



The psychological impact of adult-onset craniopharyngioma: A qualitative study of the experience of patients and clinicians

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ABSTRACT

Purpose: Individuals who experience social and emotional difficulties struggle to maintain successful social relationships and incur an increased risk of developing mood disorders. These, in turn, have a significant impact on psychological and physical wellbeing. A small number of medical studies suggest that patients with adult-onset craniopharyngioma (AoC) report poorer quality of life, however, no in-depth psychological research has been carried out. The present study aimed to capture a rich understanding of whether patients with AoC experience a psychological impact from their diagnosis and whether psychological factors may contribute to a poorer quality of life.

Method: Both patients with AoC and clinicians with experience of working with patients with AoC were invited to take part in a semi-structured interview. Participants were recruited from three geographically dispersed National Health Service (NHS) units across the United Kingdom (UK). Eight patients and 10 clinicians took part in the study. Interviews were recorded and transcribed verbatim and analysed using inductive thematic analysis.

Results: Two key themes, with multiple subthemes, were identified: 1) Patients experience psychological impacts of AoC; and 2) Patients also experience common physical symptoms.

Conclusions: Patients and clinicians recognised significant psychological impact as a result of AoC, and these impacts contributed to overall poorer quality of life. Crucially, both parties also felt that further research into psychological impact of AoC was both interesting and useful.

1. Introduction

Research has demonstrated that individuals who experience social and emotional difficulties are at an increased risk of developing mood disorders (Beck, 2008) and struggle to maintain successful social relationships (Fischer and Manstead, 2008). These, in turn, have been found to significantly impact psychological (Ishii-Kuntz, 1990) and physical wellbeing (House et al., 1988). A small number of medical studies (Dekkers et al., 2006; Wijnen et al., 2017) suggest that patients with adult-onset craniopharyngioma (AoC) report poorer quality of life, however, to date, no psychological research has been carried out. It is therefore important to understand if patients with AoC experience social and emotional difficulties, as they may be at risk of developing, or have already developed, mental health disorders.

Craniopharyngiomas are a rare type of brain tumour that cause disruption to critical structures including the hypothalamus, optic

chiasm, and ventricles and are commonly associated with endocrine dysfunction and visual impairment (Müller, 2014). Craniopharyngiomas present with a bimodal age distribution and can be split into childhood-onset craniopharyngioma (CoC), with incidence peaking between 5 and 14 years of age, and AoC, with incidence peaking between 50 and 74 years of age, with AoC accounting for between 50 and 70% of all cases (Müller, 2014). Direct treatment for the tumour, via surgical resection (with or without radiotherapy), is largely successful with a high survival rate (87–95%), however, research has demonstrated that patients experience long-term physical sequelae that, despite successful resection, continues to have a negative impact on quality of life (Müller, 2014). This medical research has naturally focused on the impact of physical health on patient quality of life, but mental health can also contribute significantly to one's sense of wellbeing (Health, 2009; Slade, 2010) and thus it is important to understand to what extent craniopharyngiomas are affecting patient's psychological functioning, as this

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may also be contributing their poorer quality of life.

Research investigating the psychological consequences of craniopharyngioma have tended to focus on CoC. A systematic review of this literature identified 11 relevant studies finding that, across studies, while physical health affected quality of life in 52% of patients, social impairment and emotional dysfunction affected quality of life in 41% and 40%, respectively, suggesting that psychological sequelae have a significant impact on patient wellbeing (Zada et al., 2013). Indeed, a recent study found that 34% of CoC patients self-report that they were receiving or had received psychological treatment (Sterkenburg et al., 2015). To date, however, only one quantitative psychological study has been carried out in CoC patients, finding that patients displayed the same degree of face aversion as autistic individuals (Hocking et al., 2020), providing one potential explanation for why CoC patients may experience difficulties with emotion regulation and social interaction.

Conversely, there is relatively little research investigating the psychological consequences of AoC. One potential explanation for this is because, by definition, AoC occurs later on in life and this may offer a buffer against the psychological impact of the tumour. For example, from a medical perspective, if AoC developed after a patient's fertility window, the tumour would not have influenced their fertility (to name one of many important biological processes occurring during adolescences and early adulthood). From a psychological perspective, adolescence is also a critical time, particularly for social development (Blakemore, 2008; Choudhury et al., 2006; Orben et al., 2020), and again, this critical window is largely unaffected for AoC patients. The small handful of medical studies that have investigated psychological aspects of AoC, however, question the extent to which this logic holds.

The first studies to investigate quality of life in AoC found that patients did indeed report poorer quality of life (Dekkers et al., 2006; Kendall-Taylor et al., 2005) and that this related to physical symptoms such as fatigue (Dekkers et al., 2006). A more recent study exploring medical records found that 23% of patients with AoC had a documented psychiatric condition and 20% had behaviour change (Wijnen et al., 2017). Crucially, these studies used established medical-based questionnaires or medical records in an attempt to measure psychological factors. However, medical questionnaires and medical records are (i) extremely limited in their ability to capture complex social and emotional phenomena; and (ii) likely to underestimate prevalence. One study that did employ well-established quantitative psychological techniques found that hypopituitary patients, 40% of whom had a craniopharyngioma, performed significantly worse on two tasks designed to measure empathy – one's ability to identify the emotions and mental states of others (Daughters et al., 2017). Thus, the limited research to date suggests that AoC patients also experience a poorer quality of life and that psychological functioning may contribute significantly to this outcome; however, the methodological limitations of these studies means that the full psychological impact of AoC is still not fully understood. Qualitative psychological research, on the other hand, offers a unique opportunity to capture, in detail, the lived experiences of patients with AoC.

The present study sought to capture, for the first time, a detailed description of AoC patients' lives outside the clinic. In particular, the study aimed to capture in-depth information about patient's social functioning and mental health. Both patients with AoC and clinicians with experience of working with patients with AoC were invited to take part in a semi-structured interview, a format that uses set questions to prompt discussion while also allowing flexibility to follow topics raised by participants. Clinicians were recruited with the view to providing an alternative viewpoint and the possibility to assess potential (mis) matches in perceptions of AoC patients' social strengths and difficulties.

2. Materials and methods

2.1. Design

The current study employed a qualitative approach using semi-structured interviews to capture participants experience with AoC. Both the study procedure and information reported in the paper follow the 'Consolidated criteria for reporting qualitative research' guidelines (Tong et al., 2007).

2.2. Participants

Patients and clinicians were recruited from three geographically dispersed NHS centres across the UK (to protect the identity of participants the specific centres will not be disclosed). Ultimately, eight patients and 10 clinicians were interviewed for the study. Table 1 provides a summary of the demographics of the two study samples. The study was approved by the NHS HRA ethics committee (21/WA/0079).

2.3. Data collection

Prior to data collection a separate interview schedule was prepared for patients and clinicians, but the same process was used to generate them, and they contained similar questions (see Supplementary Materials 1 and 2). An initial schedule was created based on previous research and the study aim, and was refined using a framework outlined in Braun and Clarke (2013, page 85). Each interview began by asking the participant basic demographic questions. The patient interview then asked about patients' discussions with their endocrinologists (e.g., "Has your endocrinologist ever discussed your psychological wellbeing with you?"), and both general (e.g., "Have you noticed any particular differences in your social life since undergoing treatment?") and specific (e.g., "Has your number of friends changes since your began treatment?") questions about their social lives. This was followed by questions about their emotional (e.g., "Do you ever struggle to control your emotions?") and mental health (e.g., "Do you have any mental health difficulties?"), along with general discussions about the impact of AoC on their life. The clinician interview asked about the clinician's specific experience with AoC (e.g., "How long have you been working with patients with AoC?"), about whether patients ever discussed social difficulty in the clinic (e.g., "Has a patient ever wanted to discuss their mental health or personal life during a consultation?"), and whether they had noticed any psychological impacts of the diagnosis (e.g., "Have you noticed any general psychological impact of posterior pituitary deficiency?"). Both interview schedules ended by asking participants whether they thought the research was important, what they specifically thought should be researched in future, and if there was anything they wanted to add.

Participants were recruited by a clinician at each NHS centre by circulating study adverts to appropriate patients and clinicians, the study therefore utilised a convenience sampling method due to the rarity of the condition. Participants who expressed interest were then sent the

Table 1

Sample characteristics of patients and clinicians who took part in the study.

	Patients	Clinicians
Sex	3 Male 5 Female	6 Male 4 Female
Age (years)	53 (31–72)	49 (37–74)
Years' Experience working with AoC		18 (9–28)
Age at diagnosis	44	
Years since diagnosis	12 (1–40)	
Hormone Replacement Therapy	8/8	
Radiotherapy	3/8	
Diabetes Insipidus	4/8	

*Bold values indicate means. Values in brackets indicate the minimum and maximum.

study information sheet and either contacted the researcher directly or gave approval for the researcher to contact them. KD then arranged all interviews via email and requested confirmation from clinicians regarding each patient's diagnosis. KD had no prior relationship with any participants before the study. Due to fluctuating COVID-19 restrictions during data collection, all interviews were conducted either by telephone or via zoom. The majority of participants took part from home, with some clinicians at work; all interviews were conducted individually and took approximately 1 h. Interviews were audio recorded and transcribed verbatim. Participants were not sent their transcripts for comments. KD also made field notes during each interview and reflected on these after each one. Data collection was finished once saturation occurred, which was discussed and agreed upon between KD and KU.

KD, a female lecturer (PhD), carried out all interviews. KD has a Masters in Social Science Research Methods which included formal training on semi-structured interviews but had not conducted interviews as part of her research before the current study, therefore, KD received additional training from KU, an experienced qualitative researcher, prior to interviews taking place. Although KD has conducted in-person quantitative research with endocrine patients in the past, including those with craniopharyngioma, she does not have *personal* experience of being an endocrine patient or visiting an endocrine clinic, and thus approached each participant as being an expert, able to provide in-depth information about their experiences. In order to address this clearly during interviews with patients, KD took time to explain that she was a researcher interested in finding out about their own unique experience and that as such patients were the experts (Berger, 2015). In so doing, it is hoped that patients felt empowered to share their experience freely, and therefore provided a more accurate representation of their social lives. For clinicians, KD explained her previous research experience with endocrine populations and the general motivation for the study, but also clearly stated she had no medical training and had never attended an endocrine clinic.

2.4. Data analysis

All interviews transcripts were uploaded into NVivo software (NVivo, 2012). Inductive thematic analysis (Braun and Clarke, 2006, 2013) was employed to analyse the data. In thematic analysis, codes are identified which represent statements centring on a similar topic that are endorsed across participants. Codes that are frequently used are compiled into larger themes, reflecting broader topics.

A subset (approx. 10%, n = 2) of transcripts were analysed by KU. Through a process of negotiated agreement (Campbell et al., 2013) coding differences were addressed, culminating in the creation of a preliminary coding scheme. All remaining interviews were then coded by KD using the formalised coding scheme which was continually refined as new codes emerged. The codes were then grouped into larger themes or sub-themes. These themes and sub-themes were discussed and refined through discussion with the research team.

3. Results

Two core themes emerged from the interviews: 1) Patients experience psychological impacts of AoC; and 2) Patients also experience common physical symptoms. Table 2 provides a summary of the themes and subthemes. For convenience, important aspects of each subtheme are highlighted in bold throughout the results section.

3.1. Theme 1 - patients experience psychological impacts of AoC

3.1.1. Core psychological symptoms

A number of core psychological symptoms were described by both patients and clinicians such as low mood, depression, and anxiety. Clinicians tended to report that "[t]he main thing that's come up are

Table 2

Themes and subthemes identified across both patient and clinician interviews regarding the impact of AoC.

Theme 1: Patients experience psychological impacts of AoC
Core psychological symptoms
Wider psychological impact
Protective factors against psychological symptoms
Discussing psychological wellbeing within endocrine clinics
Accessing professional mental health services
Theme 2: Patients also experience common physical symptoms
Core physical symptoms
Other physical symptoms
Psychological impact of physical symptoms
Individual differences

AoC; Adult-onset craniopharyngioma.

fluctuating moods, low moods, lack of motivation ...". Insight from patients suggest this is often related to a sense of loss of who they used to be: "I don't feel like I'm the same person I was before the diagnosis". When asked to describe the frequency of these periods of low mood, answers varied ("I would say at least every week." "... probably monthly or twice monthly ...") but indicated these could be relatively frequent, with several patients currently taking medication for depression. Despite this, the most common specific mental health condition referenced was anxiety. Taken together it was clear that patients did not feel they were the same person after their diagnosis, and this was supported by clinicians: "they're almost never like they were before ...".

It emerged that particular periods over the life-course of AoC could lead to poor psychological wellbeing, in particular the time around surgical intervention: "... immediately after the surgery I had a period where I was very down. As I said, very emotional. Um, probably borderline depressed if I'm honest ...". Indeed, the patients who struggled to come to terms with living with AoC as a chronic condition were more likely to describe (and be described as) experiencing low mood, which related closely to patients' sense of loss of their old selves.

Patients' self-esteem was also dramatically impacted by their diagnosis and associated changes: "I don't like myself very much" and "I'd sort of shied away from things... a lot of it did have some impact on my ability to socialize and feel confident enough to do something as well." Relatedly, some patients experienced, and clinicians reported, cognitive dysfunction, commonly referred to as 'brain fog', which was described as leading to low mood, anxiety (e.g., around medication) and low self-esteem (e.g., in social interactions).

3.1.2. Wider psychological impact

Various psychological and physical (Theme 2) symptoms had an impact on patients' social lives. Crucially, patients described a shrinking in their social network: "I found I have narrowed my group of friends." As a result, patients were engaging in fewer social interactions and for some, the interactions themselves had changed: "... I don't know if I'm interacting with people like I used to. Like I'm more aware sometimes I can say inappropriate things. And I don't cut off." There were also differences in close family relationships: "My husband kept saying to me 'I want my wife back'. I didn't... understand what he was trying to say." Changes in close relationships were also acknowledged by clinicians.

When asked about their emotional experience, some patients felt they struggled to understand other people's emotions, while others reported no concern. Patients described struggling to identify their own emotions, and in particular, to regulate their emotions: "... I didn't understand why I'd be angry or frustrated or why I'd burst into tears at the drop of a hat ...". Closely related to patients' low mood and depression, crying more frequently was a common symptom and also related to patients' sense that their overall emotional state had changed since their diagnosis: "... before... I would always be bubbly and positive ..."; something clinicians also identified: "... he's much more likely to end up sobbing. And

he... was never like that before he got the diagnosis, ever."

3.1.3. Protective factors against psychological symptoms

Importantly, there were positive, protective factors that were seen to reduce the chance of poor psychological wellbeing. Maintaining a sense of independence was viewed as being very beneficial both from clinicians: "... find it difficult to find work and then that... has an impact on their mental health." and patients: "... I went back to work... I never really realized how... beneficial that would be on my mental health." Patients who expressed mental resilience described psychological symptoms but the impact of these was less: "I have a low mood now. Like I say, I have to think... 'oh there are people worse off than me, so pick yourself up', you know."

3.1.4. Discussing psychological wellbeing within endocrine clinics

There was an interesting, and highly salient, miscommunication between patients and clinicians regarding discussions of psychological wellbeing during consultations. Patients firmly stated that their endocrinologist did not enquire about their psychological wellbeing/mental health: "[My endocrinologist] never asked about my mental health". Conversely, many clinicians believed that they did ask about mental health. Some clinicians stated that they always begin a consultation with general, open questions which they felt provided the opportunity for patients to raise any issues, including anything psychological, but patients felt these types of questions only referred to physical health or were "... more of a passing pleasantry rather than anything that's a detailed part of the conversation ...". Other clinicians stated they would only enquire about mental health if they were aware of a pre-existing condition/issue, but a few others acknowledged they didn't ask patients about their psychological wellbeing at all: "No, that doesn't specifically come up, to be honest with you, those are questions I don't ask ...". Clinicians identified that time constraints within the consultation were a barrier to discussions about psychological wellbeing. To compound the issue, they also felt that time was essential in order to develop rapport with a patient which was necessary in order for patients to feel comfortable raising psychological/mental health issues: "... patients might take a while to volunteer anything because it is so personal."

There were also differences in the extent to which patients wanted to discuss their psychological wellbeing with their clinician. Some patients felt very comfortable and wanted to discuss these issues, but others preferred to consult their general practitioner instead. That being said, patients were not always willing or able to accurately identify factors that may be affecting their psychological wellbeing. Clinicians noted that: "it'd be the partner [who would] actually raise some issues rather than the patient themselves." Indeed, within the interviews, patients didn't always demonstrate insight into their own mental ill-health despite describing common psychological symptoms or receiving treatment for psychological conditions: "No, I haven't had any treatment for depression. I'm on Prozac." In particular, some patients felt 'mental health' was an extreme term, and that one had to be "crazy" or suicidal for the term to be relevant, and it therefore didn't apply to them: "I'm not at suicide or anything like that it's just a real... sensation."

3.1.5. Accessing professional mental health services

Whilst patients had been referred within the NHS for mental health services, crucially, both patients and clinicians stated that this experience was negatively impacted by the level of resourcing available. One patient who was referred never received support due to severe time delay: "... to be honest, it took... so long for [the appointment for mental health support] to come through I never ended up doing anything with it." Clinicians expressed frustration at the extent of under resourcing: "The difficulty is... that it wasn't until... fairly recently we had that kind of [psychological] support. And not just in our own unit, but nationally, there is a lack of easy access and, to be frank, numbers of people trained to give the psychological support."

Interestingly, clinicians also felt that the traditional mental health

services available through the NHS was not appropriate for endocrine patients. They felt strongly that in order for a therapist to provide appropriate psychological support they would need a certain level of understanding about the patient's condition, and thus provide specialised psychological support: "I'm not sure how well that approach works to be fed into a, you know, a generic kind of primary care trust, run of the mill, kind of, psychology or therapy service where actually a degree of knowledge of, what admittedly, is an uncommon biological condition, is... helpful." In a similar vein, clinicians also felt they themselves were not trained to deal with psychological wellbeing/mental health and this led to frustration at not being able to help: "From our aspect is probably the hardest... situation for us because we can't do that much for them, I find. And I find that's really hard... psychological health is always going to be precious for them." Clinicians were, however, very open and receptive to implementing ways in which they could improve their services for patients: "if it facilitates me doing a better job, I'm always interested in that."

3.1.6. Theme 1 summary

Ultimately, AoC was described as resulting in patients making significant changes in their social lives: "... my social life used to revolve a lot around exercise. So that whole aspect of my life has gone." These changes often came as a result of core symptoms (both psychological and physical) but crucially these changes could lead to further psychological symptoms. When asked to reflect on how many of their patients experience the types of symptoms discussed above, clinicians stated "I would have said the majority".

There was also a strong feeling that craniopharyngiomas were extremely complicated to treat medically and was the worse condition the clinicians dealt with, specifically because of the impact the condition has on patient quality of life: "... for some [poorer quality of life] doesn't come, but for a lot of them it does... and it's only then that they realize actually what, excuse my French, f*****g b*****d condition it is. It is, it's about the most horrible condition I look after."

3.2. Theme 2 – patients also experience common physical symptoms

Core physical symptoms

The two most common physical symptoms were weight-gain and fatigue. Patients were very clear that weight-gain was the most important symptom: "Well my biggest concerns were rapid weight gain ... "; and clinicians also felt this was a big concern: "A big theme to my mind is in relation to weight and weight-gain." This was closely followed by fatigue: "Fatigue has come up a lot ... ", with patients stating: "... I'm always fatigued. I can get up at 11 o'clock in the morning if I'm not going [out]... [a]nd... have an hours sleep in the afternoon, I get very tired ...".

3.2.2. Other physical symptoms

Patients and clinicians also mentioned several other physical symptoms including low libido, with one patient reporting: "We've never had sex since [the surgery] ... ". For patients with diabetes insipidus, both patients and clinicians referenced having to manage the associated symptoms: "So, I have to make sure there's a toilet near all the time, because if I need to go, I gotta go then". Finally, patients also reported that they tended to get more headaches after treatment.

3.2.3. Psychological impact of physical symptoms

Importantly, when these physical symptoms were discussed, it was not the physical impact of these symptoms but rather the psychological impact that patients felt strongly about. For example, when discussing weight-gain patients discussed how it made them feel. Weight gain was strongly associated with poor self-esteem, low mood, and anxiety: "It's hard and it has knocked my confidence... I went to the park the other day with all my old... friends... I know they wouldn't say it, but I'm paranoid they're going to think 'oh christ, she's a lot fatter than she used to be'. But then I know they all know what I've been through. It's all me really, in my head. I don't like it. I just want to be back to my normal size." Similar

statements were also made by clinicians: "... it really does interfere with her self-esteem her weight and her interactions with her husband as well." These effects, in turn, had a negative impact on patients' social relationships and resulted in making lifestyle changes and even obsessive behaviour regarding food which was also noted by clinicians: "... so initially she had weight gain and, and then she suddenly became obsessed about losing weight."

Similarly strong associations were also found around fatigue, but with a particular impact on patient's social lives: "... it's hard to manage your energy when you don't know really how much you're going to have. Can't plan... a schedule. A friend asked if I want to go for a walk today and I'm so exhausted I can't, I'll have to cancel. Then I feel anxious about that because I'm cancelling, you know."

3.2.4. Individual differences

Clinicians were clear that there was one specific factor that determined how severe patient symptoms would be: hypothalamic involvement. Patients with extensive hypothalamic involvement were seen to be both harder to treat medically and seen to suffer the most severe symptoms, both physically and psychologically: "Every time she came in, it was just like 'oh my god, what did you do to this woman' because... her quality of life was awful."

4. Discussion

The current study employed qualitative psychological research methods for the first time to provide novel insight into the psychological impact of AoC. Thematic analysis of 18 interviews identified two core themes: 1) Patients experience psychological impacts of AoC; and 2) Patients also experience common physical symptoms.

Both patients and clinicians believed patients with AoC experience significant psychological impact as a result of their AoC diagnosis and treatment. Affective disorders such as anxiety and depression and sub-clinical or undiagnosed low mood were commonly referenced, supporting previous literature from both CoC (Sterkenburg et al., 2015) and AoC (Wijnen et al., 2017) populations. Crucially, however, patients also reported other psychological impacts, which included more subtle but important areas of social functioning. Both patients and clinicians noted changes in personality and emotional experience after treatment (including poor emotion regulation and increased propensity to cry), something that has only previously been noted in CoC (Riva et al., 1998; Zada et al., 2013). Psychological research has demonstrated that individuals who experience social and emotional difficulties are at an increased risk of developing mood disorders (Beck, 2008) and struggle to maintain successful social relationships (Fischer and Manstead, 2008). Ultimately, both can have a detrimental effect on one's psychological (Ishii-Kuntz, 1990) and also physical wellbeing (House et al., 1988). It is therefore important to acknowledge and conduct further research into both the mental health impact and more subtle areas of social functioning affected by AoC. Indeed, when asked all patients felt that further research into the psychological impacts of AoC would be useful, a sentiment that was echoed by clinicians. Patients and clinicians acknowledged a dearth of information and understanding regarding the potential psychological impact of AoC on patient's lives outside of the clinic. Patients expressed that simply hearing about other patients' experiences would be very reassuring. Thus, further research into the psychological impact of AoC is desperately needed and wanted.

In addition to developing potential psychological interventions, physiological interventions may also be appropriate. The hormone oxytocin has been found to play a vital role in the areas of social functioning identified in the current study (Bartz et al., 2011; Leppanen et al., 2017; Shahrestani et al., 2013), and there is some preliminary evidence that hypopituitary patients may present with low oxytocin (Daubenbüchel et al., 2016; Daughters et al., 2017). Thus, further research is required to investigate the therapeutic potential of oxytocin administration in this cohort (Bhargava et al., 2019; Cook et al., 2016;

Hoffmann et al., 2017).

Despite these psychological impacts there were two psychological factors identified that seemed to provide a degree of protection against, or reduce the risk of, poor psychological wellbeing. Both patients and clinicians identified that maintaining a degree of independence in a patient's life offered significant advantages. As such, restoring or maintaining vision as well as being able to return to work were of paramount importance. Relatedly, mental resilience was also identified as a protective factor. Although patients who demonstrated resilience referenced the same types of physical and psychological impacts, by employing resilience the negative impact of these symptoms was reduced. Further research should be conducted to explore both factors as potential avenues for possible intervention (Davydov et al., 2010; Ludolph et al., 2019; Rutten et al., 2013).

There was evident miscommunication regarding discussions of psychological wellbeing and mental health during clinic consultations. Patients felt strongly that their endocrinologist did not inquire about this, while many clinicians believed that they provided an opportunity to discuss mental health. To compound this problem, there were also other clinicians who avoided the topic of mental health, either due to time constraints (a long standing challenge (Dugdale et al., 1999)) or not feeling sufficiently qualified to offer psychological support. In both instances, patients are not able to discuss an important health concern with their medical professional. This void is at odds with government plans to strive for a "better quality of life as the central aim of clinical care" and that "interventions [should reflect] the psychological, social and physical needs of the service user" (9, page 60).

Crucially, however, even the clinicians who did actively discuss their patients' psychological wellbeing, described an increasing frustration with the process of referring patients for psychological support through the NHS. Mental health services around the world are under extreme pressure due to poor resourcing across a number of fronts, including human resources (Kakuma et al., 2011; Saxena et al., 2007). It is not surprising, therefore, that clinicians in tertiary care in the UK experience the same issues (Cummins, 2018), however, it does continue to highlight the need for further investment, particularly in light of the COVID-19 pandemic which has placed an even greater strain on these already limited services (Holmes et al., 2020; Hossain et al., 2020). Clinicians did note that mental health services were available for other patient cohorts, such as those coping with a cancer diagnosis (Hubert-Williams et al., 2018; Kadan-Lottick et al., 2005), and that these services were thought to be incredibly helpful for patients. Based on the current study, AoC patients may benefit considerably from access to these existing services. Particularly around the time of their surgery, which the current study highlights as a high-risk interval.

That said, clinicians with experience of referring AoC patients for mental health services within the NHS also raised concerns regarding the lack of specialised psychological support available. Clinicians strongly felt that in order for psychological interventions to be successful (or perhaps achieve greater efficacy) a degree of knowledge about AoC was required, something they felt was not currently available and which therefore suggests that mental health workers should receive training on AoC. Alternatively, clinicians may wish to engage in mental health training in order to support mental health services by engaging in "task shifting" whereby non-specialist health workers take an active role in supporting their patients' mental health (Kakuma et al., 2011). Future research should investigate whether both specialised psychological support or additional mental health training for clinicians may provide additive benefit compared to standard psychological care.

In line with previous medical studies (Dekkers et al., 2006; Patel et al., 2015; Poretti et al., 2004; Sterkenburg et al., 2015; Wijnen et al., 2017), the two core physical symptoms identified were weight-gain and fatigue. However, the present study provides novel insight into the psychological impact of these physical symptoms. Weight-gain, fatigue and low libido were strongly associated with poor self-esteem, anxiety, depression and wider social impacts upon close personal relationships

and social networks; and are comparable to previous findings regarding obesity (Luppino et al., 2010; Strauss, 2000) and sexual functioning (Trudel and Goldfarb, 2010). While tackling psychological symptoms directly should be a key target (as discussed above), given the extreme pressure on mental health services (Cummins, 2018), it may be both more practical in the short term, and within the medical expertise of clinicians, to aim to improve psychological symptoms *indirectly* by targeting core physical symptoms. This approach may be particularly beneficial if clinicians are able to explain the link between improving physical symptoms and improving psychological symptoms. The present study suggests that patients would benefit from hearing the experience of other patients and common symptoms, thus explaining the link between physical and psychological symptoms and the commonality of this experience may provide reassurance to patients that their experience is not unusual.

4.1. Strengths and limitations

Taken together the current study provides evidence of the complex psychological impact of AoC. There are, however, a few limitations that should be acknowledged. While qualitative psychological methods provide an excellent opportunity to gain a deep understanding of a particular topic (Braun and Clarke, 2006, 2014), and indeed this study provides, for the first time, an in-depth insight into the experiences of patients with AoC and clinicians who work closely with them an inherent limitation is that findings may not be generalisable to the wider population because of the sample size. Further quantitative research (e.g., survey) is therefore needed to confirm these findings in a larger sample. In addition, the study was completed between July 2021 and January 2022 a time when COVID-19 was still very salient and included periods of lock down. It is acknowledged that this unrelated external factor may have an influence on results reported here, however, we note that clinicians had been working with patients with AoC for an average of 18 years and patients had had a diagnosis of AoC for an average of 12 years. Thus, asking them to reflect on their experiences *since* diagnosis or experiences in treating *all* AoC cases mitigated this potential influence as much as possible.

A strength of this study is the inclusion of both patients and clinicians. Although patients are best placed to speak to their own experiences, by interviewing clinicians the current study was able to highlight important similarities and differences in opinions which offer practical recommendations for future practice. These include: 1) Clinicians may wish to adapt their approach to ask explicit questions about a patient's psychological wellbeing; 2) Clinicians may choose to take part in mental health training in order to facilitate "task shifting" and provide additional mental health care in the clinic where appropriate; 3) AoC patients should be able to make use of existing mental health services currently available to other patient cohorts in neurosurgery wards such as cancer patients; 4) Existing mental health services may wish to engage in workshops or training events to aid their understanding of AoC to better support their clients; and 5) Clinicians should be empowered to share the findings of this study with patients to provide information and reassurance.

5. Conclusion

The current study found a significant psychological impact of AoC. This impact ranged from discrete mental health disorders to more subtle but profound effects on interpersonal relationships and emotion regulation. Crucially, we do not make claims about the uniqueness of these experiences, indeed they are not only common in AoC but other medical conditions. We stress, however, that because of this similarity patients with AoC should therefore have access to the same levels of mental health support as these other conditions. While these findings fill an important gap in the literature, they also demonstrate a need for further research into the psychological impact of AoC.

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CRediT authorship contribution statement

Katie Daughters: Conceptualization, Funding acquisition, Methodology, Investigation, Formal analysis, Project administration, Writing – original draft, Writing – review & editing. **Katy Unwin:** Methodology, Writing – original draft, Writing – review & editing. **D. Aled Rees:** Conceptualization, Project administration, Writing – review & editing.

Declaration of competing interest

The authors declare no conflicts of interest.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.ejon.2023.102346>.

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