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Severe and enduring anorexia nervosa: Update and observations about the current clinical reality

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Abstract

Several objectives underlie the current article. First, to review historical diagnostic issues and clinical strategies for treating SE-AN. Second, to provide an overview of recent evidence informed strategies and clinical innovations for the treatment of SE-AN. Third, based on the authors' collective clinical and research experience, we offer eight observations that we believe capture the current clinical experience of patients with SE-AN. Some of these observations represent empirically testable hypotheses, but all are designed to generate a meaningful discussion about the treatment of this group of individuals with eating disorders. Finally, we hope to call clinicians, scientists, professional organizations, advocates, and policy makers to action to attend to critical issues related to the care of individuals with SE-AN. We believe that an international discussion could clarify areas of need for these patients and identify opportunities for clinical innovation that would enhance the lives of individuals with SE-AN and their families.

KEYWORDS

anorexia nervosa, disordered eating, eating disorders, severe and enduring anorexia nervosa

Anorexia nervosa (AN) is most common in girls and young women with a lifetime prevalence in women of up to 2–4% (Galmiche, Dechelotte, Lambert, & Tavoracci, 2019; Keski-Rahkonen & Mustelin, 2016; Smink, van Hoeken, & Hoek, 2013). The highest prevalence of AN has been

reported in Europe, North America, and Australasia with increasing reports from Asia (Hoek, 2016; Nakai, Nin, & Noma, 2014; Thomas, Lee, & Becker, 2016; Tong et al., 2014). AN is a very rare disorder in Africa and Latin America, while other eating disorders are common on

these continents (Kolar, Rodríguez, Chams, & Hoek, 2016; van Hoeken, Burns, & Hoek, 2016). AN has a crude mortality rate of 5% per decade and a standardized mortality ratio of around 6 (Arcelus, Mitchell, Wales, & Nielsen, 2011; Fichter & Quadflieg, 2016; Smink et al., 2013).

First-line treatments for AN typically consist of weight restoration and eating disorder focused psychotherapies, with family-based interventions for children and adolescents (Fisher, Skocic, Rutherford, & Hetrick, 2019) and various individual psychotherapies for adults (Brockmeyer, Friederich, & Schmidt, 2018; van den Berg et al., 2019; Zeeck et al., 2018). Antidepressant or antipsychotic medications have little or no effect and/or have low acceptability (Attia et al., 2019; Himmerich & Treasure, 2018). Inpatient and residential treatment is costly and reserved for those with the most severe form of the illness in many countries. Across the age range, and with best available treatments, recovery rates at 1–2 year follow-up range from 13 to 50% in recent trials (Brockmeyer et al., 2018; Fichter, Quadflieg, Crosby, & Koch, 2017). Up to 20–30% of AN patients develop a persistent and sometimes life-long form of the illness, often punctuated by a series of unsuccessful treatments (Dobrescu et al., 2019; Eddy et al., 2017; Herpertz-Dahlmann et al., 2018; Støving, Andries, Brixen, Bilenberg, & Hørder, 2011). This group of patients has come to be characterized as having severe and enduring anorexia nervosa (SE-AN) (Broomfield, Stedal, Touyz, & Rhodes, 2017; Hay & Touyz, 2015).

Our overall intention for this article is to provide an informative update on SE-AN and not a systematic review. We reviewed the most important recent articles related to SE-AN using PubMed, PsycInfo, Ovid database, and Google Scholar. Our specific aims were to: (a) briefly review historical diagnostic issues and clinical strategies for treating SE-AN, (b) provide an overview of recent evidence-informed strategies and clinical innovations for the treatment of SE-AN, and (c) offer eight observations by the authors that are thought to capture the current clinical experience of patients with SE-AN. Finally, we conclude with a summary of our thoughts and concerns about the status of SE-AN patients in our field and strategies to enhance care.

1 | ISSUES RELATED TO DESCRIBING AND DEFINING ENDURING FORMS OF ANOREXIA NERVOSA

Although AN is conceptualized as a disorder that typically lasts several years, considerable interest has emerged in describing and understanding clinical presentations that are enduring and often challenging to treat (Touyz, Le Grange, Lacey, & Hay, 2016). In a meta-analysis, Broomfield et al. (2017) emphasized two different descriptive components of this body of work: (a) different ways in which authors and clinicians have historically referred to or “labeled” this presentation, and (b) varying efforts to more formally define and classify enduring forms of AN.

Broomfield et al. (2017) delineate numerous labels that have been applied to enduring forms of AN. For example, terms including “chronic,” “severe,” “enduring,” “treatment resistant,” “recalcitrant,” and “long-standing” have all been used to characterize this population. Clinicians often express concern that these labels connote different

meanings, which may influence clinicians' perspectives on treatment, as well as patients' narratives about their illness and chances of recovery. For example, the labels of “chronic” and “treatment resistant” both may influence clinicians' views of a patient's treatability or willingness to collaborate in their treatment. Broomfield et al. (2017) indicate that while the label “chronic” is the most common term used to describe this patient population, other terms are emerging that may be less focused on an individual's treatability, such as “severe and enduring,” “longstanding,” and other variations of severity and duration labels.

The idea of creating a classification system for enduring variations of AN has also been discussed and our article builds on the pioneering work by the Australians Hay and Touyz (Hay & Touyz, 2015; Hay & Touyz, 2018; Hay, Touyz, & Sud, 2012) and other relevant papers (Ciao, Accurso, & Wonderlich, 2016; Wonderlich et al., 2012). Generally efforts to classify this population integrate three or four dimensions or variables. Examples of dimensions include duration of the illness, severity of the illness, evidence of unsuccessful treatments, and occasionally, patient age. Each of these dimensions poses difficulties for classification. For example, duration of the illness varies significantly across different definitions and has ranged from 3 to 10 years (Ciao et al., 2016). Similarly, unsuccessful treatment experience is rarely operationalized and is hard to define. Was the unsuccessful intervention an evidence-based treatment for eating disorders, any eating disorder treatment, any psychiatric treatment, or simply any type of clinical management? Also, the issue of patient age is important to consider, but difficult to place into formal classification. Whether a relatively young individual with AN (e.g., 18–22 years of age) should be labeled as “chronic” has caused considerable concern because of the labeling implications and possible impact on treatment (i.e., premature movement into a harm reduction model of care). Given the evidence that some individuals with enduring forms of the disorder ultimately do recover after decades with the illness (Eddy et al., 2017), the age and developmental status of the patient needs to be carefully considered when making treatment decisions.

Importantly, Hay and Touyz (2018) have recently provided clarification of the classification of SE-AN individuals with a proposed set of empirically testable criteria which includes a specific criterion for duration (i.e., greater than 3 years) that is similar to definitions of other enduring psychiatric disorders (e.g., psychosis; Ruggeri, Leese, Thornicroft, Bisoffi, & Tansella, 2000) and operational criteria for unsuccessful treatments (i.e., exposure to at least two evidence-based treatments appropriately delivered). Their proposed classification criteria are included in Table 1 and it should be noted that the authors

TABLE 1 Proposed criteria for “Severe and Enduring Anorexia Nervosa” (Hay & Touyz, 2018)

- (1) A persistent state of dietary restriction, underweight, and overvaluation of weight/shape with functional impairment
- (2) Duration of >3 years of anorexia nervosa; and
- (3) Exposure to at least two evidence-based treatments appropriately delivered together with a diagnostic assessment and formulation that incorporates an assessment of the person's eating disorder health literacy and stage of change

themselves recognize limitations in these criteria and strongly support empirical tests to clarify the best defining features of SE-AN (Hay & Touyz, 2018). Although there may be some debate about whether classification criteria and clear definitions are needed for SE-AN, it is worth noting that a recent practice guideline has been developed that does, in fact, propose alternative treatment strategies for individuals meeting diagnostic criteria for SE-AN (Hay et al., 2016). This development in a professional treatment guideline has significant implications for individuals with AN in general, and in particular for those meeting criteria for SE-AN. Given the clinical implications of such a taxonomy, efforts to enhance definitional clarity, along with empirical tests of the classification criteria, are encouraged.

2 | CLINICAL RECOMMENDATIONS AND TECHNIQUES FOR TREATING SE-AN (1980–2010)

In the 1980s, clinicians who treated eating disorders began to recognize the complexity of SE-AN and offered explicit recommendations regarding the treatment of this subgroup of individuals with eating disorders. A series of papers and books emerged that outlined strategies for SE-AN treatment, which differed in meaningful ways from existing eating disorder treatments. These publications (e.g., Goldner, 1989; Hamburg, Herzog, Brotman, & Stasior, 1989; Robinson, 2009; Strober, 2004; Yager, 1992) delineated a collection of clinical ideas that the authors believed should be considered and implemented in the treatment of SE-AN. The treatment approaches were neither evidence-based nor rigorously tested in robust scientific studies, but were largely based on theoretical conceptualizations and clinical experience. The following themes or ideas were highlighted across this literature. First, clinicians were generally encouraged to tolerate and manage extreme ambivalence or resistance to weight regain in SE-AN patients, as opposed to focusing primarily, or solely on weight gain (Goldner, 1989; Hamburg et al., 1989; Yager, 1992). Second, patients with SE-AN were thought to be best treated in the context of a comprehensive, multi-disciplinary team that could provide varying levels of care (Hamburg et al., 1989; Strober, 2004; Yager, 1992). Some clinicians also recommended a treatment approach that might best be considered psychiatric rehabilitation, similar to that seen in other serious psychiatric disorders, such as schizophrenia (Robinson, 2009). Third, the authors clearly encouraged modification of treatment goals in SE-AN relative to more acute cases of AN. Rather than emphasizing meal planning or significant increases in food consumption, along with weight gain, these authors generally relied on defining alternative treatment targets (e.g., quality of life, social adjustment). Fourth, some authors (Goldner, 1989; Yager, 1992) encouraged clinicians to carefully consider the judicious use of legal interventions in SE-AN, particularly regarding involuntary commitment, legal incompetency, and patients below the age of majority. Finally, there was a general suggestion to include carers in the treatment of SE-AN patients. Family inclusion could allow a variety of meaningful interventions ranging from careful historical reviews of past treatment history, psychoeducation about eating disorders,

families coming to terms with the severity of the illness, and general support of the family.

In 2012, Wonderlich and colleagues synthesized this information and highlighted the lack of empirical evidence supporting any treatment for this group of patients. These authors noted the challenging and persistent difficulties often encountered in SE-AN treatment including serious psychiatric and medical emergencies, and repeated cycles of significant behavioral change followed by serious relapse. Thus, in 2012, questions were clearly emerging regarding the best approaches to treating patients with SE-AN and concerns that these patients were often receiving a relatively unfocused intervention with uncertain intensity, duration, goals, and a lack of integration across different levels of care. Around this time, empirical studies on course of SE-AN were beginning to be considered (Eddy et al., 2017) and importantly, a randomized, controlled trial was being conducted in Australia that focused on SE-AN (Touyz et al., 2013). A summary of empirical studies on clinical course and treatment of SE-AN will be provided next.

2.1 | Empirical studies on clinical course and treatment of SE-AN

It has been widely assumed in the eating disorder field that long duration of illness predicts poor prognosis and treatment outcome in AN. However, studies on the prognostic effects of illness duration have produced surprisingly mixed results—some indicating an association with unfavorable treatment response (Steinhausen, 2002; Von Holle et al., 2008; Wild et al., 2016) and others suggesting negligible effects (Calugi, El Ghoch, & Dalle Grave, 2017; Raykos, Erceg-Hurn, McEvoy, Fursland, & Waller, 2018). It is worth noting that these studies vary significantly in terms of their definitions of SE-AN with some simply examining the effects of duration and others specifying categorical definitions of SE-AN. Furthermore, long-term follow-up studies (which now span intervals exceeding 30 years) reveal an illness trajectory characterized by progressively larger proportions of people recovering from AN over the long term (Dobrescu et al., 2019; Eddy et al., 2017; Keel & Brown, 2010; Nilsson & Hägglöf, 2005). For example, although it has been known that around 30% of AN patients recover in the first decade of the illness, recent data suggest that approximately another third of AN patients followed through a second decade will also recover (Eddy et al., 2017). Such observations challenge the ominous belief that individuals with SE-AN are “beyond help.” Studies examining the specific question of treatment responsiveness in individuals with SE-AN lend even more direct support to the idea that many SE-AN patients may indeed benefit from structured, short-term treatments.

2.2 | Treatment studies of SE-AN

In 2012, prior to the availability of any randomized controlled trials (RCTs) evaluating specific treatments for SE-AN, Hay and colleagues reviewed 11 then-available RCTs involving at least some participants

with SE-AN (Hay et al., 2012). Based on their review, these authors concluded that specialist treatment yielded better results than did nonspecialist “treatment as usual” for patients with SE-AN. Their review also indicated that, in this population, CBT adapted for AN (CBT-AN) might reduce relapse, and that adjunctive olanzapine might enhance weight gain and symptom reduction.

Since 2012, there has been only one formal RCT addressing psychotherapeutic treatment of SE-AN, conducted in 63 individuals with AN of at least 7 years duration (Touyz et al., 2013). The study compared the efficacy of 30 sessions of an adapted form of CBT-AN to that of an adapted form of Specialist Supportive Clinical Management for AN (SSCM-AN), with adjustments in both treatments prioritizing enhanced quality of life and harm reduction, rather than weight-gain and symptom-reduction. Both treatments yielded improvements in quality of life and, unexpectedly, produced modest weight gains and reductions of eating symptoms for up to 1 year post-treatment. Furthermore, the rate of attrition from both treatments was very low—15% overall. In other words, the adapted treatments led to weight gains and improved AN symptoms. A secondary analysis of the same data found that younger age, lower chronicity of illness, nonpurging illness type, and better social adjustment at baseline predicted better outcomes (Le Grange et al., 2014). Yet another secondary analysis showed that good therapeutic alliance predicted positive response on both eating-specific and generalized symptoms (Stiles-Shields et al., 2013). Together, findings from this study demonstrate that SE-AN can be treated, and point to the importance of designing treatments that optimize patient engagement.

Although not RCTs, several studies have examined the value of what is probably the best-established psychological treatment for EDs—namely CBT-E (Fairburn et al., 2009) in the treatment of SE-AN. The first, by Calugi and colleagues (Calugi et al., 2017), compared outcomes in 66 adult patients with AN who were subdivided into groups with illness duration less than, or greater than, 7 years. All participants received CBT-E as a component of inpatient treatment. Results showed that both groups displayed similar increases in body mass index (BMI) and improvements in eating symptoms and general psychopathology. After discharge, both groups showed similar rates (upwards of 40% for completers) of good outcome (BMI greater than 18.5 kg/m²) and 12-month “minimal ED symptom” rates were 32 and 33%, respectively. In a related vein, Raykos et al. (2018) examined associations between effects of illness severity or duration upon outcome in outpatient CBT-E. In a sample of 134 AN patients, it was observed that neither illness severity nor duration predicted outcome—such that patients with more severe or long-standing illnesses did as well in CBT-E as did other patients. Although the latter study was limited by attrition from follow-up, an implication is that chronicity of illness may not be as detrimental to outcome as has been thought.

Another study involved the Maudsley Model of Anorexia Nervosa Treatment for Adults (MANTRA), which is a manual-based, multimodal outpatient treatment for adults with AN (Schmidt et al., 2015; Schmidt et al., 2016; Schmidt & Treasure, 2006) recommended by NICE as a first line treatment (Hilbert, Hoek, & Schmidt, 2017;

National Institute for Health and Care Excellence, 2017). Although there have been no RCTs specifically testing MANTRA as a treatment for SE-AN, Wade, Treasure, and Schmidt (2011) documented a study of 33 outpatients treated with MANTRA in which completers had a mean illness duration of over 8 years. Improvements in BMI and eating symptoms were reported at 12-month follow-up and, notably, duration of illness was not a significant predictor of quality of outcome.

In another study, secondary analysis of a multisite clinical trial of adult outpatients with AN ($N = 187$) provide a contrast to the findings just-reviewed (Ambwani et al., 2020). Trial participants all had been offered NICE-recommended psychotherapies augmented by a short, digital treatment (Cardi et al., 2019). Patients were grouped into two categories: those in an “early stage” (illness duration <3 years; $n = 60$) and those representing a “severe and enduring” stage (SE-AN; $n = 41$), defined by level of distress (Depression, Anxiety and Stress Scale; DASS-total ≥ 60) and illness duration (≥ 7 years). Baseline comparisons between “early stage” versus SE-AN patients indicated poorer work/social adjustment and higher ED symptomatology among SE-AN patients. Comparisons between assessments conducted at baseline and then after 12 months indicated higher rates of improvement on work/social adjustment among “early stage” patients. Of note, SE-AN patients had higher rates of accessing intensive services (inpatient or day treatment) than did those with “early stage” illness.

3 | CONTEMPORARY CLINICAL INNOVATIONS IN SE-AN

3.1 | Building the evidence base

Novel behavioral, pharmacologic, and service delivery approaches to treatment are being developed and tested to address illness persistence or chronicity. Some of these attempt to impact broader issues, such as harm reduction or improvements in quality of life. Furthermore, some emerging interventions are being proposed specifically for SE-AN on theoretical grounds (i.e., as targeting possible illness maintenance factors) whereas others are being proposed for SE-AN, but not in more acute AN, because they are very resource intensive in terms of service delivery costs and personnel time required, and may have to be reserved for treatment of the most severe cases.

3.1.1 | Behavioral treatments

A range of behavioral interventions that have emerged from studies of acute AN, including altering fear learning processes via exposure or reconsolidation approaches (Murray et al., 2018) or reversing anorexic habits in relation to maladaptive food choices and more general habit-proneness (Steinglass et al., 2018), are being discussed as potentially useful for SE-AN. To the best of our knowledge, these have yet to be specifically tested in patients with SE-AN.

Cognitive inflexibility and excessive detail focus have been extensively studied in AN (Smith, Mason, Johnson, Lavender, & Wonderlich, 2018) and are thought to be associated with more enduring illness (Lang, Stahl, Espie, Treasure, & Tchanturia, 2014). One RCT involving 82 adult in-patients with a severe and/or enduring illness AN spectrum illness, studied effects of a 10-session cognitive remediation therapy (CRT) that used a range of cognitive exercises to improve cognitive flexibility and information processing (Dingemans et al., 2014). All patients received about 30 hr per week of comprehensive inpatient care with multiple treatment components, and half of these patients also received CRT as an adjunct to their treatment. Outcome measures assessed set-shifting, central coherence, ED symptoms, general psychopathology, motivation, quality of life, and self-esteem. Although the CRT adjunct did not promote more rapid weight gain, patients who received adjunctive CRT showed larger improvements on ED-related quality of life at the end of treatment and on ED symptoms at 6-month follow-up. However, a more recent trial by the same group comparing CRT with an active control therapy, both added to AN treatment as usual, did not find CRT to be superior to active control treatment (van Passel et al., 2020).

3.1.2 | Pharmacologic interventions

A range of novel medications is being considered for reduction of symptoms in SE-AN. For example, evidence has emerged demonstrating a role of oxytocin in the AN disease process. The operative mechanism may be related to homeostatic and reward-related processes involved in the regulation of food intake, but also via pathways involving attachment experiences and social cognition, all of which are impaired in AN (Giel, Zipfel, & Hallschmid, 2018; Thaler et al., 2020). Likewise, use of ketamine preparations to treat the depression that commonly accompanies SE-AN is being discussed (Price & Duman, 2019). Neither oxytocin nor ketamine have to date been trialed in SE-AN.

One small cross-over RCT tested the adjunctive use of the appetite-enhancing cannabinoid agonist, dronabinol, in 25 women with AN of at least 5 years duration. Participants were randomized to treatment with either dronabinol-then-placebo or placebo-then-dronabinol, added to treatment as usual (Andries, Frystyk, Flyvbjerg, & Støvring, 2014). During dronabinol treatment, participants gained 0.73 kg more, on average, than during placebo. In other words, the active drug induced small, but significant increases in weight gain without adverse events.

3.1.3 | Brain stimulation interventions

Neurobiological models of AN tend to focus on an altered balance between neural mechanisms related to reward and those related to cognitive control/inhibitory systems (Dalton et al., 2018). These models have motivated the development of neurostimulation techniques in EDs, both as illness probes and as potential treatment

modalities. Neurostimulation has been defined as "any intervention intended to alter nervous system function by using energy fields such as electricity, magnetism, or both" (Dalton, Campbell, & Schmidt, 2017). Contemporary therapeutic neurostimulation methods use a range of devices to enhance or suppress brain and neuronal activity for the treatment of disease, including the noninvasive methods of repetitive transcranial magnetic stimulation (rTMS) and transcranial direct current stimulation (tDCS) and the surgical method of deep brain stimulation (DBS).

Several single case studies, case series, and proof-of-concept RCTs of noninvasive brain stimulation methods (rTMS, tDCS) in AN have shown promise, improving ED symptoms, mood and decision making and are safe and well-tolerated in AN (McClelland et al., 2016; McClelland, Kekic, Campbell, & Schmidt, 2016; Van den Eynde, Guillaume, Broadbent, Campbell, & Schmidt, 2013). Recently, a feasibility sham-controlled double blind randomized controlled trial (RCT) of 20 sessions of high frequency rTMS to the left DLPFC was conducted, in 34 SE-AN patients (17/group) (Dalton et al., 2018). Treatment (vs. sham) effect sizes (Cohen's *d*) of symptom change (baseline to 4 months) were small for BMI ($d = 0.2$) and ED symptoms ($d = 0.1$), medium for quality of life and moderate to large ($ds = 0.61-1.0$) for mood. In an 18-month open follow-up, improvements in mood were largely maintained and 45.5% of the original real rTMS group were weight recovered (BMI > 18.5 kg/m²) versus only 9.0% of the original sham group (Dalton et al., submitted). The delay in BMI improvement may be due to the delayed effects of TMS on neuroplasticity (Cirillo et al., 2017).

DBS involves the surgical implantation of electrodes in targeted brain structures so as to allow for controlled, local electrical stimulation. The procedure has been studied as a treatment option for severe and refractory AN by several different groups, using different stimulation targets, including the bed nucleus of the stria terminalis, the subcallosal cingulate, the nucleus accumbens, or the ventral/capsule/ventral striatum (Sobstyl, Stapinska-Syniec, Sokol-Szawłowska, & Kupryjaniuk, 2019). The largest series to date included 16 patients with chronic and treatment-refractory AN (Lipsman et al., 2017). Subcallosal cingulate DBS was associated with an average BMI increase from 13.83 (*SD* 1.49) at pre-treatment to 17.34 after 12 months of stimulation. DBS was also associated with significant improvements in depression, anxiety, and affect regulation. These preliminary results show that DBS may be a viable treatment option in SE-AN, although several non-negligible adverse events were also reported.

3.2 | Innovation in clinical delivery systems

A range of other interventions has recently been developed to reduce harms and improve quality of life in SE-AN. For instance, Munro et al. (2014) reported on the effectiveness of an intensive treatment for SE-AN, offered by an outpatient Anorexia Nervosa Intensive Treatment Team. The treatment, which applied supportive care and cognitive components, was associated with high patient and staff satisfaction ratings, good medical safety outcomes, and low attrition over

a 3-year period. A similar outpatient approach, the Community Outreach Partnership Program (Williams, Dobney, & Geller, 2010) offered treatment to patients in their communities using goals (set by patients themselves) focused on symptom management, skill development, and understanding benefits and risks of symptoms. After an average of 25 months, 18 “chronic” patients (mean duration of illness = 15.23 years) showed significant improvements in eating symptoms and weight, but not quality of life. In a similar vein, Long, Fitzgerald, and Hollin (2012) evaluated the effectiveness of a multimodal, multidisciplinary stepped-care approach (involving inpatient and then outpatient care) for adults with AN who had been described as showing severe symptoms and/or chronic lack of improvement in outpatient treatment. The intervention involved a program of individual, group, and family-based interventions that allowed for the setting of structured short-term goals, attention to leisure pursuits and community re-integration, and work with family members and partners. Results showed 44% of the patients to be rated as “in remission” at a 4-year post-treatment follow-up, based on a criterion requiring resumption of menses, BMI over 18, and relative absence of eating-disordered attitudes and behaviors.

Finally, although quantitative outcome indices are pending, two reports address a novel “self-admission” approach to treating SE-AN (Strand, Bulik, von Hausswolff-Juhlin, & Gustafsson, 2017; Strand, Gustafsson, Bulik, & von Hausswolff-Juhlin, 2017). In the program, two beds on an adult inpatient ward are earmarked for self-admission and self-discharge. Eligible patients must have undergone at least one inpatient treatment during the past 3 years, and to have subsequently been followed in the unit’s outpatient or day-treatment services. Patients are invited to complete a contract for self-admission. A qualitative content analysis of self-admission experiences of 16 AN patients with a mean duration of illness of 15 years (range 3–42) suggested that access to brief self-admissions increased feelings of security, supported development of healthier behaviors, and helped prevent needs for longer hospitalizations.

4 | OBSERVATIONS ABOUT THE NATURE OF ACTUAL TREATMENT DELIVERED TO SE-AN PATIENTS

Although the evidence base is growing and will eventually inform new and efficacious treatments for individuals with SE-AN, we now turn our attention to a topic that is more reflective of the current state of affairs. We hope that we have effectively conveyed that there are a significant number of individuals with AN who have a complex eating disorder that is psychiatrically, medically, and socially extremely impairing. Furthermore, empirically supported treatments for these individuals are scarce, yet they represent a portion of our patient population that is in great need of care. In this section, we reflect on the nature of treatments that individuals with SE-AN are actually receiving in 2020. Based on our clinical and research experience, we present a list of observations that we believe capture the essence of these patients’ typical clinical experience. We acknowledge that these

observations reflect our perspective only, and may not accurately or completely capture the current experience of all patients with SE-AN; however, our intention is to offer these observations in an effort to generate discussion about the nature of care generally provided to individuals with SE-AN.

1. Regarding the nature of SE-AN treatment actually received, we surmise that individuals with SE-AN are particularly likely to not receive active *eating disorder treatment*. We anticipate the reasons for this are many and include treatment burnout, loss of hope, absence of community resources, absence of a treatment model appropriate for their variant of illness, absence of financial resources to pay for treatment, deep ambivalence about recovery, and the hesitancy of clinicians to take on SE-AN cases because of their complexity and potential fatality. Moreover, many individuals with SE-AN may eschew treatment as they begin to feel as if AN is integral to their identity and cannot imagine a life without the illness (Tierney & Fox, 2009).
2. For those who do receive treatment, extreme heterogeneity is likely to exist with regard to treatment strategies for SE-AN. Given the absence of a solid evidence base for this variant of AN, the mode of treatment provided emerges primarily from expert opinion and may be significantly affected by available resources in the community both of which increase variability in treatment approach.
3. Protocols similar to those used in the Australian study (Touyz et al., 2013) are unlikely to be broadly implemented in the treatment of individuals with SE-AN for several reasons. First, few treatment programs are adequately resourced to provide training, supervision, outcome monitoring, and adherence measures while providing evidence-based structured interventions to complex patients. Second, many treatment centers may have relatively few individuals with SE-AN in their active treatment census, meaning that intensive training and adherence measures would not be cost-effective. Third, clinicians may develop resistance to delivering evidence-based or manualized interventions (von Ranson, Wallace, & Stevenson, 2013). Fourth, across countries, and even within treatment settings, clinician uncertainty about the best approach to treating individuals with SE-AN, can lead to an approach that is likely to include a nonspecialized, “eclectic” psychotherapy or counseling, possibly accompanied by medical monitoring, and pharmacotherapy with a multitude of treatment “targets.”
4. Although often not receiving specialist care for their eating disorder, individuals with SE-AN are particularly likely to be high utilizers of healthcare (Stuhldreher et al., 2015). Emergency room visits, medical hospitalizations, nonspecialist psychiatric hospitalizations, consultations with a variety of medical specialties can lead to un-coordinated and expensive care. Also, we speculate that hospitalizations of SE-AN patients are complicated because of the myriad medical and psychiatric needs of these patients. Furthermore, iatrogenic problems would be more common in SE-AN than in more acute, short-term AN cases, with more dangerous polypharmacy, contradictory techniques or interventions, and differences of clinical opinion across providers.

5. For those SE-AN patients who do seek care on eating disorder inpatient units, their unique needs may not be well met by programs that are typically designed for younger and more acute cases (Bamford, Mountford, & Geller, 2016). Moreover, the presence of SE-AN patients on the unit can have complicated effects on the milieu—on other patients, on parents and family members, and on staff. Younger patients can find the presence of SE-AN patients to be frightening (although for some it can be motivating for recovery). Parents fear that their less severely ill children will learn dangerous behaviors from the SE-AN patients and of course fear that their child might also take on a chronic and severe course. Staff can become demoralized when faced with caring for too many patients with SE-AN without a clear blueprint for expectations of the hospitalization.
6. Clinicians are particularly likely to diagnose individuals with SE-AN with other psychiatric disorders (e.g., mood disorders, anxiety disorders, personality disorders, psychotic disorders) given the variety of symptoms many of these patients experience. In the presence of such a psychiatric comorbidity profile, care needs to be taken to ensure accurate identification of the most significant disorders or problems and prioritize treatment strategies accordingly. Our observation is that in SE-AN, such careful diagnostic work and associated treatment planning does not occur, routinely.
7. Patients with SE-AN are particularly likely to report having experienced extreme or coercive efforts to increase body weight, typically in higher levels of care, only to be followed by “weight relapse” and a revolving door pattern of admission and discharge. Recent national data from Denmark (Clausen, Larsen, Bulik, & Petersen, 2018) indicates that nearly one in five AN patients has experienced involuntary treatment and a small number of AN patients (i.e., 2%) experienced over 100 involuntary treatments. Repeat attempts at refeeding that include limitations of privileges, intrusive observation, forced bed rest, and involuntary treatment measures can become unproductive and even traumatic for the patient, increasing the likelihood of them refusing future interventions.
8. Finally, in countries that rely heavily on private health insurance (e.g., United States), individuals with SE-AN, especially those who are receiving disability benefits, may not have private insurance, and have a greater likelihood of relying on public insurance for health care. Many facilities do not accept public insurance (Guarda, Wonderlich, Kaye, & Attia, 2018), and we anticipate that SE-AN patients are particularly likely to travel long distances to treatment facilities that accept public insurance (if they can access them at all). Of course, this also complicates effective discharge planning. Such conditions result in treatments that are not well integrated across providers and place a severe burden on patients and their families.

5 | SUMMARY AND FUTURE DIRECTIONS

The paucity of evidence-based treatments for SE-AN is striking and underscores how much the field has neglected this most severe and potentially fatal presentation of eating disorders. We see several factors that have contributed to this neglect, including professional

factors (training, education, support), political/financial (insurance, access to care, research funding), social (stigma regarding eating disorders), and geographical (absence of treatment facilities in close proximity to patients).

As a first step we need data to clarify the nature and boundaries of SE-AN to inform a possible diagnostic subcategory that can be considered for codification in subsequent versions of the DSM and ICD. That said, as stated by many master clinicians who have written on the topic, having SE-AN does not necessarily relegate one to a lifelong illness, as many can, and do, recover—at least partially. Critical research topics include how best to engage and retain people with SE-AN in treatment; how best to support parents and loved ones who are in caretaker roles; optimal tailoring of existing treatments or development of new treatments for SE-AN; and a deeper understanding of the specific biology of SE-AN and how it differs from more transient AN cases in order to advance the evidence base and drug discovery. As we discover more about the genetics and neurobiology of SE-AN, and how it differs from more transient cases, our ability to predict who might be at greatest risk of developing a chronic course may improve and inform personalized medicine approaches designed to obviate poor outcomes. Given that many centers only have small numbers of SE-AN patients on their caseload, collaborative efforts across several clinical research centers with complementary research resources in the genetics and bio-behavioral processes underlying SE-AN are required to achieve these scientific goals. Moreover advancing SE-AN treatment outcome research by the careful application of innovative adaptive designs may make better use of resources and facilitate translation to clinical practice (Pallmann et al., 2018).

In parallel to these efforts, we recommend enhanced clinical training in delivering care to individuals with SE-AN. The goals of this training would be to increase clinician confidence in treating this population, to guide the collaborative setting of achievable treatment goals, and to develop support or supervision paradigms to obviate demoralization and burnout. Furthermore, given the growing consideration of integrating palliative care or hospice into the treatment of some individuals with SE-AN, we believe that discussions with ethicists/legal experts, clinicians, scientists and patient/carer organizations, are needed to better conceptualize these approaches and develop protocols for clinicians, patients, and families to ensure the best interests of the patient are preserved. Furthermore, staff who work in palliative care and hospice may benefit significantly from educational activities regarding the nature and treatment of eating disorders.

We strongly encourage professional organizations and funders to acknowledge that the limited understanding of and paucity of treatments for SE-AN represent a crisis in our field. Patients with SE-AN report feeling marginalized and often distance themselves from treatment centers that are ill-prepared to meet their needs. Alternatively, SE-AN patients are also rejected by clinical centers due to limitations in health insurance and funding support. SE-AN patients who are aged 18 or over, may exclude parents from clinical decision making even when they are desperately ill and cognitively impaired, so privacy issues must be addressed as part of this crisis. With this article as a springboard, we hope to motivate action across sectors (policy, insurance,

professional) to develop a comprehensive strategy to appropriately address the needs of this challenging and neglected population.

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DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no new data were created or analyzed in this study.

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