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Burden of disease in Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS) – a scoping review

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INTRODUCTION

- ME/CFS is a **severe and chronic multi-systemic disease**
- Symptoms** include post-exertional malaise, strong fatigue, cognitive impairments and pain
- Aetiology of the disease is still unknown, **no therapy, no diagnostic biomarkers**
- Prevalence** is estimated at 0.17% to 0.37% in Germany and 0.42% in the USA, with **women** being affected **three times** as often as men
- Increasing** prevalence as a result of the SARS-CoV-2 pandemic expected

METHODS

- Scoping review following the **PRISMA-ScR guidelines**
- Research question:** What is the burden of disease of ME/CFS?
- Search Algorithm: ME/CFS AND burden of disease
- Inclusion criteria:** any study design, only full-text & peer-reviewed publications
- Exclusion criteria:** publications in languages other than German or English
- Databases: PubMed, Web of Science, Scopus, ABI Inform Complete, CINAHL, PsycInfo, supplemented by citation searching

RESULTS

- 20 studies** published between 1996 to 2023 were included
- Studies from 6 different countries, but mainly US (n=10) and Europe (n=6)
- Reported indicators for burden were **costs** (n=16), **disability-adjusted life years (DALYs)** (n=3), **employment rates** (n=1) and **school attendance** (n=1)

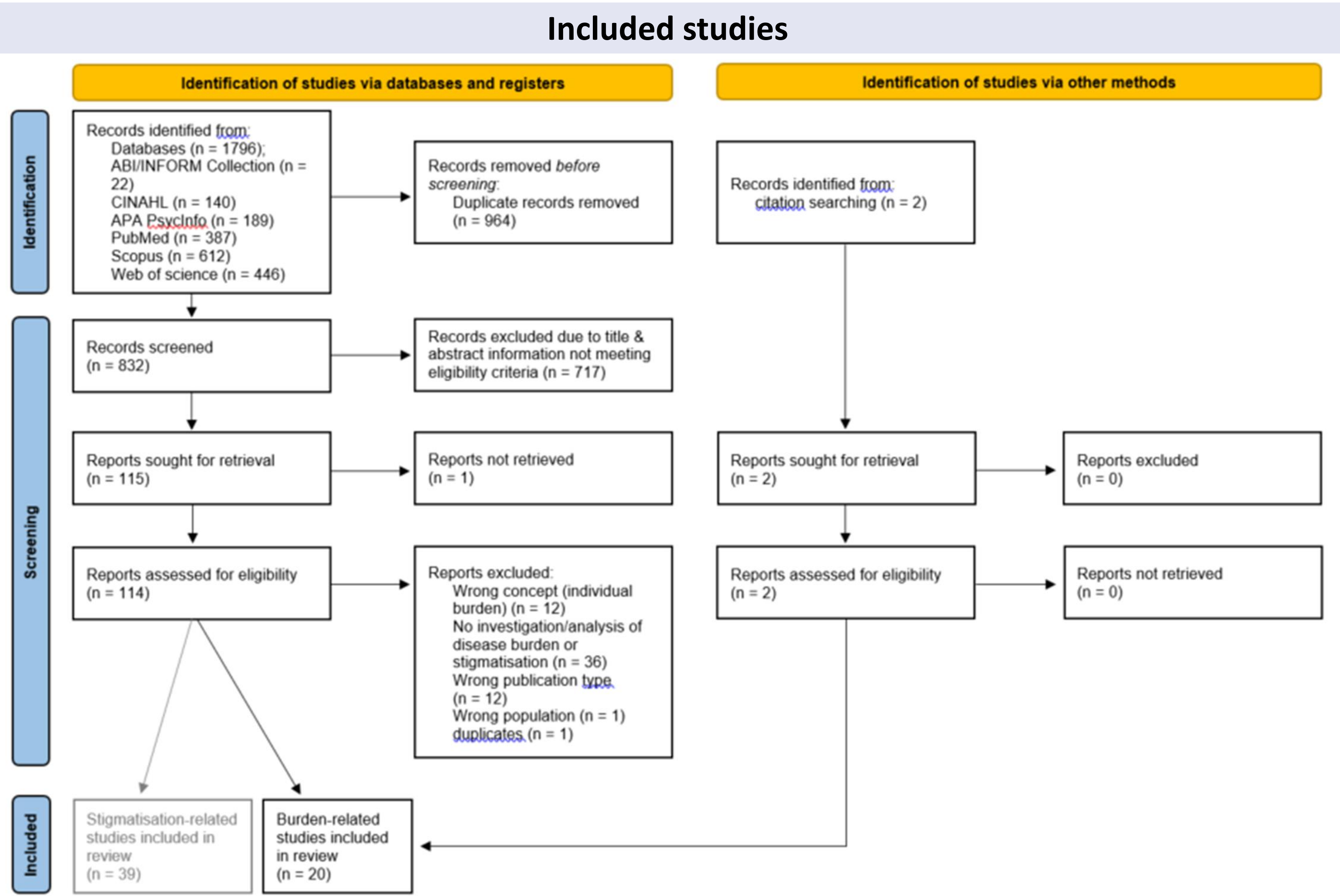


Fig. 1 PRISMA flow diagram

Quality assessment

- No tool exists to assess all different study types included in this review
- Therefore, use of a **modified version** of the Mixed Methods Appraisal Tool (MMAT) supplemented with Section A of the Critical Appraisal Skills Programme (CASP) Systematic Review Checklist
- Overall **moderate study quality**
- Quality score ranging from 0% (n=1) to 100% (n=1) with an **average quality of 63%**

DALYs

- DALYs for ME/CFS in the US population:
 - increased** from 0.714M (2013) to 0.733M (2017) in people aged ≥ 13 years
 - increased** from 0.921M (2020) to 5.77M (2022) in people aged ≥ 5 years with the last DALY based on a 10% after-COVID ME/CFS onset rate
- DALYs were **higher** than those of other diseases such as multiple sclerosis, autism, or HIV/AIDS

CONCLUSION

- ME/CFS imposes a substantial health, social and economic burden of disease
- Discrepancies in estimates are probably due to **differences** in study samples, methodologies, cost components, and health care systems
- Due to a lack of robust prevalence estimates and an assumed underdiagnosis of ME/CFS, the burden is likely still **underestimated**
- Future research is needed to reduce the burden and its impact and to improve the quality of care for individuals with ME/CFS.

Costs of ME/CFS																	
Authors	Close et al.	Zhao et al.	Araja et al.**	Araja et al.	McCrone et al.	Sabes-Figuera et al.	Collin et al.	Reynolds et al.	Jason et al.***	Jason et al.****	Jason & Mirin***	Jason & Mirin****	Lin et al.	Jeffery et al.	Valdez et al.	Mirin et al.***	Mirin et al.****
Country	Australia	Australia	Latvia	Latvia	UK	UK	UK	USA	USA	USA	USA	USA	USA	USA	USA	USA	USA
Year of study	2020	2023	2021	2021	2003	2010	2011	2004	2008	2008	2021	2021	2011	2014	2019		2022
Year to which costs relate	2019	2021	2020	2020	2000	2006	2010	2002	2005	2005	2020	2020	2005	2010	2016		2022
Total costs p.p.*	42,439	34,256	2,498	2,916	43,796	21,806	119,611	32,683	33,463	42,948	35,717	47,066	17,734	14,490	37,615	34,430	45,869
% Direct costs	30%	15%	100%	100%	6%	13%			10%	30%	12%	33%	28%	100%	100%	12%	34%
% Drug costs	9%	24%				5%	5%		49%	63%			44%	26%			
% Outpatient costs	27%	32%				62%	75%		51%	37%			41%				
% Inpatient costs	11%	4%				0%	7%						16%				
% Other costs	53%	41%				33%	13%										
% Indirect costs	70%	85%			94%	87%	100%	100%	90%	70%	88%	67%	72%			88%	66%
% Productivity losses	93%	87%				12%	70%	100%	100%	100%	100%	100%	100%			100%	100%
% Informal care costs	7%	13%				88%	30%										
Total cost per country* in M	8,129	746 - 5,452		46	56,688		529	15	27,975	35,905	40,611	57,520	116,261	760	65,383 - 130,765	149,259 - 258,953	209,886 - 363,048
Prevalence	0.76%	0.1% - 0.76%		0.80%	2.60%		0.12%	0.24%	0.42%	0.42%	0.42% (adults), 0.75% (children)	0.42%	2.50%	0.02%	0.519% - 1.038%	0.42% (adults), 0.75% (children); supplemented by 5% - 10% after-COVID ME/CFS onset rate	
Age range		≥15		17-81	16-75	16-75	18-64	18-69	≥18	≥18	≥5	≥5	18-59	18-64	0-89		≥5
*p.a., PPP & inflated in 2022 USD **included undiagnosed patients only ***community sample ****tertiary sample																	

Table 1 Costs of ME/CFS by included study

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