Prevalence and incidence of CFS/ME in Europe

EUROMENE: Working Group 1
"Epidemiology"

Belgrade, September 7th, 2017

Steps followed: 1. Systematic search

- **Databases & Keywords combination**

  - **Scopus**: ({epidemiology} OR {prevalence} OR {incidence}) AND ({chronic fatigue syndrome} OR {myalgic encephalomyelitis} OR {CFS/ME} OR {ME/CFS})

  - **Web of Science**: ("epidemiology" OR "prevalence" OR "incidence") AND ("chronic fatigue syndrome" OR "myalgic encephalomyelitis" OR "CFS/ME" OR "ME/CFS")

Steps followed: 1. Systematic search

- **Databases & Keywords combination**

  - **Pubmed**: ("Fatigue Syndrome, Chronic"[Mesh] AND (("Incidence"[Mesh] OR "Epidemiology"[Mesh] OR "epidemiology" [Subheading]) OR "Prevalence"[Mesh] OR "Cross-Sectional Studies"[Mesh]))

Steps followed: 1. Extended search

- References of included papers
- Citations of included papers
- EUROMENE survey by email

Steps followed: 2. Exclusion criteria

- Review
- Non-European studies
- Biased samples (e.g., **vaccines, virus infection**)
- Secondary or tertiary care (i.e., high-risk groups)
- Innappropriate case definition (e.g., **Oxford criteria**, **CFS-like illness**)
- Children and adolescents
- Double report

Example of exclusion

The epidemiology of chronic fatigue in the Swedish Twin Registry

BIRGITTA EVENGÅRD[1], ANDREAS JACKS[1], NANCY L. PEDERSEN[2,3]

Assessment of chronic fatigue

When the telephone interview for chronic fatigue was designed in 1996–1997, no generally recognized assessment instrument was available. Therefore, we designed a screening module for chronic fatigue that closely emulated the CDC consensus criteria for CFS (Fukuda *et al.* 1994). The following data were collected. The stem question was 'Have you felt abnormally tired during the last six months?' and defined fatigue. The time-frame was the 6 months prior to interview as assessment of lifetime fatigue was believed to be considerably less reliable. Only subjects who endorsed this item were asked further questions. Subjects were then asked about the continuousness of fatigue in the previous 6 months and about the duration of continuous fatigue. Impairment was considered present if subjects considered themselves 'too tired to live a normal life', that fatigue had caused social problems, or that fatigue had caused $\geqslant 25\%$ work incapacity. Finally, subjects were asked about eight ancillary symptoms during the period of abnormal tiredness (substantial impairment in short-term memory or concentration; sore throat; tender lymph nodes; muscle pain; multi-joint pain without swelling or redness; headaches of a new type, pattern, or severity; unrefreshing sleep; and post-exertional malaise lasting more than 24 hours). The presence of $\geqslant 4$ of these ancillary symptoms are an integral part of the definition of CFS (Fukuda *et al.* 1994).

Steps followed: 3. Quality assessment

- **Tool**:
  - Joanna Briggs Institute-Checklist for Prevalence Studies

- **Main advantage**:
  - Short and easy to apply

- **Main disadvantage**:
  - Not widely used

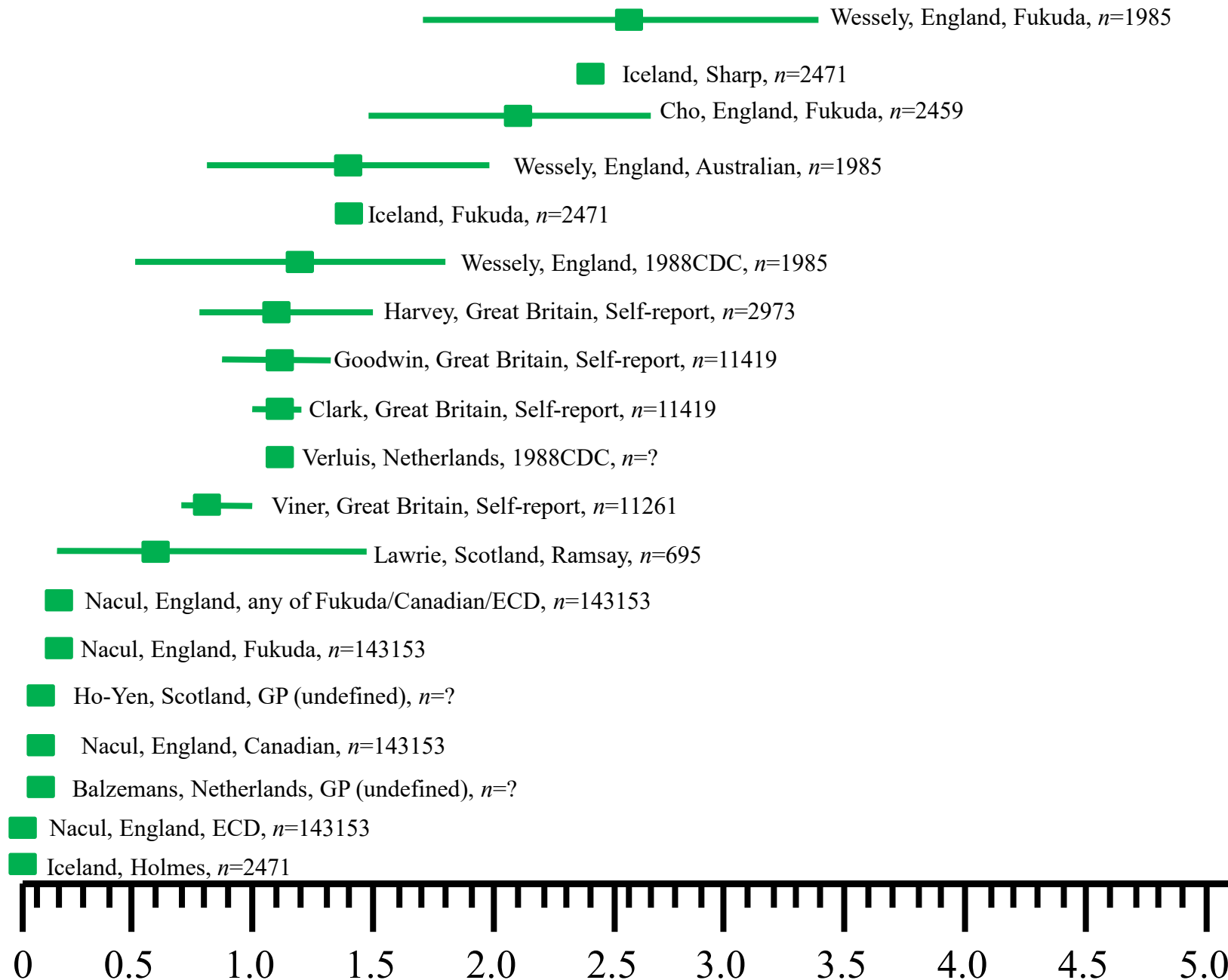|	Yes	No	Unclear	Not applicable
1. Was the sample frame appropriate to address the target population?	☐	☐	☐	☐
2. Were study participants sampled in an appropriate way?	☐	☐	☐	☐
3. Was the sample size adequate?	☐	☐	☐	☐
4. Were the study subjects and the setting described in detail?	☐	☐	☐	☐
5. Was the data analysis conducted with sufficient coverage of the identified sample?	☐	☐	☐	☐
6. Were valid methods used for the identification of the condition?	☐	☐	☐	☐
7. Was the condition measured in a standard, reliable way for all participants?	☐	☐	☐	☐
8. Was there appropriate statistical analysis?	☐	☐	☐	☐
9. Was the response rate adequate, and if not, was the low response rate managed appropriately?	☐	☐	☐	☐

Results: 1. Flow-chart
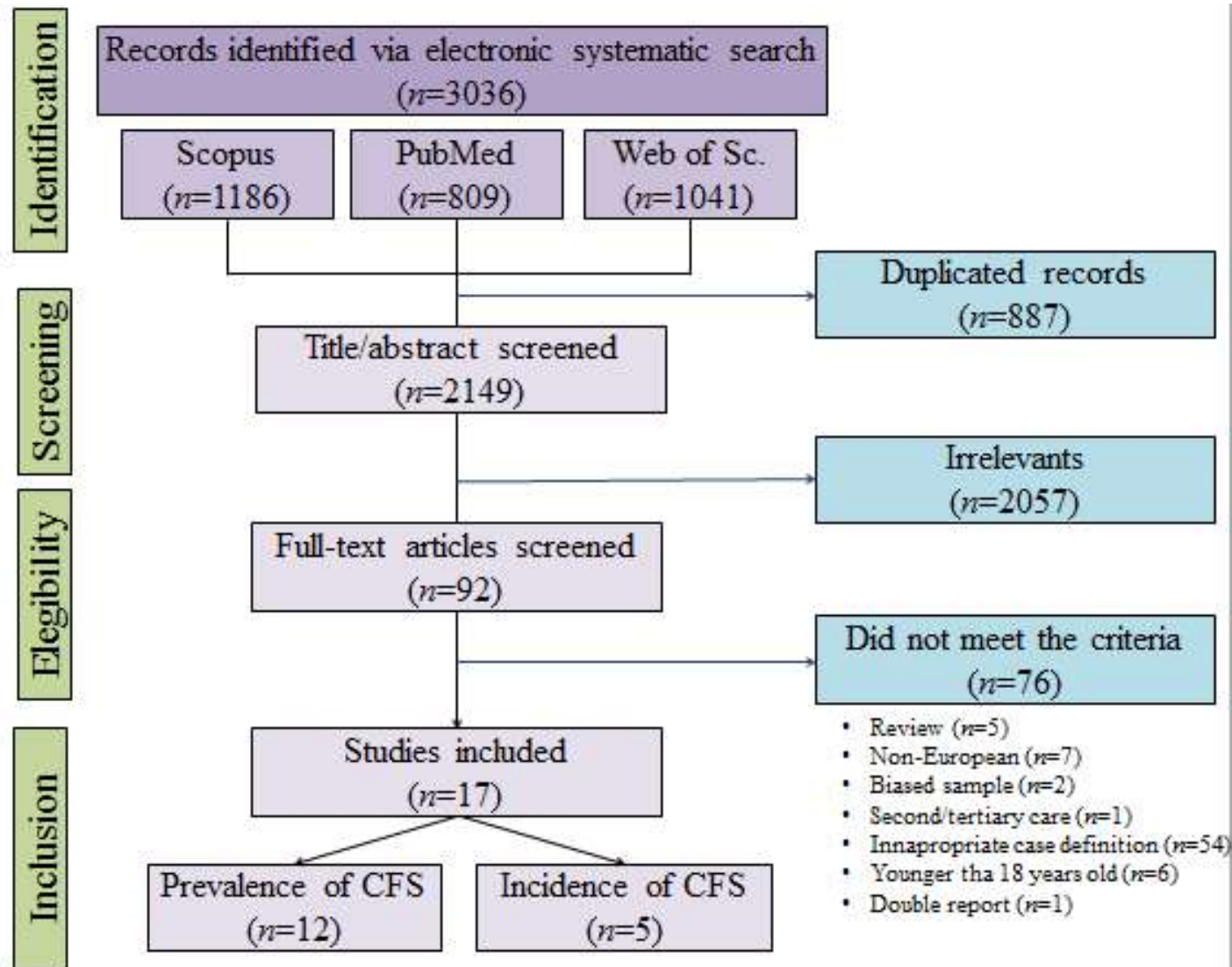
Results: 1. Flow-chart
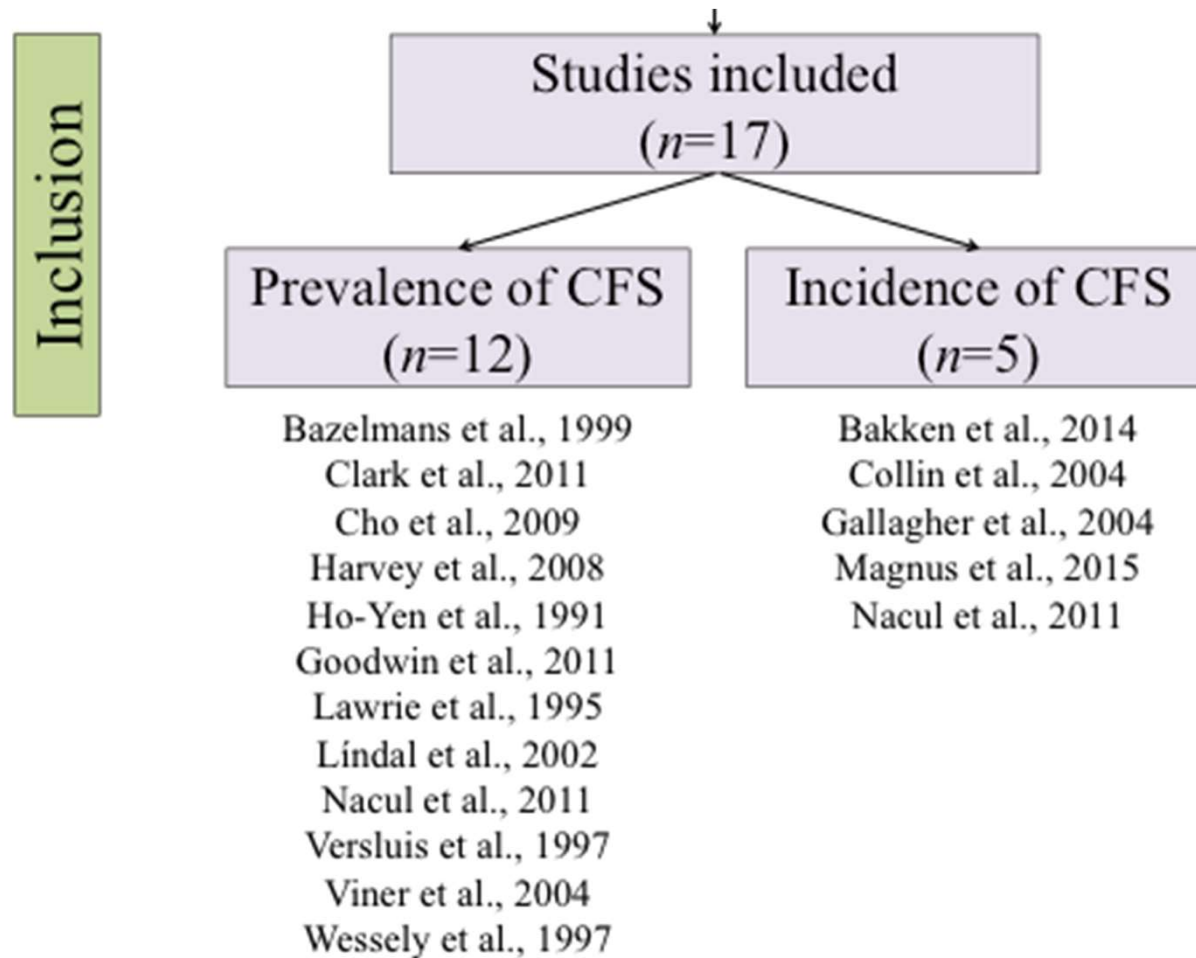
Results: 2. Prevalence

Point prevalence (%) of CFS/ME in Europe

Iceland, Lloyd, *n*=2471

Wessely, England, Fukuda, *n*=1985

Iceland, Sharp, *n*=2471

Cho, England, Fukuda, *n*=2459

Wessely, England, Australian, *n*=1985

Iceland, Fukuda, *n*=2471

Wessely, England, 1988CDC, *n*=1985

Harvey, Great Britain, Self-report, *n*=2973

Goodwin, Great Britain, Self-report, *n*=11419

Clark, Great Britain, Self-report, *n*=11419

Verluis, Netherlands, 1988CDC, *n*=?

Viner, Great Britain, Self-report, *n*=11261

Lawrie, Scotland, Ramsay, *n*=695

Nacul, England, any of Fukuda/Canadian/ECD, *n*=143153

Nacul, England, Fukuda, *n*=143153

Ho-Yen, Scotland, GP (undefined), *n*=?

Nacul, England, Canadian, *n*=143153

Balzemans, Netherlands, GP (undefined), *n*=?

Nacul, England, ECD, *n*=143153

Iceland, Holmes, *n*=2471

0   0.5   1.0   1.5   2.0   2.5   3.0   3.5   4.0   4.5   5.0

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Great Britain (as a whole or single countries)

| 0 | 0.5 | 1.0 | 1.5 | 2.0 | 2.5 | 3.0 | 3.5 | 4.0 | 4.5 | 5.0 |

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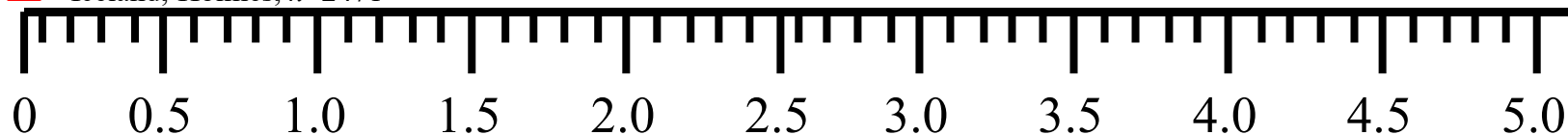Nacul, England, Fukuda, *n*=143153

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Nacul, England, ECD, *n*=143153

Iceland, Holmes, *n*=2471

Other EU countries

0   0.5   1.0   1.5   2.0   2.5   3.0   3.5   4.0   4.5   5.0

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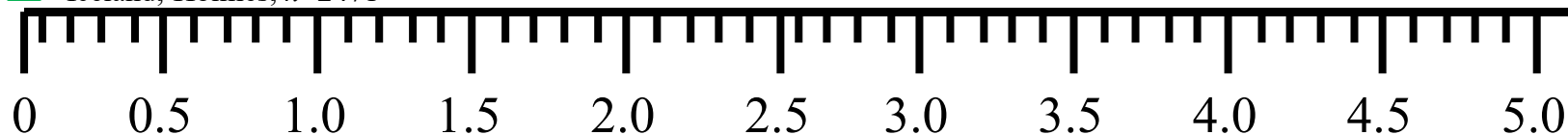Nacul, England, Fukuda, *n*=143153

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Iceland, Holmes, *n*=2471

Fukuda case definition

0   0.5   1.0   1.5   2.0   2.5   3.0   3.5   4.0   4.5   5.0

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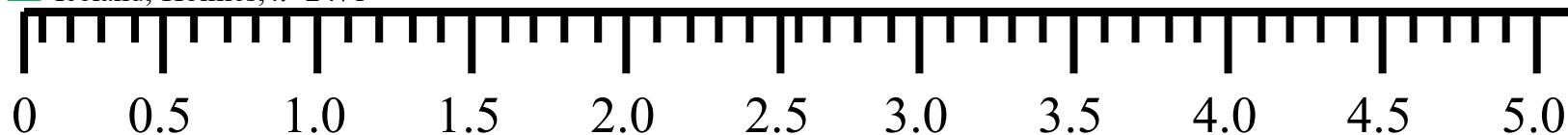Nacul, England, Fukuda, *n*=143153

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Iceland, Holmes, *n*=2471

Self-reported CFS/ME

0   0.5   1.0   1.5   2.0   2.5   3.0   3.5   4.0   4.5   5.0

Results: 3. Incidence

Ref Adults studies	Country	Sample, total (women, %)	Follow-up (months)	Case definition	Incidence (95% CI); cases per 100000 person-years)[1]
Gallagher et al., 2004	UK	2,400,000	84	Read coding	10 (n/a) for females 4 (n/a) for males
Nacul et al., 2011	England	143,153 (n/a)	12	Any of the following: 1994 CDC/Fukuda, 2003 Canadian, or ECD	15.0 (n/a)
Collin et al., 2017	UK	n/a (n/a)	156	Read coding	14.8 (14.5 to 15.1)
Bakken et al., 2014	Norway	22,173,710 (50%)	60	ICD-10 coding	25.8 (25.2 to 26.5)
Magnus et al., 2015	Norway	4,822,337 (50%)[2]	38	ICD-10 coding	24.96 (n/a)

Results: 4. Quality prevalence studies

Author (year)	P1	P2	P3	P4	P5	P6	P7	P8	P9	Score
Lawrie SM et al. (1995)	1	1	1	1	UN	1	1	1	1	8
Wessely S et al. (1997)	1	1	1	1	1	1	1	1	1	9
Viner R et al. (2004)	1	1	1	1	1	0	1	1	1	8
Harvey SB et al. (2008)	1	1	1	1	1	0	1	1	1	8
Ho-Yen DO et al. (1991)	1	1	1	UN	0	0	0	0	1	4
Nacul LC et al. (2011)	1	1	1	1	1	1	1	UN	1	8
Clark C et al. (2011)	1	1	1	1	1	1	1	1	1	9
Goodwin L et al. (2011)	1	1	1	1	1	1	1	1	1	9
Cho HJ et al. (2009)	1	1	1	1	1	1	1	1	1	9
Líndal E et al. (2002)	1	1	1	UN	0	0	0	0	1	4
Bazelmans E et al. (1997)	1	1	1	0	0	0	0	0	1	4
Versluis RG et al. (1997)	PAPER NOT FOUND									

Results: 4. Quality incidence studies

Author (year)	P1	P2	P3	P4	P5	P6	P7	P8	P9	Score
Bakken IJ et al. (2014)	1	1	1	1	1	1	N/A	1	1	8
Magnus P et al. (2015)	1	1	1	1	1	1	N/A	1	1	8
Gallengher AM et al. (2004)	1	1	1	UN	1	1	1	UN	1	7
Nacul LC et al. (2011)	1	1	1	1	1	1	1	UN	1	8
Collin SM et al. (2017)	1	1	1	1	1	1	1	1	1	9

Future: Do we want to improve the review?

- If yes,
    - Prospero registration (final design)
    - Double check (e.g., review, data extraction)
    - Ask authors for further details (e.g., prevalence by gender, age ranges).
    - Better quality assessment.
    - Meta-analyses: Global and specific ones.
    - **Other suggestion(s)?**

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