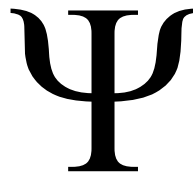




DET PSYKOLOGISKE FAKULTET



*The prevalence of orthorexia in exercising populations.
A systematic review and meta-analysis*

HOVEDOPPGAVE
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Sammendrag

Hensikt: Ortorexia nevrosa (ON) er foreslått som en ny type spiseforstyrrelse, karakterisert med en overopptatthet av å spise kvalitativt sunn mat. Per dags dato finnes det ikke systematisk litteraturoversikt eller meta-studie som viser prevalens av ON blant personer som trener regelmessig. Målet med denne studien var derfor å vurdere studier som oppgav prevalensen av ON hos personer som trente regelmessig, for å kalkulere en overordnet prevalens med en random effekt meta-analytisk tilnærming. Og undersøke assosiasjoner av ON prevalens og fysisk aktivitet, ved å bruke en random-effekt meta-regresjon.

Metode: Det ble utført systematisk litteratursøk i følgende databaser: PubMed, Embase, Web of Science, PsychInfo, CHINAL, Google Scholar and Open Net. Følgene søketermologi ble benyttet: Orthore* AND (prevalenc* OR incidenc* OR frequen* OR cut-off OR epidem*). Totalt ble 613 artikler gjennomgått av to individuelle anmeldere. 24 studier ble kodet og kvalitetsvurdert (Holy m.fl). Metaregresjons-analysen inkluderte tre uavhengige variabler (kjønn, sportstype og utvalgsstørrelse).

Resultat: Den overordnede prevalensen av ON var 55,3% (95% CI = 43.2 – 66.8). Cochran Q var 11,436.38 ($df = 23$, $p < 0.0000$), og I^2 var 98.4%, som indikerte en høy heterogenitet på tvers av studiene. Ingen av de uavhengige variablene (kjønn, sportstype og utvalgsstørrelse) var signifikant relatert til variasjonen i ON prevalens.

Diskusjon: Prevalensen av ON blant fysisk aktive populasjoner var svært høy. Forskjellen mellom studiene var stor, men kunne ikke forklares av de inkluderte moderatorene. En fjerdedel av studiene hadde moderat risiko for skjevhet. Flertallet av studiene spesifiserte ikke relevant demografisk informasjon om utvalget, og informasjonen om sportstype var ofte ikke oppgitt.

Abstract

Aim: Orthorexia nervosa (ON) is a proposed type of eating disorder that is necessary to research further. There are currently no systematic reviews or meta-analyses concerning the prevalence of ON in the exercising population. The aim of this study was to review studies that report the prevalence of ON in people who exercise, to calculate an overall prevalence by a random effects meta-analysis approach, and investigate the association of ON prevalence and physical activity using a random-effects meta-regression.

Method: Systematic searches were conducted in the following online databases: PubMed, Embase, Web of Science, PsychInfo, CHINAL, Google Scholar and OpenNet. The following

search terms were used: Orthore* AND (prevalenc* OR incidenc* OR frequen* OR cut-off OR epidem*). 613 unique hits were reviewed of two blinded authors. 24 studies were coded and assessed for risk of bias (Holy et.al). The meta-regression included three independent variables (sex, type of sport, and sample size).

Results: The overall prevalence of ON in the exercising population was 55.3% (95% CI = 43.2–66.8). Cochran's Q was 11,436.38 ($df = 23$, $p < 0.0000$), and the I^2 was 98.4%, indicating high heterogeneity across studies. None of the independent variables were related to ON prevalence.

Discussion: The overall prevalence of ON in exercising populations was very high. The between-study disparity was large but could not be explained by the proposed moderators. One fourth of the studies had moderate risk of bias. The majority of the studies did not specify relevant demographic information about the sample, and information about the type of sport was frequently missing.

Keywords: Review, Meta-regression, Meta-analysis, Orthorexia, Prevalence, Frequency, Exercise, Eating disorder

Plain English summary

Orthorexia nervosa (ON) is a proposed type of eating disorder characterized by an excessive preoccupation with eating healthy food of high quality. This proposed disorder is found among sports athletes in some studies, and there is an assumed link between ON and physical activity in general. In this systematic review and meta-analysis we were looking at the overall prevalence of ON in the exercising population. We searched in scientific databases and ended up with 24 of 613 articles that met the rigorous inclusion criteria. We found that the overall prevalence of ON in the general exercising population was 55.3%. This prevalence was thought to be predictable by sex, type of sport, and sample size, but we found no significant associations. This may be due to poor study quality and to lack of demographic information of the participants in some of the included studies. Other predictors worth investigating might be age, student status, status as vegetarian/vegan, psychiatric comorbidity, and level of physical activity. This knowledge can help prevent, and provide the right treatment for people at risk of developing ON.

Introduction

Orthorexia nervosa (ON), literally meaning “correct appetite”, is a relatively new type of eating disorder and was named by Steven Bratman in 1997 (1). He observed that some people became obsessive and dysfunctional in their way of trying to eat a perfect diet. In the book *Health Food Junkies* (2) he describes this disorder in detail. Orthorexics typically restrict their food consumption according to what they believe is pure and healthy because their main motivation is to achieve “optimal health” (3). However, such food restrictions may lead to nutritional deficiencies, malnourishment, and unwanted/unhealthy weight loss because of their elimination of certain food groups and nutrients (4, 5). In 2004 the first peer-reviewed article about ON was published in which a tool, ORTO-15, to measure the disorder was presented (6). Today the ORTO-15 and the Orthorexia Self-Test (also known as the Bratman Orthorexia Test) (7) are the most commonly used ON instruments.

Ten years after the first peer-reviewed paper were published, Moroze and colleagues proposed the first diagnostic criteria (8). Several definitions and diagnostic criteria for ON have since been proposed, but consensus has so far not been reached and ON is yet to be included as a formally recognized disorder in any psychiatric diagnostic system (9). Papers about its prevalence and its correlates as well as case-studies seemed to make up a major proportion of the academic literature regarding ON (8, 10-12).

As eating disorders in general are well known in the sport and exercise population (13-17), it comes as no surprise that instances of ON are also linked to peoples’ physical activity level (18-21). Physical activity and good diet are both regarded as important factors in achieving optimal physical health, and therefore the association between physical activity and ON has been studied and documented extensively (13). Despite the fact that there is a link between exercise and ON (13), prevalence studies of ON in sport and exercising populations present large discrepancies in terms of results (22).

In the same way that eating disorders in general have a higher incidence and prevalence in certain groups, it is conceivable that the same also applies to ON. It has been shown that women are at higher risk of eating disorders than men (23) based on the assumption that women, compared to men, add greater value to their physical appearance. For many women a thin and slim body is the ideal, and this is fuelled by social media exposure to retouched bodies and models (24, 25). Thus, a higher prevalence of ON is expected to be found among women.

Because eating disorders are often linked to a distorted self-image (26), it is likely that those in question have a strong focus on themselves. Studies suggest that athletes in

individual sports suffer more frequently from stress, anxiety, and depression than those involved in team sports, probably because individual athletes are more concerned about goals, whereas team sport members are more motivated by having fun. In addition, the performance of individual athletes is more under scrutiny than that of team sport members (27, 28). Based on the above, we assumed that ON occurs more frequently in individual sports or types of exercise than in team sports and exercises.

Although sample size probably affects estimates less often in prevalence studies than in trial studies, there is still some evidence to suggest that small study effects may influence prevalence estimates (29). Hence, sample size should be investigated as a potential moderator for prevalence estimates across ON studies.

As mentioned above, the prevalence of ON in exercising/sports samples seems to vary considerably. So far, no systematic review or meta-analysis of the ON prevalence within this population has been conducted, although a meta-analysis can provide a quantitative synthesis of the individual findings as well as identify potential variables moderating the prevalence figures across studies. Against this backdrop we conducted a meta-analysis of all prevalence studies of ON in exercising/sports samples. In addition, a meta-regression including sex, type of sports (individual, team, mixed/unknown), and sample size was conducted to identify potentially relevant moderators.

Methods

Protocol and guidelines

This systematic review and meta-analysis was pre-registered with PROSPERO (CRD42022301749) and adhered to the guidelines found in the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) procedure (30, 31) as well as the recommendations of the Meta-analysis of Observational Studies in Epidemiology (MOOSE) (32). The first PROSPERO registration for this metanalysis only covered “athletes” as a sample, but it was changed to “exercising populations” in general due to extremely few studies. Figure 1 presents the literature search and selection process. See Appendix A for a completed PRISMA-guideline checklist (31).

Systematic search strategy

Systematic searches and a comprehensive literature search were conducted in five electronic databases –PubMed, Embase, Web of Science, PsychInfo, and CHINAL. In addition, we searched through Google Scholar and OpenNet in order to identify potential grey literature in

the field. The literature searches were conducted between January 20, 2022, and February 1, 2022. The following keywords were used: Orthore* AND (prevalenc* OR incidenc* OR frequen* OR cut-off OR epidem*). Reference lists of included articles were further hand-searched for identification of relevant articles for inclusion. No restrictions in terms of time frame were used. The search strategy and the keywords were approved by a librarian at the University of Bergen.

A total of 1,300 hits (including the first 200 hits in Google Scholar) were identified from the database search. First, all the hits were transferred to Endnote Online X9 – which automatically removed duplicates. Second, the remaining hits were transferred to the Covidence software (33) that was used for the remaining screening and data extraction. After removing duplicates (by software and manually) a total of 76 articles were assessed for eligibility. Of this pool a total of 24 articles met the inclusion criteria and were consequently included in the meta-analysis.

Study selection criteria

The key inclusion criteria for the articles in this systematic review and meta-analysis were as follows. 1) The study informed about the prevalence of ON among athletes or people who exercise frequently. “Frequent exercisers” were defined as subjects who exercised at least once week, and all types of physical activities and sports were included. Students studying sports-related themes were included based on the assumption that a high interest in sports most often presupposes to being active in sports participation. See reference (34) for the definition of athletes used. 2) The study presented original data on the prevalence of ON. 3) The study was published in any European language. 4) The study used a tool for measuring ON where the procedure provided a categorization of pathological ON (e.g., based on cut-off scores or an interval that was categorized as either “ON”, “risk of ON”, or “tendencies of ON”. In the analysis we used the most liberal cut-off scores (the scores that provided the highest prevalence) if the study reported more than one. This decision was based on the most liberal cut-off score for the ORTO-15 as recommended by the scale constructors (35).

The exclusion criteria were as follows: 1) participants stemming from a clinical sample only, 2) studies reporting the prevalence of “healthy orthorexia”, 3) studies only reporting mean scores on ON measures, hence failing to report proportions/percentages scoring above cut-offs/categorization of ON, and 4) studies based on qualitative data, case studies, interviews, case reports, or reviews. Two reviewers (SMH and JB) preformed title/abstract and full text screening independently of each other. A third reviewer (SP)

participated in the final discussion about the included articles. Discrepancies were resolved through discussions.

Data extraction

The following study and participant characteristics were extracted from the identified studies and coded into a data extraction template, including author, year of publication, country and continent, study type, sample type, type of selection, sex, age range (mean \pm SD), age category, total sample size, percentage exercisers, percentage women, frequencies of exercising, type of ON measurement, cut-off / score-interval used for measurement of ON, type of sport (individual, team, mixed/unknown), prevalence of ON, and response rate.

In the categories “country and continent”, we stated the country/continent of the origin of the manuscript and for the participants. If several countries/continents were mentioned, we selected all that applied.

In the category “sample type”, we chose between the following four options: 1) general population, 2) students, 3) athletes/exercisers, and 4) others. A specific category was scored if 75% or more of the sample consisted of that category. If the sample consisted of several categories, then all the categories could be coded for the respective study. We defined the sample as athletes or exercisers if the sample fit with the definition of athletes being used (34), if the sample was defined as yoga-practitioners, if the whole sample was selected from a fitness-gym or a CrossFit-centre, or if the article authors specifically named the sample as athletes. If the study did not provide demographic information for the exercising population, we used the information about the whole sample.

The category of age contained five different age-groups: 1) adolescents 15–18 years, 2) young adults 18–34 years, 3) adults 35–64 years, 4) older adults 65+ years, and 5) mixed ages. The sample ended up in one of the first four categories if 75% or more belonged to one specific age category. The category “type of selection” had two options: 1) random population sampling and 2) non-random sampling.

The category “percentage exercisers”, includes the proportion of the total sample was categorized as frequent exercisers/athletes if the participants performed a sport, or a specific exercise form (e.g., yoga) for at least once a week they were categorized as exercisers/athletes. Participants who specifically reported that they rarely or never performed any physical activity were excluded from the “exercising sample”. Participants reporting to be physically active or very active were categorized within the exercising sample. Students

called “students athletes” or students belonging to a field of sports studies at a university were categorized as athletes/exercisers and were included in the exercising sample.

In the category “frequency of exercising”, we chose between four levels: 1) exercising 1–3 times a week, 2) exercising 4–6 times a week, 3) athletes, or 4) unknown exercise frequency. The sample were categorized in one of the first three categories if 75% or more of the sample belonged to that specific category.

In the category “type of sport” we differed between three codes. 1) Individual sport – which contained athletes/exercisers such as runners, dancers, gymnastics, boxers, wrestlers, martial artists, cyclists, figure skaters, Olympic weightlifters, powerlifters, body-builders, and CrossFitters. Also people belonging to a fitness gym or yoga centre were included in this category on the basis that they do not work together on a team to achieve a goal. 2) Team sport – which contained every sport where one performs as a team, e.g., football, hockey, rugby, netball, cheerleading, lacrosse, baseball, handball, basketball, or floorball. 3) Mixed/unknown – which was used if both individual and team sports athletes/exercisers were included or if the study did not inform about which type of sport/physical activity the participants performed. The athletes/exercisers ended up in one of the two first categories if 75% or more of the sample consisted of that category.

The extraction was conducted independently by two reviewers (SMH and JB). Disagreements were resolved through discussions.

Risk of bias assessment

All the included studies were assessed for risk of bias using a modified quality assessment checklist for population-based prevalence studies developed by Holy et.al. (36). This risk of bias assessment contains 10 items reflecting different characteristics of the included articles aiming at evaluating their internal and external validity. Each of the 10 items was scored either “yes” (0-points, low risk of bias) or “no” (1-point, high risk of bias). High risk of bias was indicated as follows: 1) the target population was not representative of the national population, 2) the sampling frame was not representative of the overall target population, 3) the sample was not randomly selected, 4) the response rate was less than 65%, 5) data were collected from a proxy, 6) no acceptable definition or delimitation of ON was used, 7) the ON measurement instrument was not shown to have reliability or validity, 8) the same mode of data collection was not used for all participants, 9) the shortest prevalence period for the parameter was not suitable, and 10) the numerator(s) or denominator(s) were not suitable. Based on the 10 items, a composite risk of bias score was calculated: high risk of bias (7–10

points), moderate risk of bias (4–6 points), or low risk of bias (0–3 points). Two reviewers (SMH and JB) conducted the risk of bias assessment independently of each other, and in case of disagreement consensus was reached through discussions.

Data synthesis and analysis

For all types of coding (inclusion/exclusion, study characteristics extraction, and risk of bias evaluation), percentages of initial agreement between the two reviewers were calculated. The prevalences and the corresponding 95% confidence intervals (95% CIs) of the included studies were synthesized using a random-effects model, which did not assume that the included studies came from the same population of studies (37). The between-study variance was estimated by using the Der Simonian and Laird approach (38). Heterogeneity was assessed based on Cochran's Q and I^2 statistics, the latter of which reflects the proportion of variation in the observed effects that is due to variation in true effects (39). According to Higgings et al. (40) an I^2 of 0% suggests no heterogeneity, 25% suggests low heterogeneity, 50% suggests medium heterogeneity, and 75% suggests high heterogeneity. Publication bias was investigated by the Egger test (41) and Duval and Tweedie's trim and fill procedure. The latter is based on a funnel plot with the largest and most precise studies situated at the top (y-axis) of the funnel plot with the effect size situated along the x-axis. The trim-and-fill procedure trims off asymmetric outlying studies and replaces them with studies around the centre, whereupon an adjusted effect size and 95% CI are calculated (42). Finally, a random-effects meta-regression analysis was conducted to examine whether the following predictors explained heterogeneity in ON prevalence: a) percentage women, b) type of sport (1 = individual, 2 = team, 3 = mixed/unknown, where the latter category comprised the reference), and c) sample size. The meta-analysis and the meta-regression analysis were conducted using the Comprehensive Meta-Analysis 3.0 software (43).

Availability of data, code and other materials

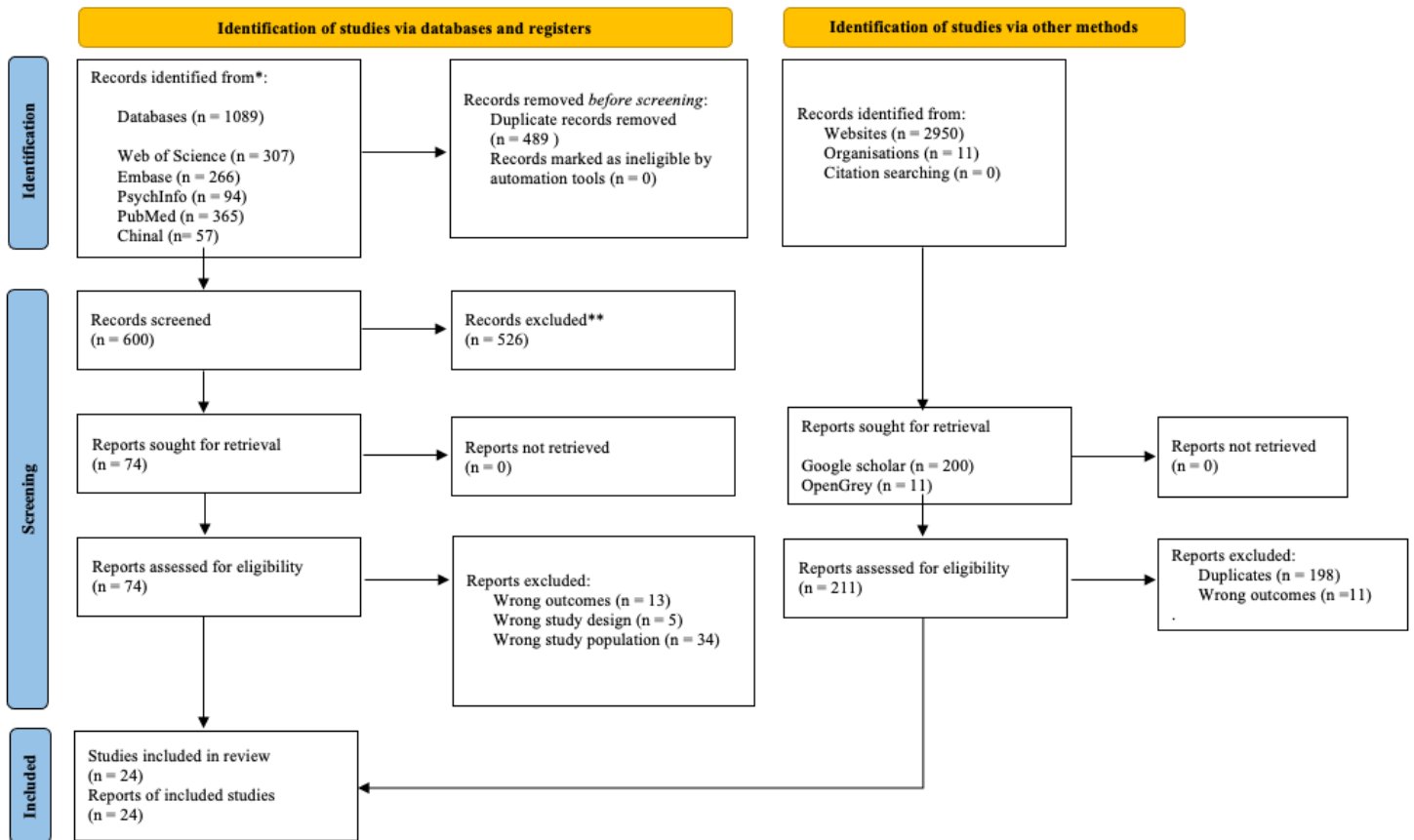
Data extracted from included studies; data used for all analyses; analytic code; any other materials used in the review can be provided by the corresponding author (stine.hafstad@hotmail.com).

Figure legends

FIGURE 1. PRISMA 2020 (30) flow diagram for new systematic reviews for ON prevalence.

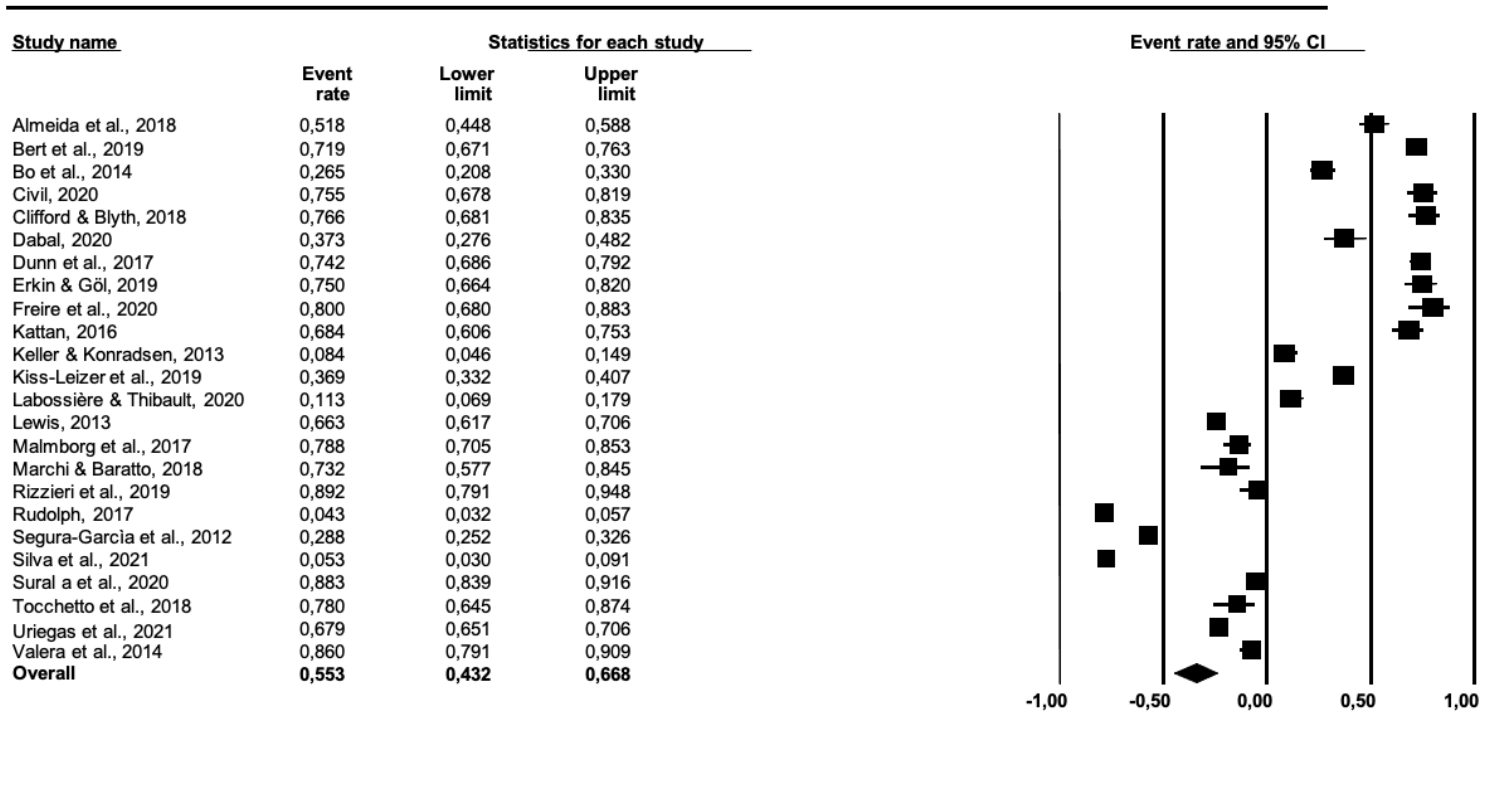
FIGURE 2. Forest plot of the included studies.

FIGURE 1



NOTE: There were found 600 unique hits from databases, and 13 unique hits from grey literature. 74 studies from databases, and 2 from grey literature were screened in full-text.

FIGURE 2



Results

Description of the studies

Of the 24 included studies, publication years ranged from 2012 (44, 45) to 2021 (46, 47). Studies were conducted in Portugal ($k = 4$: (19, 48-50)), Italy ($k = 3$: (18, 51, 52)), the US ($k = 3$: (22, 44, 46)), Brazil ($k = 2$: (47, 53)), Turkey ($k = 2$: (54, 55)), Poland ($k = 2$: (56, 57)), and one study each from the UK (3), Germany (58), Sweden (59), Denmark (45), Hungary (60), Lebanon (61), Spain (62), and Canada (63).

Samples were mostly recruited from fitness gyms, sports universities, or specific groups of athletes ($k = 17$: (3, 18, 19, 44-49, 52-56, 58, 62, 63)). For six studies the sample was recruited from regular universities (students) (22, 50, 51, 57, 59, 61), and one study was based on a general population sample (60). All of the studies had cross-sectional study designs and used a non-random selection method – except for one study that employed a random selection of participants (61). All of the studies were peer-reviewed research papers, except for two studies that were theses (one master theses (61) and one doctoral theses (44)) and one study published in a journal for Danish nurses (45).

The majority of studies ($k = 18$: (3, 18, 19, 22, 44, 46, 48-53, 56, 57, 59, 61-63)) assessed ON using the ORTO-15 questionnaire (6). In addition, three studies (54, 55, 60) used the ORTO-11 questionnaire (64), one study (45) used the Orthorexia Screen (65), one study (58) used the Düsseldorf Orthorexia Scale (66), and one study (47) used the Teruel Orthorexia Scale.

The studies included a total of 7,592 participants, ranging from 41 (50) to 1,090 (46) respondents with a mean of 316 (SD = 3) participants. In total 4,288 participants were females, and 3,304 were males. **Table 1** presents further characteristics of the included studies.

Prevalence estimates and heterogeneity

The results of the meta-analysis are presented as a forest plot (**Figure 2**). The overall prevalence across all 24 studies was 55.3% (95% CI = 43.2–66.8). Cochran's Q was significant ($Q = 1,436.38$, $df = 23$, $p < 0.0000$), suggesting heterogeneity across the prevalence estimates, and the I^2 statistic was 98.4%, indicating very high heterogeneity.

Correlates of ON prevalence

Because of the significant heterogeneity, a meta-regression analysis based on a random-effects model was conducted including percentage female, type of sport (individual = 0, team

= 1, mixed/unknown = 2), and sample size as predictors. The results are present in **Table 3**.

Overall, the regression model was not significant ($Q = 5.97$, $df = 4$, $p = 0.2011$, pseudo $R^2 = 0\%$).

Percentage female ($b = 0.018$, $p = 0.23$), individual sport and team sport ($b = -0.442$, $p = 0.43$, $b = 0.658$, $p = 0.511$; mixed/unknown comprised the reference), and sample size ($b = -0.002$, $p = 0.09$) were accordingly unrelated to ON prevalence among the exercise populations.

Publication Bias

The results of the Egger test ($b = 0.312$, 95% CI = $-6,891$ to $7,52$, $t = 0.090$, p 1-tailed = 0.465 , p 2-tailed = 0.929) did not suggest publication bias. The funnel plot suggested a minor deviance from symmetric distribution, thus suggesting a lack of studies to the left of the distribution. The Duwal and Tweedies' trim-and-fill procedure trimmed three studies and consequently implied a minor change in the overall estimated prevalence (51.4%, 95% CI = 39.6%–63.9%)

TABLE 1. Characteristics of prevalence studies of exercise populations.

Study ID	Country	Selection	Sample type	Age range	Age Mean \pm SD	ON instrument	Cut-off	N	% Female	Sport type	ON prev%	RR %
Almeida et al., 2018	Portugal	Non-random	Exercisers	18-59	32.8 \pm 11.6	ORTO-15	35	193	58.5	Individual	51.8	100
Bert et al., 2019	Italy	Non-random	Athletes	18-40	26.5 \pm 5.4	ORTO-15	40	367	25.5	Mixed/unknown	71.9	100
Bo et al., 2014	Italy	Non-random	Students	Not provided	19.9 \pm 1.8	ORTO-15	35	200	32.5	Mixed/unknown	26.5	87.0
Civil, 2020	Turkey	Non-random	Athletes	18-37	22.9 \pm 4.3	ORTO-11	27	143	31.5	Individual	75.5	100
Clifford & Blyth, 2018	UK	Non-random	Students & athletes	18-27	21 \pm 1.0	ORTO-15	40	116	58.0	Team sport	76.6	100
Dabal, 2020	Poland	Non-random	Students	18-35	23.9 \pm 4.7	ORTO-15	35	83	49.6	Mixed/unknown	37.3	100
Dunn et al., 2017	USA	Non-random	Students	Not sited	21.7 \pm 4.8	ORTO-15	40	264	68.0	Individual	74.2	93.5
Erkin & Göl, 2019	Turkey	Non-random	Athletes	18-56	30.5 \pm 9.2	ORTO-11	27	118	92.4	Individual	75.0	77.1
Freire et al., 2020	Brazil	Non-random	Exercisers	18-34	26.6 \pm 7.8	ORTO-15	40	60	63.3	Individual	80.0	100
Kattan, 2016	Lebanon	Random	Students	18-30	21.7 \pm 1.9	ORTO-15	40	152	84.0	Mixed/unknown	68.4	100
Keller, 2013	Denmark	Non-random	Exercisers	16-29	22 \pm 3.4	OS	7	119	53.8	Individual	8.4	100
Kiss-Leizer et al., 2019	Hungary	Non-random	General population	18-72	29.7 \pm 10.2	ORTO-11	14-19	635	79.2	Individual	36.9	100
Labossière & Thibault, 2020	Canada	Non-random	Students & athletes	Not provided	21.3 \pm 1.7	ORTO-15	35	133	71.4	Mixed/unknown	11.3	31.6
Lewis, 2013	USA	Non-random	Students & athletes	18-23	20.5 \pm 1.2	ORTO-15	40	427	44.0	Team sport	66.3	71.0
Malmborg et al., 2017	Sweden	Non-random	Students	19-29	22.8 \pm 2.2	ORTO-15	40	118	54.2	Mixed/unknown	78.8	66.1
Marchi & Baratto, 2018	Portugal	Non-random	Students	18-39	21 \pm not provided	ORTO-15	40	41	93.9	Mixed/unknown	73.2	96.5
Rizzieri et al., 2019	Portugal	Non-random	Exercisers	20-59	29.9 \pm 7	ORTO-15	40	65	55.4	Individual	89.2	100
Rudolph, 2017	Germany	Non-random	Athletes	Not provided	29.4 \pm 11.6	DOS	30	1008	44.5	Individual	4.3	100
Segura-Garcia et al., 2012	Italy	Non-random	Athletes	16-45	22.3 \pm 4.7	ORTO-15	35	577	32.8	Mixed/unknown	28.8	86.8
Silva et al., 2021	Brazil	Non-random	Exercisers	18-60	27.8 \pm 5.1	TOS	50-75	226	36.3	Individual	5.3	100
Surala et al., 2020	Poland	Non-random	Athletes	14-39	20.9 \pm 4.7	ORTO-15	40	273	45.8	Mixed/unknown	88.3	100

TABLE 1. Continued

Tocchetto et al., 2018	Portugal	Non-random	Students & athletes	Not sited	23.5 ± 1.4	ORTO-15	40	50	46.0	Mixed/unknown	78	100
Uriegas, 2021	USA	Non-random	Students & athletes	18-40	19.6 ± 1.4	ORTO-15	40	1090	69.4	Mixed/unknown	67.9	98.6
Valera et al., 2014	Switzerl and	Non-random	Athletes	20-55	37 ± 6.7	ORTO-15	40	136	65.5	Individual	86.6	23.4

TABLE 2. Risk of bias/methodological quality (36) of the included studies.

Study ID	1. N-representativeness	2. N-frame	3. Randomization	4. Non-response bias	5. Primary data	6. Operationalization	7. Instrument	8. Consistency	9. Period	10. Estimation	Total Risk score	Risk Category
Almeida et al., 2018	1	1	1	0	0	0	0	0	0	0	3	Low
Bert et al., 2019	1	1	1	0	0	0	0	0	0	1	4	Moderate
Bo et al., 2014	1	1	1	0	0	0	0	0	0	0	3	Low
Civil, 2020	1	1	1	0	0	0	0	0	0	0	3	Low
Clifford & Blyth, 2018	0	1	1	0	0	0	0	0	0	0	2	Low
Dabal, 2020	0	1	1	0	0	0	0	0	0	0	2	Low
Dunn et al., 2017	1	1	1	0	0	0	0	0	0	0	3	Low
Erkin & Gül, 2019	1	1	1	0	0	0	0	0	0	0	3	Low
Freire et al., 2020	1	1	1	0	0	0	0	0	0	0	3	Low
Kattan, 2016	1	0	0	0	0	0	0	0	0	0	1	Low
Keller & Konradsen, 2013	1	1	1	0	0	0	0	0	0	0	3	Low
Kiss-Leizer et al., 2019	1	1	1	0	0	1	0	0	0	0	4	Moderate
Labossière & Thibault, 2020	1	1	1	1	0	0	0	0	0	0	4	Moderate
Lewis, 2013	1	1	1	1	0	0	0	0	0	0	4	Moderate
Malmborg et al., 2017	0	1	1	1	0	0	0	0	0	0	3	Low
Marchi & Baratto, 2018	1	1	1	1	0	0	0	0	0	0	4	Moderate
Rizzieri et al., 2019	0	1	1	0	0	0	0	0	0	0	2	Low
Rudolph, 2017	0	1	1	0	0	0	0	0	0	0	2	Low
Segura-Garcia et al., 2012	1	1	1	0	0	0	0	0	0	0	3	Low
Silva et al., 2021	1	1	1	0	0	1	0	0	0	0	4	Moderate
Surala et al., 2020	0	1	1	0	0	0	0	0	0	0	2	Low
Tocchetto et al., 2018	1	1	1	0	0	0	0	0	0	0	3	Low
Uriegas et al., 2021	1	1	1	0	0	0	0	0	0	0	3	Low
Valera et al., 2014	1	1	0	1	0	0	0	0	0	0	3	Low

Item score: (0: low risk, 1: high risk). §Total quality/risk score: [range (0–10): high quality/low risk (0–3), moderate quality/risk (4–6), poor quality/high risk (7–10)].

TABLE 3. Results of meta regression of percentage female, type of sport and sample size on ON prevalence among the exercising populations.

Predictor	Coefficient	SE	95% CI	Z-value	2-sided p
Percentage female	0.0176	0.0145	-0.0109, 0.0460	1.21	0.226
Individual sport ¹	-0.4420	0.5602	-1.5000, 0.6559	-0.79	0.430
Team sport ¹	0.6575	0.9990	-1.3006, 2.6155	0.66	0.511
Sample size	-0.0016	0.0010	-0.0035, 0.0003	-1.71	0.087

Note: SE = standard error, 95% CI = 95% confidence interval, ¹Mixed sports/unknown comprised the reference category.

Discussion

A total of 24 studies fulfilled the inclusion criteria and were consequently included in the meta-analysis. The studies yielded an overall ON prevalence of 55.3%. The dispersion of effect sizes was significant, ranging from 4.3% (58) to 89.2% (49). The prevalence of ON was overall very high across the included studies and suggests that approximately over half of the population are suffering from it, indicating that ON is a frequent eating disorder among exercising populations. Hence, a significant proportion of those who aim to achieve and/or maintain good physical health may be prone to a preoccupation with healthy eating to a dysfunctional level (18, 48, 55, 56, 58, 60). Accordingly, focusing on prevention and treatment of ON among exercising populations should be prioritized (67). The reported prevalence of ON was considerably higher compared to the prevalence of eating disorders in general among athletes, which is estimated to be 13.1% (14), but is still comparable to the ON prevalence in the general population (41% estimated by the ORTO-15) (65).

Although these findings can be regarded as support for the high prevalence of ON in exercising populations, the findings should still be interpreted with caution. ON is currently not acknowledged as a diagnostic entity in formal psychiatric nosology (ICD-11 and DSM-V), and thus there is still a lot of discussion about the diagnostic criteria and methods and tools used for its classification (65). Furthermore, pathological ON is often overlapping with healthy orthorexia and orthorexic behaviours (68). Accordingly, many have criticized the methods used to assess ON for being too unspecific and insensitive and for having various cut-offs and for being based on different definitions of ON (4, 8, 68-70). The ambiguity associated with the ON diagnosis and its measuring tools can lead to false positive results and inflated prevalence rates. On the other hand, it cannot be ruled out that the high prevalence reflects real problems, mirroring the more invisible symptoms and characteristics of ON in contrast to the more tangible symptoms associated with other eating disorders such as anorexia nervosa and binge-eating disorder (71, 72). In addition, the high prevalence of ON can also reflect a willingness to report, and even over-report, the relevant symptoms, which in contrast to other eating disorders may be regarded as desirable, which seems to be the case for symptoms of exercise addiction (73).

In all, 11 of the 24 studies consisted of students, where the mean age across those studies was 18.8 years (3, 22, 44, 46, 48, 50, 51, 57, 59, 61, 63). This may also have an impact on the prevalence. Another factor that may affect the prevalence is geographical location. Only seven (22, 44, 46, 47, 53, 61, 63) of the included studies were conducted outside Europe, and only three (47, 53, 61) of these stemmed from non-western countries. The

predominance of studies from western countries may be yet another reason for the high prevalence rates because studies on other eating disorders (e.g., anorexia nervosa) show a higher incidence in western countries compared to non-western countries (65, 74).

The high heterogeneity in terms of prevalence rates of ON found in this review resonates well with the differences in ON prevalence reported from the general population, where it varies between 6.9% and 75.2% (65). This large disparity in prevalence might be explained by the same factors as mentioned above, however it may also reflect divergence on other study characteristics as well. In order to elucidate this further in terms of the present study, a meta-regression analysis with three independent variables was conducted. The independent variables included sex (percentage females), type of sport (individual, team sport, mixed/unknown), and sample size. Females were hypothesized to show higher prevalence than men (23), those involved in individual sports were hypothesized to be more predisposed for ON than those involved in team sports (75), and sample size was hypothesized to be inversely related to the prevalence.

However, none of the independent variables were significantly associated with the prevalence rates. This implies that other study characteristics may explain the heterogeneity. Possible candidate variables in this context are risk of bias and other sample characteristics. It is further conceivable that athletes compared to more recreational exercisers put stronger emphasis on diet and thus would score higher on orthorexic tendencies. Other potential moderators entail age (76), student status (77), status as vegetarian/vegan (78), psychiatric comorbidity (79), and level of physical activity(80). These should thus be investigated in future studies.

Limitations of the included studies

Notably, one fourth of the included studies had moderate risk of bias, typically associated with low external validity. Further, it should be noted that only one of the included studies used random selection to recruit the sample (61). Two of the included studies were not published in a peer-reviewed journal (44, 61), and two of the included studies did not use a specific cut-off and rather used a score-interval that resulted in “high risk” in terms of internal validity regarding defining ON (47, 60). Hence, future studies should improve especially on these study dimensions in order for the field to move forward. Further, it is conspicuous that all studies were based on a cross-sectional design. Some studies had limitations in terms of reporting, as only demographic information of the total sample, and not specifically for those included as exercisers were reported in eight studies (22, 50, 51, 57, 59-61).

Limitations and strengths of the present meta-analysis

Almost half (18, 46, 48, 50-52, 56, 57, 59, 61, 63) of the included studies lacked information about the type of sport and were consequently grouped as unknown for this parameter. In addition, detailed demographic information about the exercising sub-samples was sometimes lacking, hence descriptive data for the whole samples was in these cases included in the analysis. This represents limitations regarding sex and type of sport as moderators. Still, it should be noted that the authors of the present meta-analysis contacted the study authors in an attempt to obtain missing information. The present meta-analysis targeted the inclusion of grey literature, as recommended for the calculation of non-biased estimates in meta-analyses (37). No restriction in terms of time frame were applied, articles in all European languages were included, and the meta-analysis was conducted in line with the updated PRISMA guidelines (30), and these all are strengths of the present study. Although searches were conducted in several relevant databases, we cannot rule out that some relevant papers were not included. All prevalence data and quality assessments of the included studies were coded independently by two reviewers. The inter-rater reliability of the study screening procedure was provided by the Covidence software and showed a proportional agreement of 96.0% in the title/abstract screening (Cohen's kappa = .82) and 94.7% in the full-text screening (kappa = .88). This indicates a substantial agreement between the two reviewers. Still, for some parameters there were substantial disagreements. This is not necessarily a problem (81), and disagreement encouraged discussion between the reviewers, finally resulting in consensus being reached. Some of the included articles had with several cut-offs, of which the most liberal was selected, which to some degree may have inflated some of the single as well as the overall prevalence estimates.

Conclusions and recommendations for further studies

The present meta-analysis revealed a high prevalence of ON, albeit with a large disparity between studies. Still, none of the moderators (sex, type of sport, and sample size) could explain this heterogeneity. To expand our knowledge in this field, we need a better definition and agreed-upon diagnostic criteria of ON. Sensitive and reliable measurements of ON should also be developed. In addition, more longitudinal studies are warranted in order to identify predictors and not just correlates of ON. Prevalence studies including respondents from multiple sports, with wider age-ranges, from non-western countries, and based on representative samples will also help advance the field. More knowledge of actual ON

prevalence and predictors are instrumental in order to estimate the actual need for treatment and to develop targeted preventive efforts.

Abbreviations

CI: confidence interval; df: degrees of freedom; I^2 : heterogeneity; ON: orthorexia nervosa; ORTO-15: Orthorexia Nervosa 15 Questionnaire; SD: standard deviation

Ethics approval and consent to participate

Not applicable.

Consent for publication

Not applicable.

Availability of data and materials

The data are available in the selected studies. The dataset that was generated after extracting the themes from the selected studies is available upon a substantiated request to the corresponding author.

Competing interests

The authors declare that they have no competing financial interests.

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None.

Authors' contributions

Study conceptualization, literature search, data analysis, and coding of studies were conducted by SMH and JB. SMH contributed to interpretation of data, writing, and revising the work critically for important intellectual content. All authors read and approved the final version of the work to be published and agree to be accountable for all aspects of the work in ensuring that questions to the accuracy of any part of the work are appropriately investigated and resolved.

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Authors' information

SMH is a student at the faculty of psychology at University in Bergen (UiB). JB is co-author and an exchange student from Germany.

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Appendix A

PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
TITLE			
Title	1	Identify the report as a systematic review.	P. 1
ABSTRACT			
Abstract	2	See the PRISMA 2020 for Abstracts checklist.	P. 2-3
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of existing knowledge.	P. 4
Objectives	4	Provide an explicit statement of the objective(s) or question(s) the review addresses.	P. 5
METHODS			
Eligibility criteria	5	Specify the inclusion and exclusion criteria for the review and how studies were grouped for the syntheses.	P. 6
Information sources	6	Specify all databases, registers, websites, organisations, reference lists and other sources searched or consulted to identify studies. Specify the date when each source was last searched or consulted.	P. 5
Search strategy	7	Present the full search strategies for all databases, registers and websites, including any filters and limits used.	P. 5
Selection process	8	Specify the methods used to decide whether a study met the inclusion criteria of the review, including how many reviewers screened each record and each report retrieved, whether they worked independently, and if applicable, details of automation tools used in the process.	P. 6-7
Data collection process	9	Specify the methods used to collect data from reports, including how many reviewers collected data from each report, whether they worked independently, any processes for obtaining or confirming data from study investigators, and if applicable, details of automation tools used in the process.	P. 7-8
Data items	10a	List and define all outcomes for which data were sought. Specify whether all results that were compatible with each outcome domain in each study were sought (e.g. for all measures, time points, analyses), and if not, the methods used to decide which results to collect.	P. 9
	10b	List and define all other variables for which data were sought (e.g. participant and intervention characteristics, funding sources). Describe any assumptions made about any missing or unclear information.	P. 7-8
Study risk of bias assessment	11	Specify the methods used to assess risk of bias in the included studies, including details of the tool(s) used, how many reviewers assessed each study and whether they worked independently, and if applicable, details of automation tools used in the process.	P. 8
Effect measures	12	Specify for each outcome the effect measure(s) (e.g. risk ratio, mean difference) used in the synthesis or presentation of results.	P. 9
Synthesis methods	13a	Describe the processes used to decide which studies were eligible for each synthesis (e.g. tabulating the study intervention characteristics and comparing against the planned groups for each synthesis (item #5)).	P. 9
	13b	Describe any methods required to prepare the data for presentation or synthesis, such as handling of missing summary statistics, or data conversions.	P. 7-8
	13c	Describe any methods used to tabulate or visually display results of individual studies and syntheses.	P. 9
	13d	Describe any methods used to synthesize results and provide a rationale for the choice(s). If meta-analysis was performed, describe the model(s), method(s) to identify the presence and extent of statistical heterogeneity, and software package(s) used.	P. 9
	13e	Describe any methods used to explore possible causes of heterogeneity among study results (e.g. subgroup analysis, meta-regression).	P. 9
	13f	Describe any sensitivity analyses conducted to assess robustness of the synthesized results.	P. 8
Reporting bias assessment	14	Describe any methods used to assess risk of bias due to missing results in a synthesis (arising from reporting biases).	P. 8
Certainty assessment	15	Describe any methods used to assess certainty (or confidence) in the body of evidence for an outcome.	P. 9
RESULTS			
Study selection	16a	Describe the results of the search and selection process, from the number of records identified in the search to the number of studies included in the review, ideally using a flow diagram.	P. 13
	16b	Cite studies that might appear to meet the inclusion criteria, but which were excluded, and explain why they were excluded.	P. 11
Study characteristics	17	Cite each included study and present its characteristics.	P. 14-15
Risk of bias in studies	18	Present assessments of risk of bias for each included study.	P. 15
Results of individual studies	19	For all outcomes, present, for each study: (a) summary statistics for each group (where appropriate) and (b) an effect estimate and its precision (e.g. confidence/credible interval), ideally using structured tables or plots.	P. 12
Results of syntheses	20a	For each synthesis, briefly summarise the characteristics and risk of bias among contributing studies.	P. 15
	20b	Present results of all statistical syntheses conducted. If meta-analysis was done, present for each the summary estimate and its precision (e.g. confidence/credible interval) and measures of statistical heterogeneity. If comparing groups, describe the direction of the effect.	P. 15
	20c	Present results of all investigations of possible causes of heterogeneity among study results.	P. 13
	20d	Present results of all sensitivity analyses conducted to assess the robustness of the synthesized results.	P. 13-14
Reporting biases	21	Present assessments of risk of bias due to missing results (arising from reporting biases) for each synthesis assessed.	P. 13-14
Certainty of evidence	22	Present assessments of certainty (or confidence) in the body of evidence for each outcome assessed.	P. 13
DISCUSSION			
Discussion	23a	Provide a general interpretation of the results in the context of other evidence.	P. 16
	23b	Discuss any limitations of the evidence included in the review.	P. 17
	23c	Discuss any limitations of the review processes used.	P. 18
	23d	Discuss implications of the results for practice, policy, and future research.	P. 17-18
OTHER INFORMATION			
Registration and protocol	24a	Provide registration information for the review, including register name and registration number, or state that the review was not registered.	P. 5
	24b	Indicate where the review protocol can be accessed, or state that a protocol was not prepared.	Protocol not made
	24c	Describe and explain any amendments to information provided at registration or in the protocol.	P. 5
Support	25	Describe sources of financial or non-financial support for the review, and the role of the funders or sponsors in the review.	None
Competing interests	26	Declare any competing interests of review authors.	None
Availability of data, code and other materials	27	Report which of the following are publicly available and where they can be found: template data collection forms; data extracted from included studies; data used for all analyses; analytic code; any other materials used in the review.	P. 9

From: Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *BMJ* 2021;372:n71. doi: 10.1136/bmj.n71

Appendix B

Author guidelines for reviews at “Journal of Eating Disorders”: (Vancouver reference style).

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