CASE REPORT



Pathways to orthorexia nervosa: a case series discussion

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Abstract

Background Orthorexia nervosa (ON) has gained increasing interest in the last 2 decades. Although a consensus on the diagnostic boundaries of ON has not yet been reached, there is some evidence for an overlap with eating disorders, obsessive—compulsive disorder, and psychotic disorder. Most of the knowledge about ON has emerged from studies of non-clinical and at-risk populations and is focused on differential diagnosis; therefore, further clinical studies are needed to better outline the ON phenomenon in a real-life setting.

Objective This case series aims at describing clinical cases that developed symptoms suggestive of ON after being diagnosed with a prior psychiatric disorder and then discussing them in light of possible clinical pathways.

Methods Four women consecutively admitted to an outpatient unit for the treatment of eating disorders were diagnosed with ON through a clinical interview, according to Dunn and Bratman's criteria and self-administered questionnaire assessment (ORTO-15), and were considered to be eligible for this case series study. Psychiatric anamnestic data were collected retrospectively.

Results The anamnesis revealed that all patients were previously diagnosed with a psychiatric disorder (i.e. obsessive–compulsive disorder, bulimia nervosa, illness anxiety disorder, and psychotic disorder) before developing ON.

Conclusion Past literature focused on differential diagnosis between ON and other psychiatric disorders. This is the first description of clinical cases in a real-life setting that started with different psychiatric disorders and later developed symptoms suggestive of ON. These cases have generated a new research question on the possibility that different psychiatric disorders may associate with a later onset of ON.

Level of evidence Level V, descriptive study.

Keywords Orthorexia nervosa · Case series · Pathway · Differential diagnosis · Comorbidity

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Introduction

Orthorexia nervosa (ON), despite great controversy, has gained increasing interest in the last 2 decades. Although the first description of the phenomenon seemed clear [1], subsequent definitions have shown significant heterogeneity of interpretations of the concept [2].

This heterogeneity could partly explain why, despite various diagnostic criteria being proposed [3–6], ON has not yet been recognized as a psychiatric disorder. Regardless of the lack of consensus on a shared and official definition of the phenomenon, a recent narrative review stated that both an obsessive focus on healthy eating and a consequent clinically significant impairment need to be present for the diagnosis of ON [2].



A major debate has arisen over whether ON should be considered as a distinct disorder or a variant of other psychiatric disorders [7]. Most research on ON has focused on epidemiological studies in the general population or at-risk samples and has discussed ON in terms of differential diagnosis with eating disorders (EDs), obsessive—compulsive disorder (OCD), and psychotic disorder. Clinical studies were limited to a few case reports [4, 8–11] describing the clinical presentation and management of cases falling into the ON diagnosis. To this extent, the Orthorexia Nervosa Task Force (ON-TF) is actually demanding more qualitative studies to help in outlining clinical aspects and diagnostic controversies [2].

To the best of our knowledge, only two studies reported cases of ON with a prior [12] or later diagnosis [4, 13] of other psychiatric disorders. As most of the past literature discussed differential diagnosis and as diagnostic criteria do not agree with the need to exclude them to make a diagnosis of ON, herein we present four clinical cases of patients who were diagnosed for a prior psychiatric disorder of interest and at some point in the follow-up developed symptoms suggestive of ON. Thus, this manuscript aims at exposing four different presentations of ON and discussing them in light of possible clinical pathways.

Methods

The study took place at the Centre for Clinical Research and Treatment of Eating Disorders in Catanzaro (Italy), allocated within the Magna Graecia University outpatient service of general psychiatry. This tertiary multidisciplinary service provides outpatient mental health care to adolescents, adults, and the elderly with EDs. The assessment includes structured clinical interviews with psychiatrists and psychologists expert in the field of EDs and the administration of self-report questionnaires. Before any treatment takes place, patients are referred to other specialized settings whenever necessary.

The case series described herein consisted of four patients followed up at the university outpatient unit of the general psychiatric or ED unit for a psychiatric disorder other than ON and who were subsequently referred to the ED unit for the development of symptoms compatible with ON. All patients were interviewed by experienced psychiatrists and screened for psychiatric disorders (e.g. mood disorder, anxiety disorder, and psychotic disorder) according to the Structured Clinical Interview for DSM-5 (SCID-5-CV) [14] and for EDs through the Eating Disorder Examination [15]. Dunn and Bratman's criteria were used during the clinical interview to investigate the symptoms of ON. These criteria were chosen because they are the most recent, they are exhaustive in describing ON and they do not consider other

previous or concurrent psychiatric disorders as exclusion criteria for the diagnosis of ON. The ORTO-15 self-rated questionnaire [16] was administered to patients in support of the clinical diagnosis. A more restrictive cut-off was considered for ORTO-15 (pathological score < 35, instead of < 40), as previously reported in other studies [13, 17].

Table 1 summarizes the clinical presentation of the four cases according to Dunn and Bratman's criteria and other clinical aspects of interest. Figure 1 illustrates the four cases in light of possible clinical pathways.

Patients gave their written informed consent for the collection and management of their medical records and for the publication of this case report. All procedures were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Case 1

A.A., a 32-year-old woman, was referred to the outpatient unit of the ED centre to investigate her recent and robust weight loss.

At the age of 27, A.A. was diagnosed with OCD. She described her initial doubts about her capabilities to understand her feelings and to take action (e.g. buying a house or not, getting married or not), turning into doubtful obsessions about choosing between "the right and the wrong" in everyday life. The compulsions were mainly mental actions about "reconsidering events" to solve the doubtful obsessions and reduce anxiety. Her daily functioning was completely disrupted. She underwent drug therapy (sertraline 100 mg/day) and weekly psychotherapy (cognitive behavioural therapy, CBT). Full recovery was achieved after 2 years of combined therapy. As maintenance, drug treatment was gradually interrupted in favour of individual CBT sessions (every 2 weeks) and follow-up visits at the outpatient psychiatric unit, in accordance with the preference of the patient.

At the time of consultancy, she mentioned that after a "stressing period" she became worried about the healthiness of food and her ability to take care of herself ("have I done it in the right way?"; "am I able to make the right choices for my own well-being?"; "perhaps I am not even able to choose my meals correctly"). Her eating habits turned to strict restriction and highly selective choices towards food that was non-canned and not treated with additives and preservatives, such as "organic food". More than an hour a day was dedicated to looking for these products and she observed precise rules in washing and cooking meals. She described eating "healthy" as the best way to overcome her worries about unsafety and her inability to take care of herself, as well as being rewarding. On the contrary, situations in which she could not exert control on meals, such as dinners with



Table 1 Diagnostic criteria according to Dunn and Bratman and further clinical aspects of interest of cases

	Case 1	Case 2	Case 3	Case 4
Dunn and Bratman's criteria				
Criterion A: Obsessive focus on "healthy" eating, as defined by a dietary theory or set of beliefs whose exaggerated emotional distress in relationship to food choices perceived as unhealthy; weight loss must but this is not the primary goal. As evidenced by the following		-	•	-
1. Compulsive behavior and/or mental preoccupation regarding affirmative and restrictive dietary practices believed by the individual to promote optimum health	✓	✓	✓	✓
2. Violation of self-imposed dietary rules causes exaggerated fear of disease, sense of personal impurity and/or negative physical sensations, accompanied by anxiety and shame	✓	✓	✓	✓
3. Dietary restrictions escalate over time and may come to include elimination of entire food groups and involve progressively more frequent and/or severe "cleanses" (partial fasts) regarded as purifying or detoxifying. This escalation commonly leads to weight loss, but the desire to lose weight is absent, hidden or subordinated to ideation about healthy eating	✓	✓	✓	✓
Criterion B: The compulsive behavior and mental preoccupation become clinically impairing by any	of the follow	ving		
1. Malnutrition, severe weight loss or other medical complications from restricted diet	✓		\checkmark	✓
2. Intrapersonal distress or impairment of social, academic or vocational functioning secondary to beliefs or behaviors about healthy diet	✓	✓	✓	✓
3. Positive body image, self-worth, identity and/or satisfaction excessively dependent on compliance with self-defined "healthy" eating behavior	✓	✓	✓	✓
Other clinical aspects of interest of cases				
Unbalanced diet		✓	✓	✓
Weight loss	✓		✓	✓
Psychological distress	✓	✓	✓	✓
Physical health problems	✓		✓	✓
Need to detox when healthy eating fails	✓	✓		✓
Sense of virtue related to healthy eating	✓	✓	✓	✓
Judgement of other's unhealthy eating	✓		✓	✓
Loss of social connection	✓		✓	✓
Ideas source	Unknown	Internet	Internet	Unknow

friends or parties, made her anxious and irritable, and she progressively started to refuse invitations. Her body mass index (BMI) was 17.9 kg/m² and the ORTO-15 score was 34. Body dissatisfaction, drive for thinness, and other pathological eating behaviours were not present.

Case 2

F.F., a 24-year-old woman, was admitted to the outpatient unit of the ED centre for a regular follow-up visit. At the age of 20, after a period of severe restriction and weight loss, she started bingeing and vomiting to minimize weight gain. She was diagnosed with bulimia nervosa and enrolled into a nutritional rehabilitation programme, plus weekly sessions of CBT. After 6 months, the bingeing and purging symptoms were drastically reduced, weight was restored and she began to feel more comfortable with her shape and weight. She returned to her studies and exams and then began a love affair. One year later, she no longer satisfied the diagnostic criteria for an ED and decided to stop the psychotherapy sessions. During a follow-up visit 12 months later, the previous ED symptoms were no longer detectable, but she had

become more selective with healthy food and eating style, turning to gluten-free and almost completely vegetarian meals. She no longer focused on calories but on "healthy features of food"; her current BMI was 20.3 kg/m² and the ORTO-15 score was 34. Referring to the past, she felt appalled that she had hurt her body and affirmed that she had learnt to take care of herself by "eating healthy", with "the right way and timing" of cooking. She confirmed that she had lost some friends due to her "healthy behaviors" but had met new ones who also shared her new "eating lifestyle".

Case 3

M.M. was 45 years old and the mother of an 11-year-old male patient suffering from somatic symptom disorder along with severe weight loss. Because the patient was underage, both parents were involved in the nutritional rehabilitation programme. At that time, a sturdy attitude and excessive meticulousness, especially with the cooking methods (preference for uncooked food), together with anxiety and intolerance, became more and more evident until M.M. asked for an outpatient psychiatric visit for herself. The anamnesis



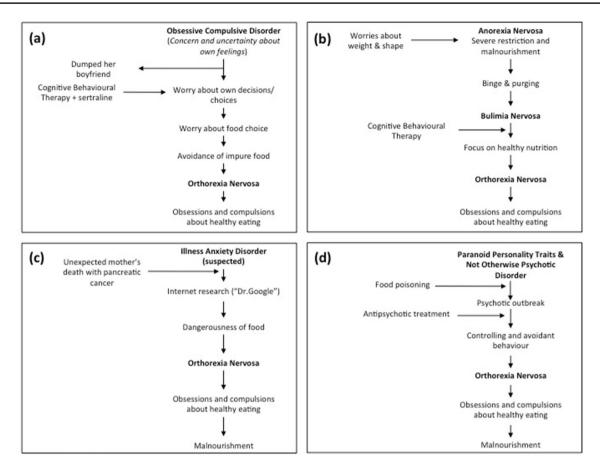


Fig. 1 Pathways to orthorexia nervosa from: a obsessive—compulsive disorder, b anorexia nervosa, c illness anxiety disorder, d paranoid personality trait

revealed a strong worry about contracting severe physical illness during adolescence and young adulthood that was never treated pharmacologically or with psychotherapy. When her mother died of pancreatic carcinoma some years before, M.M. investigated the causes of the disease, including foodstuffs. She convinced herself that some foods and cooking with unsuitable materials or for inappropriate times might lead to fatal illness. Because of this concern, she bought new crockery, pots and pans, with particular attention to the thickness and quality of the materials, and she instructed herself on "not risky" food and on proper cooking strategies. She definitively learned to buy only "safe food" and to cook in a ritualistic way for all the family. Her diet allowed only the use of integral foods, the total elimination of the addition of fats for cooking (e.g. butter, oil, sour cream, etc.) and steaming or grilling as the only cooking techniques. Detoxifying purges (i.e. fasting and purifying herbal teas) were not infrequent if this dietary pattern could not be followed in social situations.

No symptoms of body dissatisfaction or body weight control had ever been present. Even if her weight loss was not critical (BMI = 14.9 kg/m²), as it was for her child, general

malnutrition was evident in the whole family. The ORTO-15 score was 32.

Case 4

G.G., a 39-year-old woman, was accompanied by her father who was asking for help in dealing with her weight loss and strange eating behaviours.

At the age of 25, she was diagnosed with a paranoid personality trait and a psychotic disorder not otherwise specified; symptoms were successfully treated with antipsychotic medication in a few months. After some years of being well, she suffered from delusional thoughts about being killed by food poisoning at her aunt's house during dinner and received pharmacological treatment, with prompt resolution of the episode. When questioned about the event, the patient stated that she was wrong in thinking that her aunt was planning to kill her and that it was much more likely that the food was "not good". Since then, she began to enquire about food preservation (e.g. chemical treatments, additives, etc.) and cooking methods and to worry about "dangerous food", qualitatively restricting her diet. She became a vegetarian



and refused to buy food except directly from the grower. She also decided to take only nutraceutical and homeopathic drugs and started buying them on the internet, fearing that conventional drugs may contain traces of "unhealthy" components. Eventually, she became a homeopath and because of her enduring beliefs about the "unhealthy" components of drugs, she became chronically affected by easily treatable diseases (e.g. bronchitis). Her interpersonal relationships gradually shrunk because she found it difficult to share her food and life ideas with her peers.

At the time of her visit, she was severely underweight (BMI=151 kg/m²) and exhibited strict beliefs and behavioural rules about which food is "safe and healthy" and how to manage with it (ORTO-15 score = 33). The reason why she accepted a specialized visit was the "possibility to enlarge her knowledge about eating healthier" than she was actually doing.

Discussion

As the literature has tried to discriminate between ON and EDs, OCD, psychotic disorders and illness anxiety disorders, and the diagnostic criteria do not agree with the need to exclude them before making a diagnosis of ON, it seemed reasonable and meaningful to present clinical cases that were already diagnosed with these psychiatric disorders and then developed symptoms of ON.

Even though there is not a shared consensus on the definition and diagnostic criteria of ON, based on the clinical presentation, the clinical interview and the anamnesis, together with the pathological scoring on the self-report questionnaire (ORTO-15) and the satisfaction of the diagnostic criteria proposed by Dunn and Bratman, all cases were compatible with the latest synoptic definition of ON by Cena et al. [2].

It is well recognized that some clinical features of ON resemble other mental disorders, with a substantial overlap of symptoms and subsequent difficulty in making a differential diagnosis (Fig. 2). This was particularly true that these cases had already been diagnosed with the mental disorders that need to be excluded before diagnosing ON.

In the first case, the patient was previously diagnosed with OCD, which shares many symptoms with ON (i.e. time-consuming obsessions and compulsions). Nonetheless, the obsessions about healthy food and the time-consuming behaviours and rituals were somehow different from the doubtful obsessions and compulsions the patient experienced before. In the first instance, the patient asked for help because her worries were "unreasonable and excessive" and she felt overwhelmed by obsessions. In contrast, when ON was diagnosed, she considered her worries as "proper and reasonable" and her rituals as reassuring and self-caring.

One possible explanation for this lack of insight could be the worsening or relapse of the primary disorder; OCD is often a chronic condition and the patient was no longer under pharmacological treatment. On the other hand, the egosyntonic nature of the obsessive content has already been mentioned as discriminating between ON (mostly egosyntonic) and OCD (mostly egodystonic) [18].

In the second case, the starting point was bulimia nervosa, successfully treated with CBT. As the patient became aware of the possible physical consequences of food restriction and bingeing/purging, she started focusing on health instead of weight and shape, and she turned from controlling the calories and quantities towards favouring the quality of meals. In other words, "drive for thinness" changed into a "drive for health". Case 2 (as with Case 1) considered herself to be fully recovered. Previous research demonstrated that ON symptoms are frequent among ED patients after treatment [13] and that it can be explained as evolution of the ED, as well as an iatrogenic-like side effect of psychotherapy or the intellectualization of symptoms moving towards a socially acceptable position/behaviour. Moreover, even if the patient recovered from her concerns about weight and body dissatisfaction and her BMI was normal, it is worth mentioning that ED symptoms may sometimes persist in a "hidden/silent" form, as suggested by Musolino et al. ("healthy anorexia") [19] and Bratman ("covert anorexia") [1]. Her new "eating style" had driven her to change her social environment and choose new friends who supported her eating concerns.

The third patient was never diagnosed or treated for anxiety symptoms, but symptoms of a possible illness anxiety disorder were discovered in her early anamnesis (i.e. preoccupation with acquiring a serious illness and a high level of anxiety about health). The fear of illness turned into obsession about the risk of illness caused by unhealthy foods or cooking procedures after her mother's death from pancreatic cancer. At the time of her visit, she did not meet the criteria for a somatic or illness anxiety disorder: she did not have one or more somatic symptoms, did not frequently use healthcare services and did not fear having an undiagnosed fatal disease. She definitely had no insight of her pathological state; instead, her concerns and rituals about cooking were perceived as "the best way to avoid illness and taking care of her family". In his first conceptualization of ON, Bratman argued that the fear of illness may come before the onset of ON [1], as well as OCD and illness anxiety disorder. To the best of our knowledge, an evaluation of the association between illness anxiety disorder and ON is still missing, with some evidence of an association between anxiety in general and health anxiety with ON [7, 20, 21].

In the fourth case, the patient had already been treated for a psychotic disorder not otherwise specified. The content of her first delusion concerned "poisoned food", which





Fig. 2 Differential diagnosis between orthorexia nervosa and anorexia nervosa, obsessive—compulsive disorder, illness anxiety disorder, and psychotic disorder

after treatment turned from "I was poisoned" into "I got gastrointestinal problems because of bad food, because just what is pure from preservatives and additives can be considered healthy". ON has already been associated with the psychosis spectrum [12] and some authors have defined the theoretical food-related thoughts suggestive of a comorbid or future psychosis [22, 23]. These "food-related magical thoughts" were not present from that moment; instead, the patient cared more about the content of preservatives, artificial flavours and additives, exposure to pesticides or supplements, processing, packaging and unhealthy cooking (symptoms more typical of ON).

Pros and cons for/against a clinical diagnosis of orthorexia nervosa

It is not clear if these cases should be considered as re-exacerbations of the primary disorders with a different pathological content. To date, there is no consensus among the proposed diagnostic criteria about the need for excluding the exacerbation of another mental disorder that directly helps the clinician to clarify if ON is a distinctive disorder or a multiform expression of a primary disorder, especially if another mental disorder has already been diagnosed. Among the diagnostic proposals [2], only Setnick [3] and Moroze [4] criteria addressed the need for excluding that symptoms are a mere exacerbation of another mental disorder, such as anorexia nervosa, bulimia nervosa, OCD, schizophrenia or other psychotic disorder.



Deciding whether two coexisting clinical entities should be considered the expression of a single disorder or two distinct phenomena is often speculative, as we are very far from knowing in depth how mental disorders interrelate and affect each other in terms of onset, psychopathology and outcome [24]. This is particularly true in the case of ON cooccurring with other mental disorders, as there is not enough knowledge about the ON phenomenon per se in real life, or the strength of its association with other mental disorders, to prioritize one disorder over another (i.e. privileging the diagnosis of OCD when OCD and ON are co-occurring).

On the other hand, another explanation may be that mental disorders that share some dimensional psychopathological features with ON may facilitate its onset through a diagnostic continuum (clinical pathways). In this regard, previous studies have shown that ON, OCD, and anorexia nervosa are characterized by personality traits that include perfectionism, rigid thinking, excessive devotion, hypermorality and a preoccupation with details and perceived rules [25–30], as well as poor performance in cognitive tasks (i.e. set shifting, a marker of cognitive flexibility) [31–36]. The evaluation of personality, or cognitive functions, as well as other psychological dimensions of interest, was beyond the aim of the present study and no further evidence can be furnished in this regard. Although a dimensional approach generally complicates rather than simplifies classification systems, it may help to clarify those clinical cases that transit from ON to other mental disorders, and vice versa.

Undoubtedly, social pressure towards a healthy lifestyle, as well as the diffusion of news about the dangers of food (e.g. pesticides, preservatives, cooking procedures, quality of nutrients, etc.), need to be taken into account. Being healthy and having an idealized healthy lifestyle have nowadays taken a leading role and conform to the cultural ideals of our society. We previously hypothesized ON to be an 'upgrade' in terms of the trend and social acceptance of more discriminated mental disorders [13]. Thus, it can be inferred that some mental disorders that share common psychopathological dimensions, together with social pressure, could favour the development of clinical ON.

Given its descriptive nature, the present study does not provide clinical information generalizable to the whole population affected by ON. Furthermore, the cases are described up to the moment of ON diagnosis but a follow-up evaluation of the clinical pictures is missing. These limitations have prevented the capture of information on the intrinsic stability of ON symptoms over time and the possible reemergence of key symptoms of the primary disorder.

On the other hand, the present manuscript has the strength of presenting clinical cases of ON; most of the literature data on ON are provided by studies on specific groups, such as athletes [7], doctors [13, 18], dieticians [19, 20, 37] and artists [38], that are considered to be at higher risk but these

data do not show the clinical presentation in the real world. As far as we know, this is the first study reporting clinical cases of ON whose onset was years after a prior psychiatric disorder; given their particular clinical presentations, a provisional argumentation of the complexity that links ON with other mental disorders of interest has been raised. Bearing in mind the great debate ongoing in the literature—the ON-TF efforts to define diagnostic boundaries and decide whether ON should be considered as a distinct disorder or a variant of an existing disorder—this case report should be considered as provisional evidence for possible clinical pathways leading to ON as a later diagnosis.

In clinical practice, clinicians dealing with mental health should investigate symptoms of ON among patients suffering from OCD and psychotic disorders and also past symptoms suggestive of other mental disorders when faced with patients diagnosed with ON. Eating behaviours and preferences could hide clinically relevant information enabling a more complete diagnosis. More research is needed in clinical samples to clarify the nature, strength and temporality of the association between ON and other mental disorders of interest.

What is already known on this subject?

ON share common symptoms with ED, OCD, and psychotic disorder and past research dealt with differential diagnosis with these mental disorders.

What does this study add?

This study provides an argumentation about the possibility that ED, OCD, illness anxiety disorder and psychotic disorder may lead to ON across a diagnostic continuum, and that the relation among these disorders might be considered in term of pathways, rather than differential diagnosis.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent Patients gave their informed consent before collecting any data from their medical records.



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