0.5 to 1%²⁸.

Spondyloarthritis

Two studies provided estimates of the prevalence of ankylosing spondylitis^{13,29} (Table IX). In the aforementioned study of Costa *et al* (2004)¹³ participants were questioned about medical diagnosis of ankylosing spondylitis while Bruges-Armas *et al* (2002) studied recruited subjects over 50 years (52.0% males) by clinical examination²⁹. Although different data collection methods were used, the estimated prevalence of ankylosing spondylitis was 0.6% in both studies. The prevalence of spondyloarthritis assessed by dorsal, lumbar, and pelvic radiography, was 1.6% (95%CI: 0.8-2.7)²⁹. No cases of psoriatic arthritis were identified¹³.

Other systemic rheumatic diseases

We identified three studies reporting on the prevalence of other systemic rheumatic diseases^{13,28,30} (Table X). The study aforementioned by Costa *et al* (2004) estimated the self-reported prevalence of systemic lupus erythematosus in 0.2% (95%CI: 0.1-0.8)¹³. Gouveia *et al* (2005), reporting cases in children under 5 years diagnosed at a single hospital (Hospital Fernando Fonseca), provided a yearly incidence of Kawasaki disease of 8.2 per 100 000 children³⁰. In the aforementioned study of Abreu *et al* (2006) the proposed prevalence, for Portugal, for systemic lupus erythematosus ranged from 0.015 to 0.05%, for scleroderma ranged from 0.04 to 0.25%, for Sjögren's syndrome was 3% and for Behcet's disease ranged from 0.01 to 0.3%²⁸.

Pediatric rheumatic diseases

In 2004, Salgado *et al* reported prevalence and incidence estimates of pediatric rheumatic diseases and juvenile idiopathic arthritis³¹ (Table XI). The patients were identified in the pediatric rheumatology outpatient clinic and cases were defined according the International League of Associations for Rheumatology criteria. Among children under 16 years of age, one in every 1 785 children had or

Table VII. Frequency of fibromyalgia (studies characteristics and results)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition	Estimates			
				Study population	% males	Sample size	% participation
Branco J, 2010 ²⁷	2003-2006	Telephone interview	Diagnostic criteria from the LFESSQ	General Population (over 15 years old)	48%	4 517	n/av

LFESSQ - London Fibromyalgia Epidemiological Study Screening Questionnaire; n/av - Not available; LFESSQ-4 – meeting the 4-pain criteria alone from the LFESSQ; LFESSQ-6 – meeting both the 4-pain and the 2-fatigue criteria from the LFESSQ; M – males; F – females; 95%CI - 95% confidence intervals.
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LFESSQ - London Fibromyalgia Epidemiological Study Screening Questionnaire; n/av - Not available; LFESSQ-4 – meeting the 4-pain criteria alone from the LFESSQ; LFESSQ-6 – meeting both the 4-pain and the 2-fatigue criteria from the LFESSQ; M – males; F – females; 95%CI - 95% confidence intervals.

had had a pediatric rheumatic disease, and one in 3 225 children had or had had juvenile idiopathic arthritis. The incidence was calculated for the previous three years using the pediatric catchment population (<16 years) and was 8.4 per 100 000 children-year for pediatric rheumatic diseases and 4.3 per 100 000 children-year for juvenile idiopathic arthritis³¹.

No studies estimating the frequency of periarthritis rheumatic diseases or microcrystal-related arthropathies in Portugal were identified from the literature search.

Methodological quality appraisal

Overall the studies included were of low reporting and methodological quality. Major flaws jeopardizing external validity of these studies were related to low response rates in total sample⁴¹ while minor flaws were related to random samples restricted to geographic area^{13,21,24,25,30,31} and to lack of assessment and address of sampling bias^{13-17,19,20,22,24,26,27,29-31,35,37,39,40,42-44}. External validity was also compromised by poor reporting in these issues: response rate, exclusion rate from the analysis or whether sample bias was address. Low internal validity was related to major flaws in the definition of incidence or prevalence (e.g. when severity of the disease could have been relevant but was not assessed)^{13-17,19,22,24,26,36-38,41,42,44} and to the use of not validated methods^{13-17,26,29,32-34,36,37,39,40,42-44}. Minor flaws in internal validity included use of methods poorly validated²⁷ and access to unreliable data sources to measure prevalence/incidence^{19,21,23,25,30,31}. In almost every study, low internal validity was also consequence of not reported information about validity and/or reliability of the methods employed.

Discussion

In Portugal, a large amount of information is available derived from a wide variety of methods resulting in broad ranges of frequency estimates. We identified 32 studies reporting the incidence or the prevalence of rheumatic diseases in the Portuguese population and published during the last decade. This systematic review retrieved information for nine of the 11 rheumatic conditions targeted by the *National Program against Rheumatic Diseases*. Most studies computed prevalence, being back pain and work-related musculoskeletal disorders

Table VIII. Frequency of rheumatoid arthritis (studies characteristics and results)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition	Study population			% participation	Age distribution	Estimates
				% males	Sample size	Age groups (years):			
Costa L, 2004 ¹³	1999-2003	Face to face interview	History of medical diagnosed rheumatoid arthritis (self-reported)	Adult population of the city of Porto	36.4% 238	70.0%			Lifetime prevalence: Both sexes: 1.6% (95%CI:1.0-2.5) M: 0.0% (95%CI:0.0-1.1) F: 2.5% (95%CI:1.5-3.9)

M – males; F – females; 95%CI - 95% confidence intervals.

the most frequent rheumatic conditions assessed.

The timeline of the search was selected aiming to optimize the relation between the sensitivity desired and the resources available, in order to improve yield as much as feasible. We identified a review of studies estimating the prevalence of rheumatic diseases in the Portuguese population, performed since 1976, and published in 2002⁵. From this review we were able to learn that there were few studies published before 2000 and most of their results were published outside widely available journals (grey literature). To appraise these and other studies published before 2000 would require an extensive search of the grey literature available in Portugal, a time-consuming process with a very low efficiency. Bearing in mind the aim of this paper we think that limiting the timeline of the research strategy did not substantially decrease the quality and the yield of the search.

Rheumatic diseases

The overall prevalence of rheumatic diseases in Portugal varied between 14.5% and 24.0%. The difference in the estimates found in the Portuguese population could be related to the definition of the rheumatic disease but also to survey design factors and to the age and gender distribution of the study population⁴⁶.

Comparability of frequency measures from different populations is limited owing to methodological and cultural differences. For instance, the use of a list of rheumatic diseases is expected to produce higher prevalence estimates than an open and more general description of rheumatic disease⁴⁶. The description and meaning of different rheumatic diseases differ between medical specialists and general public, from different cultures and languages, and can also result in highly different estimates⁴⁶. A recent study performed in the general Portuguese population showed that there are incorrect beliefs about rheumatic diseases being the identification of rheumatic disease the major flaw⁴⁷.

The main limitation of national health surveys, such as the *4th National Health Survey*, remains in the impossibility of using standard clinical criteria to define cases, and rather relying on self-reported symptoms and diagnoses. The estimate of frequency obtained by self-report reflects much more than the occurrence of the condition. It reflects a set of constructs such as symptoms, access to health care and the awareness of disease. If, on the one hand, validation studies have supported the

Table IX. Frequency of spondyloarthritis (studies characteristics and results)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition	Study population	% males	Sample size	% participation	Age distribution	Estimates
Bruges-Armas J, 2002 ²⁹	1994	Clinical examination	European Spondyloarthropathy Study Group diagnostic criteria to spondyloarthritis and modified New York diagnostic criteria to ankylosing spondylitis	Caucasian residents of Terceira Island, Azores (over 50 years old)	52.0%	490	52.4%	Mean age: M - 66 years F - 67 years	Spondyloarthritis prevalence: Both sexes: 1.6% (95%CI:0.8-2.7) M: 2.7% (95%CI: 1.1-5.6) F: 0.4% (95%CI:0.0-2.5) Ankylosing spondylitis prevalence: Both sexes: 0.6% (95%CI:0.1-1.8) M: 1.2% (95%CI:0.2-3.4) F: 0.0% (95%CI:0.0-1.6)
Costa L, 2004 ¹³	1999-2003	Face to face interview	History of medical diagnosed ankylosing spondylitis or psoriatic arthritis (self-reported)	Adult population of the city of Porto	36.4%	1 238	70.0%	Age groups (years): ≤40: 33.9% 41-59: 36.2% >59: 29.9%	Lifetime prevalence (ankylosing spondylitis): Both sexes: 0.6% (95%CI: 0.3-1.3) M: 0.4% (95%CI:0.1-1.8) F: 0.8% (95%CI:0.3-1.7) Lifetime prevalence (psoriatic arthritis): Both sexes: 0.0% (95%CI:0.0-0.4) M: 0.0% (95%CI:0.0-1.1) F: 0.0% (95%CI:0.0-0.6)

M – males; F – females; 95%CI - 95% confidence intervals.

Table X. Frequency of other systemic rheumatic diseases (studies characteristics and results)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition	Study population	% males	Sample size	% participation	Age distribution	Estimates	
									Yearly incidence rate: 8.2 per 100 000	Lifetime prevalence:
Gouveia C, 2005 ³⁰	1996-2003	Clinical report consultation of patients hospitalized	American Heart Association diagnostic criteria for Kawasaki disease	Residents in the catchment area of Fernando da Fonseca Hospital (children under 5 years old)	n/ap	n/ap	n/ap	n/ap		
Costa L, 2004 ¹³	1999-2003	Face to face interview	History of medical diagnosis of systemic lupus erythematosus (self-reported)	Adult population of the city of Porto	36.4%	1 238	70.0%	Age groups (years): ≤40: 33.9% 41-59: 36.2% >59: 29.9%	Both sexes: 0.2% (95%CI:0.1-0.8) M: 0.2% (95%CI:0.1-1.4) F: 0.3% (95%CI:0.1-1.0)	

n/ap – Not applicable ; M – males; F – females; 95%CI - 95% confidence intervals.

accuracy of self-report to measure prevalent chronic conditions and severe diseases which require treatment⁴⁸, on the other, one study which assessed the agreement between musculoskeletal diseases' self-reporting, with six months in between, reached different conclusions⁴⁶. Although case ascertainment from self reports has sub-optimal accuracy, for the assessment of diseases that are characterized by pain and functional limitation, individual subject is our single best source of information⁴⁶.

One of the solutions to improve diagnostic validity in population studies seems to be the collection of information on medication to confirm self-reported cases^{49,50}. However, when we use medications to define disease in an epidemiologic study, we assume that all cases of the disease have been properly diagnosed and treated. This excludes undiagnosed, untreated, misdiagnosed, and mistreated cases and limits the study of early manifestations of disease. Using this approach to define disease also requires a meticulous medication data collection⁴⁹.

Case definition using clinical and complementary diagnostic examination provides better sensitivity and specificity. However, many rheumatic conditions still have no accepted universal case definition and clinical diagnosis methods are not easily applicable to general population samples.

Osteoarthritis

Costa *et al* (2004) estimated the prevalence of osteoarthritis in the Portuguese population based on self-reported history of disease¹³. Frequently, estimates of the prevalence of osteoarthritis are based on radiographic surveys^{6,48,51} which hampers comparisons with this study. In fact, there is a dissociation in the prevalence estimates between self-reported diagnosis of osteoarthritis and studies employing clinical and radiological criteria⁴.

The prevalence of osteoarthritis is usually higher in the knee than in the hip⁴ and those results were found in the Portuguese population. Comparing to a Dutch study, which used a similar methodology, the prevalence of knee and hip osteoarthritis in Portugal was slightly lower, except for knee osteoarthritis in women⁴⁶.

Back pain

Numerous studies on back pain epidemiology have been published over the past 10 years in Portugal. The estimate of low back pain in the pre-

Table XI. Frequency of pediatric rheumatic diseases (studies characteristics and results)

First author, publication year, reference	Period of data collection	Collecting data method	Case definition	Estimates			
				Study population	% males	Sample size	Age distribution
Salgado M, 2004 ³¹	2001-2004	Clinical report consultation	International League of Associations for Rheumatology diagnostic criteria	Residents in Centro region of Portugal (children under 16 years old)	n/ap	n/ap	n/ap
Salgado M, 2004 ³¹	1988-2004	Clinical report consultation	International League of Associations for Rheumatology diagnostic criteria	Residents in Centro region of Portugal (children under 16 years old)	n/ap	n/ap	n/ap

n/ap – Not applicable.

vious week found by Rabiaias *et al* (2003)³³ was much higher than the point prevalence of low back pain in Spain estimated in 14.8% (95%CI: 12.2-17.4)⁵².

To summarize and compare the results across these studies is difficult because of the heterogeneity of methods used. Particularly in this condition, studies showed substantial variation in case definition and distinct classification criteria, which affected the number of cases identified and probably explains apparent major differences in prevalence estimates of back pain⁴. Moreover, the results of studies focusing on symptoms are highly dependent on the formulation and wording of questions and this probably explains different patterns found in the prevalence of back pain when case definition and sampling frames are similar⁴⁸.

The lack of standardization and validation of the terminology and classification of back pain (frequency or duration of pain) also leads to confusion between prevalence and incidence estimates. In fact, as a consequence of the unstable and episodic nature and uncertainty of onset of back pain, the traditional epidemiological concept of incidence case definition is difficult to mainstream to this condition⁴.

In 2003, *World Health Organization* has recommended the characterization of back pain as period prevalence measures, defining acute (less than seven days), sub-acute (more than seven days) and chronic pain (more than 42 days)³. However, we only identified three studies defining duration of pain episodes that could be classified according to this criteria^{13,15,16}.

Work-related musculoskeletal disorders

A broad range of studies on the prevalence of musculoskeletal diseases or complaints among different occupational activities have been published recently in Portuguese workers. The results of the *Fourth European Working Conditions Survey* reported Portugal as one of the European countries where a high prevalence of back pain and muscular pain in shoulders, neck and/or upper/lower limbs related to occupational activity was observed. Furthermore, the estimates for Portugal were higher than European average which were 24.7% for back pain and 22.8% for muscular pain in shoulders, neck and/or upper/lower limbs³⁸.

Comparing the results of the first cross-sec-

tional study on the prevalence of WRMDs within large Portuguese companies by Cunha-Miranda *et al* (2010)¹⁹ with studies performed in other countries, clear differences were identified. The authors attributed these differences to presumable specificities of the Portuguese business sector or to differential clinical relevance given by Portuguese occupational physicians, or by patients themselves, compared to other countries¹⁹.

There are limitations in directly comparing estimates between different occupational activities because, regarding WRMDs in particular, the differences in definitions and questionnaires used might be part of the explanation for the findings.

Osteoporosis and osteoporotic fracture

The burden of osteoporosis can be estimated indirectly through its single most relevant outcome, the incidence of fragility fractures. We identified four articles estimating the incidence of hip fracture^{21,23,25,34}, of which three used hospital records^{21,23,25}. In fact, for these conditions which lead more or less inevitably to hospital admission, the total number of cases within a region and a time frame is relatively easy to obtain through hospital records⁴.

In this review, the differences in estimates between the studies can be related to different selection criteria for patients' ages, and comparisons must be carefully made since the incidence of hip fractures is strongly age dependent⁴.

Comparing the incidence of osteoporotic fractures obtained in Portugal with other European estimates, the overall incidence was higher than in Germany and lower than in Greece, Norway, Denmark and Finland. Comparing with Spain, the incidence in Portugal was lower in women and higher in men²³.

Lucas *et al* (2008) and Bernardo *et al* (2003) estimated osteoporosis prevalence using bone mineral density (BMD) measurement^{22,24}. Implicit in this definition of osteoporosis is the relationship between BMD decay and rise in fracture risk. However, when we aim to assess the burden of osteoporosis the densitometry based estimate of prevalence has limited value and in the future, the burden of osteoporosis will probably be assessed through clinical risk factors for fracture⁴.

Fibromyalgia

In an European survey estimating the prevalence of fibromyalgia in five countries, Branco *et al* (2010) identified no statistically significant differences in

the prevalence in Portugal compared to other countries²⁷. The main limitation of this study was the assessment of prevalence assuming positive-predictive values of LFESSQ-4 and LFESSQ-6 in the general population similar to those calculated in the rheumatology outpatient sample while the predictive value in the latter is probably substantially higher than in the former²⁷.

Rheumatoid arthritis

Costa *et al* (2004) estimated the prevalence of rheumatoid arthritis using self-reported information¹³. The validity of self-reported diagnosis of rheumatoid arthritis has been reported to be between 7 and 96%⁴⁹.

Several studies provided estimates on the prevalence of rheumatoid arthritis in defined populations and, although these studies have a number of methodological limitations, the remarkable finding is the uniformity of rheumatoid arthritis prevalence estimates in developed populations - approximately 0.3% to 1.0% of the adult population^{4,53}. The prevalence of rheumatoid arthritis in the Portuguese population was higher than aforementioned but similar to figures obtained in other settings when the disease was assessed by self-report⁴⁶.

Spondyloarthritis

The prevalence of 0.6% for ankylosing spondylitis found in the Portuguese population by Bruges-Armas *et al* (2002)²⁹ was slightly higher than other estimates reported in different populations, varying from 0.036% to 0.10%⁵³, but similar to the prevalence of ankylosing spondylitis in Greece assessed recently [0.3% (95%CI: 0.3-0.9)]⁵⁴.

Costa *et al* (2004) identified no cases of psoriatic arthritis in a sample of urban Portuguese adults¹³. A recent review described prevalence estimates for psoriatic arthritis varying from 1 case per 100 000 population, in Japan, to 420 cases per 100 000 population, in Italy⁵⁵.

Other systemic rheumatic diseases

The prevalence found by Costa *et al* (2004)¹³ in a sample of the Portuguese population was not very different from estimates previously reported. One of these studies validated self-reported diagnoses of systemic lupus erythematosus, by reviewing available medical records, revealing a prevalence of 0.12%⁵⁶. The validity of self-reported diagnosis of lupus has been estimated between 21% and 84%⁴⁹.

Pediatric rheumatic diseases

The majority of available studies on epidemiology of chronic arthritis in childhood is clinic-based and thus is susceptible to numerous biases⁵³. The few existing population studies have reported an approximate prevalence of juvenile rheumatoid arthritis between 1 and 2 per 1 000 children, and an incidence between 11 and 14 per 100 000 children⁵³. In the studies assessing the incidence of juvenile idiopathic arthritis published since 1995 the estimates ranged from 10 to 22.6 per 100 000 children³¹. Comparing these figures with the Portuguese estimates, according to Salgado *et al* (2004), it seems to exist a dispersion of the Portuguese patients (which may be examined by other medical specialties) or under diagnosis of cases³¹.

Methodological issues in estimating the frequency of rheumatic diseases

To better assess the burden of rheumatic diseases in the population it is important to obtain accurate estimates of the current proportion of people suffering from these conditions as well as their time trends and geographic distribution.

The estimate of rheumatic diseases frequency represents a particularly complex methodological challenge. The difficulty in defining a case, the lack of clear distinction between the different disorders and the difficulty in case ascertainment are specific problems when undertaking rheumatic disease epidemiology. Moreover, many of the rheumatic diseases are so rare that the required sample size of the surveys to ensure accuracy are probably in excess of what is feasible or cost-effective⁴⁸.

The diversity of rheumatic diseases, regarding both etiology and frequency, argues for the importance of using sources of information collected at different levels of care. Therefore, while health surveys of the general population can provide valid and accurate estimates of the frequency of highly prevalent diseases, their accuracy is very limited when studying less frequent conditions, with worse prognosis, such as systemic rheumatic diseases. To characterize the frequency of these uncommon diseases it is more appropriate to collect data at differentiated levels of health care⁴⁸. However, it is important to perceive that data collected at each level reflect the distribution of disease related factors, such as frequency and prognosis, but also the organization of the health system, collecting information systems, geographical distribu-

tion of the services, referral protocols and socio-demographic characteristics of the population. Moreover, as musculoskeletal conditions may have either an acute or a gradual onset and their outcomes vary from complete health recovery to chronic progressive course, incidence could be more relevant to measure frequency for some conditions whereas prevalence is the only measurement available for others⁴.

The use of standardized criteria to identify cases of disease is critical to make meaningful comparisons between studies. Thus, since 2003, certain basic requirements were made by the Scientific Group on the Burden of Musculoskeletal Conditions of the *World Health Organization* to access data that can be compared across musculoskeletal conditions⁴. These requirements included the use of agreed definitions for each condition in all future studies; the use of agreed age bands for reporting data; the collection of data separated by gender and the development of guidelines for uniform data collection.

Also in 2003, the *Department of General Practice and Community Medicine of the University of Oslo* recommended indicators and sources of information which can be used across the European Community to measure and monitor the impact of musculoskeletal conditions, as unspecified musculoskeletal conditions (widespread and localized), rheumatoid arthritis, osteoarthritis, and osteoporosis, at a national and community level⁷.

Methodological quality appraisal

Overall the methodological quality and reporting quality of the studies included in our review was heterogeneous. Few studies collected data from a representative sample of the population, used data-collection strategies that would minimize the risk of bias or described measures to address potential sources of bias.

Publication bias was not assessed in this review. Since these studies focus on prevalence or incidence estimates and not effect sizes, there is no reason to believe that they would be subject to publication bias.

Conclusion

There is a broad spectrum of health data generated by instruments of heterogeneous quality for this group of diseases. Theoretically, taken toge-

ther, these could allow to characterize, directly and indirectly, the burden of musculoskeletal conditions in the general population and on the Portuguese health system. However, we have identified important gaps in the knowledge about rheumatic disease epidemiology. The major gaps are related to the lack of studies estimating frequency of inflammatory arthropathies, such as gout and spondyloarthritis, and disabling pain syndromes that are not clearly defined.

In order to be able to develop preventive strategies and to measure the results of programs, such as the *National Program against Rheumatic Diseases*, it is necessary to have accurate baseline information on the present situation of rheumatic diseases occurrence. However, it is essential to take into account that, efficient and accurate frequency estimates should make use of multiple sampling frames, defined according to the frequency and the pathophysiology of each condition.

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