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CASE REPORT

Delayed avoidant restrictive food intake disorder diagnosis leading to Ogilvie's syndrome in an adolescent

Valérie Bertrand[1,2] · Caroline Dhenin[1] · Pierre Déchelotte[2,3] · Mathieu Faerber[4]

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Abstract

Purpose Avoidant restrictive food intake disorder (ARFID) was recently characterized, according to the DSM-5 classification, as a feeding and eating disorder (FED). However, ARFID remains poorly known by most pediatricians, but also by psychiatrists and primary care professionals. Despite the fact that patients with ARFID generally have a higher BMI than patients with anorexia nervosa, our purpose was to highlight the fact that they may present severe nutritional deficiencies and major somatic complications when the diagnosis is delayed.

Method We describe here a case of a 16-year-old boy who presented with severe undernutrition (BMI = 11.5) leading to Ogilvie's syndrome, which resolved with enteral refeeding. Because of undernutrition, very bad dental condition, and encopresis, some physicians wrongly suspected child neglect, but retrospective analysis of his personal history revealed a long-term FED and sensory specificities that led to the final diagnosis of an ARFID–autism spectrum disorder (ASD) association. A literature review was conducted on the ARFID somatic complications.

Conclusion The training of health professionals in the clinical forms of pediatric FED, including ARFID, is necessary, to promote early diagnosis and prevent poor nutritional outcomes. In this case the association of ARFID–ASD and the delay in access to specialized care favored by the disadvantaged social environment led to severe gastrointestinal complications.

Level of evidence V, descriptive study.

Keywords ARFID · Autism spectrum disorder · Undernutrition · Ogilvie's syndrome · Feeding and eating disorders · Mistaken child abuse

Introduction

Avoidant restrictive food intake disorder (ARFID) was recently characterized, according to the DSM-5 classification, as a feeding and eating disorder (FED), like anorexia nervosa (AN) and other more typical FED [1] (Table 1). However, ARFID remains poorly known by the pediatric community, but also by psychiatrists and primary care professionals, which can lead to diagnosis delays [2, 3]. Although the BMI of new patients with ARFID is generally higher than that of patients with AN [4], there may be an overlap, and the BMI can be very low, as shown in this case report, with severe undernutrition leading to Ogilvie's syndrome, in a context of a very delayed diagnosis of the ARFID–autism spectrum disorder (ASD) association and a subsequent delayed access to specialized care.

Case presentation

A 16-year-old boy was accompanied by his family to the emergency department for bloating and abdominal pain. The family explained that the symptoms had started several months before but had worsened recently, and that he wore sanitary pads because of recent diarrhea. The boy had missed many days of school over the past 3 months, but he refused to take treatment and to do the blood tests

✉ Valérie Bertrand
valerie.bertrand@ch-havre.fr

1 Pediatric Unit, Le Havre Hospital, BP 24, 76083 Le Havre cedex, France

2 INSERM U1073, UNIROUEN, Normandie University, Rouen, France

3 Department of Nutrition, Rouen University Hospital, Rouen, France

4 Autism Reference Center, Department of Psychiatry, Rouen University Hospital, Rouen, France

Table 1 Avoidant/restrictive food intake disorder (ARFID): DSM-5 diagnostic criteria

A. An eating or feeding disturbance (e.g., apparent lack of interest in eating or food; avoidance based on the sensory characteristics of food, concern about aversive consequences of eating) as manifested by persistent failure to meet appropriate nutritional and/or energy needs associated with one (or more) of the following:
1. Significant weight loss (or failure to achieve expected weight gain or faltering growth in children)
2. Significant nutritional deficiency
3. Dependence on enteral feeding or oral nutritional supplements
4. Marked interference with psychosocial functioning
B. The disturbance is not better explained by lack of available food or by an associated culturally sanctioned practice
C. The eating disturbance does not occur exclusively during the course of anorexia nervosa or bulimia nervosa, and there is no evidence of a disturbance in the way in which one's body weight or shape is experienced
D. The eating disturbance is not attributable to a concurrent medical condition or not better explained by another mental disorder. When the eating disturbance occurs in the context of another condition or disorder, the severity of the eating disturbance exceeds that routinely associated with the condition or disorder and warrants additional clinical attention

requested by a physician. He was severely undernourished (BMI 11.5, weight 31.9 kg, z-score: − 6.14 SD, height 168.5 cm, z-score: − 0.93 SD) (Fig. 1). Heart rate was 110/min, and blood pressure was 10/8 mmHg. He had no fever and no vomiting. His abdomen was bloated, with no mass on palpation. He had lanugo on his back. Dental condition was very bad (Fig. 2). Laboratory findings on admission showed hypokalemia, hypoalbuminemia, iron deficiency anemia, extremely low vitamin C and D levels, low levels of vitamin A, prothrombin and creatinine, and high urea (Table 2). An abdomen X-ray revealed major colonic distension measuring 11 cm (Fig. 3). The patient was hospitalized, fasted because of the paralytic ileus and risk of colic perforation, and perfused with a sodium chloride solution with low glucose concentration, enriched with potassium, phosphorus and vitamins to prevent refeeding syndrome. Most caregivers in the department suspected child neglect at this time.

The patient lived in an underprivileged social environment; however his two sisters were in good health, with good nutritional status and good physical hygiene. The mother explained that, since childhood, he had a history of constipation with encopresis, for which he had received laxative treatments and rectal enemas that traumatized him; the relationship with the physician was bad, and since then he refused to take any further treatment. No psychological evaluation had been proposed for this encopresis. He also had long-term selective and restrictive eating behavior, accepting mostly proteins and starchy foods, refusing fruits and vegetables, but the mother received no medical help for that. He had validated his middle school French certificate, but social integration at high school had been difficult. The mother explained he often refused to obey her, meals were long and difficult, his screen time was very high, and that she did not know how to manage her son.

During hospitalization, somatic and psychosocial care was provided. An abdominal scan confirmed Ogilvie's syndrome, without fecal impaction, volvulus or aorto-mesenteric clamp. Other causes of colectasia (*Clostridium difficile* infection, lead poisoning, porphyria, scleroderma, diabetes, hypothyroidism) were excluded. Screening for secondary causes of undernutrition and diarrhea (Crohn's and celiac disease, tuberculosis, thyrotoxicosis, glucocorticoid insufficiency, malignancies, cerebral disorders, genetic, cardiac and metabolic diseases, bacterial and parasitic infection) was negative. Careful rectal intubations and a short colonoscopy allowed a partial deflation of the left colon. Anorectal manometry showed high external anal sphincter pressure, rectal hyposensitivity and a megarectum. Colonic transit time was prolonged (147 h). Medullary MRI was normal. Finally, a secondary visceral myopathy, due to severe undernutrition, was suspected, which was confirmed after complete resolution of colectasia with refeeding (first parenteral then enteral nutrition). Stool consistency and encopresis also improved. A dental check-up revealed numerous cavities, four teeth were removed, and multiple pulpectomies and scaling were performed.

Questioning of the mother and the patient revealed that he displayed sensory disturbances since the age of 3 years (tactile, visual, olfactive) explaining that he could not brush his teeth, had food selectivity, chewing disorders, and could not walk on the sand or stand to have water in his ears. He also described a phobia of swallowing. A specialized oral therapy of his sensory disturbances was initiated, and progressively the patient managed to eat moderate portions, despite persistent selectivity. Psychiatric assessment did not find any history of physical or sexual abuse. Some stereotypies appeared with refeeding. Because of these sensorial, eating and psychosocial specificities, an ARFID–ASD association was suspected, and the patient was referred to an ASD reference center, which confirmed this diagnosis. Before the ASD diagnosis was confirmed, a social survey was conducted at home, following an initial physician alert; unfortunately, social workers had already applied to the judge for a foster care placement, as they did not understand this clinical situation. After explanation of the boy's illness, the request for placement, which could have aggravated the

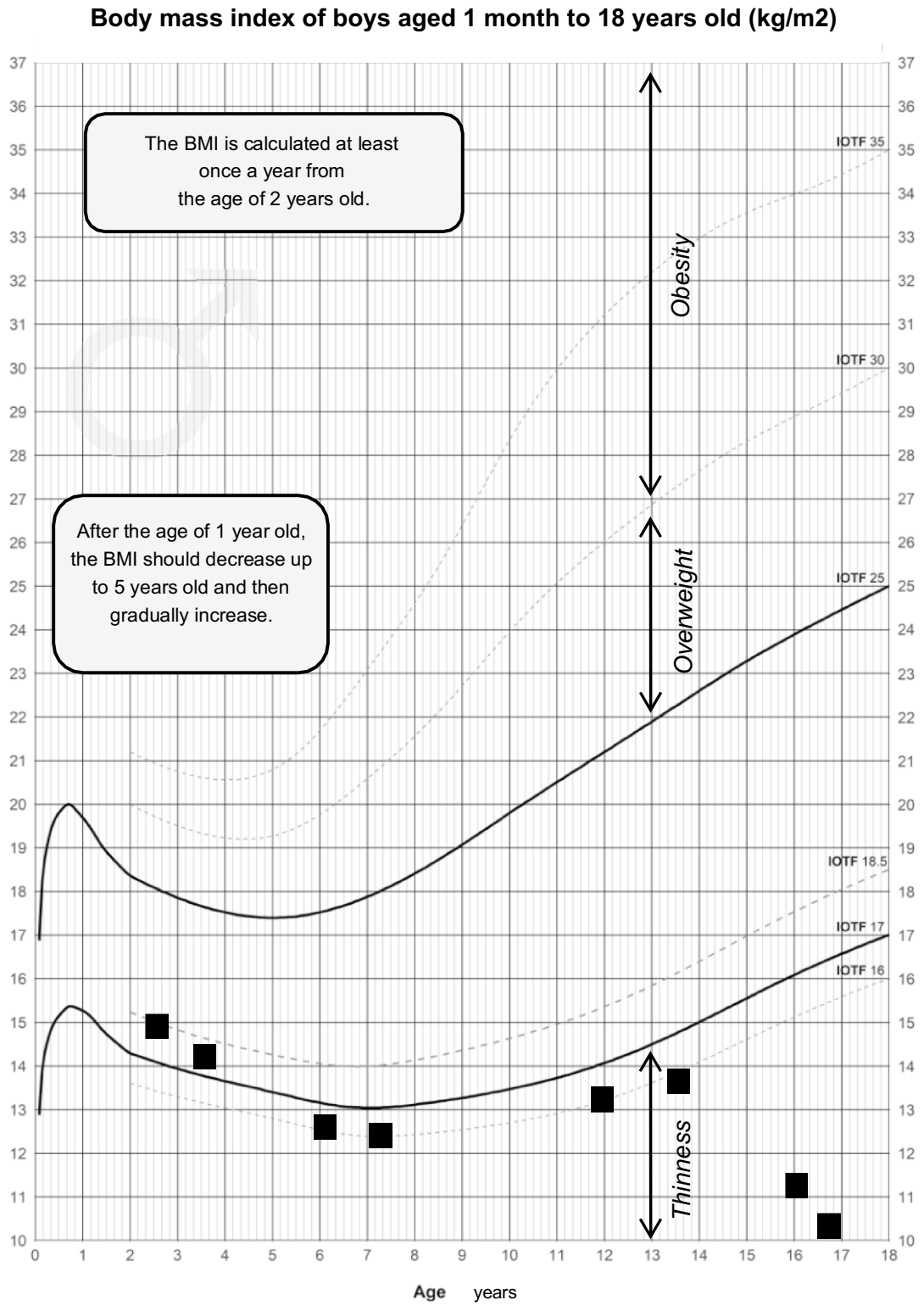

Fig. 1 Patient's BMI French growth curves

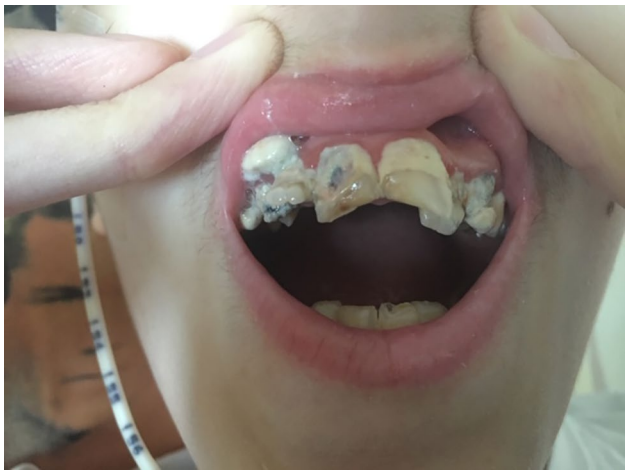

Fig. 2 Photo of patient's dental condition

symptoms, was canceled. A psychosocial and nutritional multidisciplinary care program was proposed, and he was able to return home after 5 months in hospital. During the early COVID19 pandemic, he relapsed and his food intake decreased, but a new hospitalization was beneficial. Currently the boy lives at home, and has a special needs teacher, professional follow-up, financial aid for disability, and nutritional and psychiatric care.

Discussion

This case illustrates the difficulty for pediatricians and general practitioners to identify ARFID in children, probably due to a lack of knowledge of ARFID. Indeed, pediatricians are more aware of feeding disorders in young children, and typical eating disorders in adolescents [AN, bulimia nervosa (BN)], and probably do not know well the DSM-5 classification. Without early diagnosis and adapted care, children with ARFID can be undernourished, especially if associated with ASD, with serious somatic complications, as reported in this case. Indeed, ASD is often associated with FED, mainly selective eating behavior, picky eating, food refusal, preference for specific textures or smells and also ARFID [5, 6].

ASD is generally diagnosed in children after the age of 4 years, when typical autism behaviors can be identified (echolalia, hand flapping, body rocking, language delay, etc.). However, diagnosis may be delayed for many years, when symptoms are limited. In this case report, the lack of major intellectual deficit and the low socioeconomic context may explain the fact that neither the pediatrician nor school professionals suspected an ASD. The investigation revealed, however, that in childhood he had problems with social interaction and communication, poor quality of eye modulation, some restricted interests, and walking on tiptoes. Moreover, as FED are frequently associated with ASD, severe food selectivity in a child (particularly < 10 different eating foods) should alert pediatricians to the possibility of an ASD and encourage them to screen for sensorial and social disorders. In this case, dental problems were more likely due to ASD because of poor hygiene due to major sensory impairment than to ARFID. The boy also had encopresis, which is often encountered in young children with ASD. Unfortunately, neither selectivity behavior with sensorial disorders nor encopresis alerted physicians when the boy was young. ASD can be also associated with other comorbidities including gastrointestinal problems (abdominal pain, constipation, diarrhea, bloating), immune

Table 2 Laboratory results at admission (Additional table for online-content)

Laboratory measures	Value	Reference range
Blood tests (abnormal values)		
Kalemia (mmol/L)	3.2	3.5–4.5
Albuminemia (g/L)	27	30.9–49.5
Hemoglobin (g/dL)	8.3	13–17
Mean cell volume (fL)	56.8	82–98
Ferritin (ng/mL)	5	22–322
Vitamin C (mg/L)	0.7	4.6–14.9
Vitamin D (ng/mL)	4.6	> 20 ng/mL
Vitamin A (µg/L)	195	257–715
Prothrombin level (%)	66%	70–100
Urea (mmol/L)	8.3	3.2–8.2
Creatinine (µmol/L)	41	44–97
Calcium (mmol/L)	2.12	2.18–2.6
Glucose (mmol/L)	6.9	4.1–5.9
Phosphorus (mmol/L)	2	0.78–1.65
Total white blood cell count (G/L)	12.4	4–10
Platelet count (G/L)	778	150–400
Blood tests (normal values)		
Erythrocyte sedimentation rate (mm/h)	5	1–15
C-reactive protein (mg/L)	2.2	0–5
Sodium (mmol/L)	136.3	132–146
Bicarbonates (mmol/L)	21	20–31
Chlorides (mmol/L)	100	99–109
Serum lipase (UI/L)	17	12–53
Liver transaminases TGO, TGP (UI/L)	9, 17	9–40, 13–40
Alkaline phosphatase (UI/L)	85	52–171
Gamma-glutamyl transferase (UI/L)	8	0–73
Total bilirubin (µmol/L)	5	5–21
Creatine phosphokinase (UI/L)	35	46–171
Vitamin E (mg/L)	7.5	5.6–10.3
Vitamin B12 (pg/mL)	299	211–911
Vitamin B9 (ng/mL)	6.07	> 5.38
Cholesterol (mmol/L)	4.2	< 5.18
Serum lactate dehydrogenase (UI/L)	317	208–378

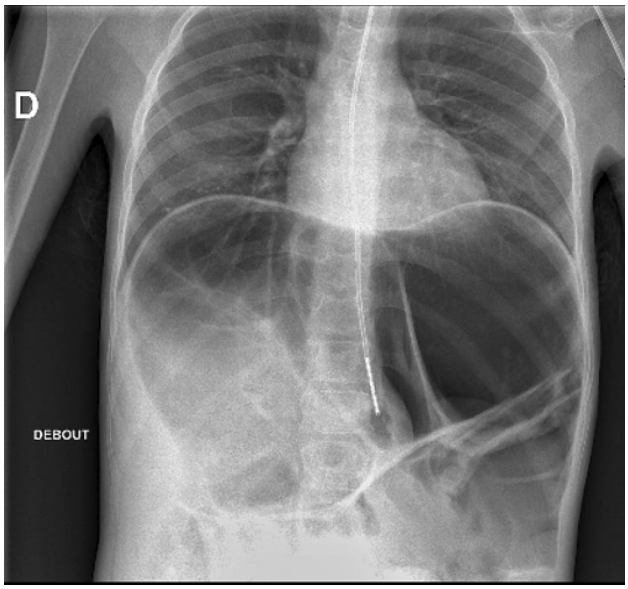

Fig. 3 Abdomen X-ray at admission showing colectasia (11 cm)

disorders, epilepsy, sleep problems, attention-deficit/hyperactivity disorder, anxiety, and depression. When ASD is suspected, children should be immediately referred to an expert ASD center. Indeed, early diagnosis and intervention from the age of 2 years are associated with a better intellectual, behavioral and functional prognosis [7].

ARFID is described, from the age of 1 year until adolescence, and in adults. ARFID have frequent psychiatric comorbidities (attention-deficit/hyperactivity disorder, anxiety). Gastrointestinal symptoms are also frequent (abdominal pain, reflux, nausea, diarrheas) [8, 9], and digestive diseases (achalasia, celiac or Crohn's disease) must be excluded. To our knowledge, this is the first case of ARFID and ASD with Ogilvie's syndrome. This syndrome, also called "acute colonic pseudo-obstruction", is rare, and can result in a severe cecal perforation in the absence of treatment. Usually, it is mostly described in adults with malignancy, sepsis, systemic illness, electrolyte imbalances, narcotic usage, or surgery [10], but any form of very severe undernutrition may lead to colonic distension. In this case, we suspected that the association of ASD and ARFID induced a severe undernutrition. The diagnosis of ARFID could have been made in early- to mid-adolescence when weight was falling off the growth curves, and should have prevented this severe digestive complication, by preventing undernutrition. Severe nutritional deficiencies are rarely reported in ARFID: some cases in adolescents reported scurvy or vitamin B9 B12 deficiencies, including one subacute degeneration of the spinal cord due to B12 vitamin deficiency [9, 11, 12]. A recent literature review of ASD–ARFID cases with vitamin deficiencies found 76 published cases in children and adolescents (vitamin C 69%, vitamin A 17%, and vitamin B12, D, B1), and only 22.9% were underweight [13]. However, as ARFID is still not well known by health professionals, we hypothesize that nutritional deficiencies are poorly screened and, therefore, still poorly described in the literature. A child with suspected ARFID (marked restrictive or selective eating behavior) should be referred to an FED specialist. Screening for nutritional deficiencies should be systematic. Despite the fact that there is currently not well-established treatment, many interventions are advised: counseling of parents (avoid pressure to eat, structure mealtimes), dietetics, desensitization with oral and exposure therapy, relaxation, and cognitive therapy in older children. Treatments such as olanzapine, mirtazapine, and cyproheptadine have been proposed [9, 14].

ASD and FED are generally not mistaken for cases of neglect. However, ASD may lead to child physical neglect and FED can be associated with a history of physical, sexual or emotional child abuse, in particular for AN, BN or binge eating disorders. To our knowledge, there is no published case of ARFID mistaken for neglect, which may reflect the recent characterization of ARFID in DSM-5. In this case, the question of the existence of care neglect was relevant, but in defense of the mother, she had been very poorly attended by health professionals when the child was young, whether by the pediatrician or the family doctor. She found herself helpless afterward, and thereafter no psycho-educational support was offered to the patient, either by the pediatrician, the family doctor or the school professionals during all these years. When the boy was a teenager, the underprivileged socio-familial context probably delayed access to care and so contributed to the severe clinical condition.

In conclusion, the training of health professionals in the clinical forms of pediatric FED, including ARFID, is necessary to promote early diagnosis and to prevent poor nutritional and psychopathological outcomes. It is important for physicians to be aware of this type of clinical presentation to screen for ASD in young children with selective eating, and conversely to screen for FED in children with ASD, especially when weight is falling off the growth curves. The association of these two severe psychiatric comorbidities, the delay in diagnosis by primary care professionals and the delay in access to specialized care favored by the disadvantaged social environment led to severe somatic complications in adolescence in this case.

What is already known on this subject?

ARFID is mainly known by mental health professionals. Patients generally have a higher BMI than patients with anorexia nervosa, and moderate associated gastrointestinal symptoms.

What does this study add?

Delayed diagnosis of ARFID and ASD can induce severe undernutrition and gastrointestinal symptoms like Ogilvie's syndrome. Training of pediatricians in the clinical forms of eating disorders is necessary.

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Author contributions VB conceptualized and designed the case report, collected and analyzed data, drafted the initial manuscript, and revised the manuscript. CD analyzed data, and critically reviewed and revised the manuscript. PD analyzed data, and critically reviewed and revised the manuscript for important intellectual content. MF analyzed data, and critically reviewed and revised the manuscript for important intellectual content. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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Declarations

Conflict of interest The authors have no conflict of interest to declare.

Ethical approval The authors have checked that they complied with the specific and ethical requirements of their institution.

Informed consent Informed consent of parents was obtained to publish the minor patient's data.

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