

## Original Article

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**Abstract**

**Objectives.** Meningiomas are the most common, primary intracranial tumor and most are benign. Little is known of the rare patient group living with a malignant meningioma, comprising 1–3% of all meningiomas. Our aim was to explore how patients perceived quality of daily life after a malignant meningioma diagnosis.

**Methods.** This qualitative explorative study was composed of individual semi-structured interviews. Eligible patients ( $n = 12$ ) were selected based on ability to participate in an interview, from a background population of 23 patients diagnosed with malignant meningioma at Rigshospitalet from 2000 to 2021. We performed an inductive thematic analysis following Braun and Clarke's guidelines.

**Results.** Eight patients were interviewed. The analysis revealed 4 overarching themes: (1) perceived illness and cause of symptoms, (2) identity, roles, and interaction, (3) threat and uncertainty of the future, and (4) belief in authority. The perceived quality of daily life is negatively impacted by the disease. Patients experience a shift in self-concept and close interactions, and some struggle with accepting a new everyday life. Patients have a high risk of discordant prognostic awareness in relation to health-care professionals.

**Significance of results.** We provide a much-needed patient-centered perspective of living with malignant meningioma: quality of life was affected by perception of threat and an uncertainty of the future. Perception of illness and the interpretation of the cause of symptoms varied between subjects, but a common trait was that patients' identity, roles, and interactions were affected. Shared decision-making and a strengthened continuity during follow-up could aid this rare patient group.

**Background**

Meningiomas are the most common primary intracranial tumor arising from the meninges surrounding the brain. Three grades of meningioma are defined based on the World Health Organization (WHO) classification of central nervous system tumors (Louis et al. 2016). Most meningiomas are indolent, slow-growing tumors, classified as WHO grade 1 meningiomas (approximately 80% of all meningiomas) according to the 2016 and 2021 WHO classifications. WHO grade 2 meningiomas comprise 15–20% of all meningiomas, and only 1–3% of all meningiomas are WHO grade 3 that can be defined as a primary cancer of the meninges: a malignant meningioma.

The incidence and prevalence of all meningiomas will increase in the future, partly because of more incidental findings and partly because of an aging population (Achey et al. 2019). Moreover, new mutational markers will likely result in an increase in the prevalence of WHO grade 3 meningiomas from 1–3% to 7–8%, as the new WHO 2021 classification includes TERT promoter mutations and homozygous deletion of CDKN2A/B (Louis et al. 2021). Thus, patients with malignant meningioma will most likely become more common despite their present rarity. The primary treatment for a malignant meningioma is surgical removal followed by adjuvant radiotherapy. Despite optimal treatment, the diagnosis is associated with significant morbidity and mortality, and up to 75% of malignant meningiomas recur within 1 year (Kshetry et al. 2015; Maier et al. 2020; Sá-Marta et al. 2021). Median overall survival in this patient group is approximately 3–4 years, but survival times vary widely between patients (Champeaux et al. 2015; Maier et al. 2022; Sá-Marta et al. 2021; Sughrue et al. 2011). The malignant meningiomas comprise a challenging clinical entity for several reasons. First, their rarity limits large datasets in which typical survival spans and outcomes can be assessed. Moreover, the cohorts hitherto compiled have highly varying survival and progression-free survival spans within

the cohorts (Champeaux et al. 2015; Sá-Marta et al. 2021; Sughrue et al. 2010). Second, limited treatment options are available, and targeted therapies and chemotherapy have not been successful in improving survival (Goldbrunner et al. 2021). Third, patients with WHO grade 3 meningioma are probably met with threat mitigating and threat enhancing information. The threat of the initial intracranial tumor is quickly mitigated by “most likely benign” radiological findings and information on how surgery can be curative. Radiology cannot distinguish meningioma phenotypes with certainty (Li et al. 2019). After a WHO grade 3 diagnosis, patients are referred to oncologists for radiotherapy and must face a life-threatening disease as WHO grade 3 meningiomas will recur and are classified as a cancer.

Studies on perceived quality of daily life are non-existing in patients with malignant meningioma. Perceived quality of daily life can be defined as a perception that reveals the subjective evaluation of everyday life (George 2006). Among all patients with meningioma, recent articles state long-term reduction in health-related quality of life (HRQOL) (Haider et al. 2021; Zamanipoor Najafabadi et al. 2017), a higher socioeconomic burden (Wirsching et al. 2020), and an elevated use of antidepressants (Thurin et al. 2021) following meningioma surgery. A recent study identified tumor size, histologic grade, and epileptic burden to be associated with worse HRQOL (Haider et al. 2021). Thus, patients with malignant meningioma are at high risk of experiencing these burdens and loss of HRQOL. Patients with benign meningioma have reported emotional distress after meningioma surgery and found themselves in a paradox: a benign tumor, that is potentially malignant, and facing extensive surgery as the only treatment (Wenström et al. 2012). Moreover, patients with intracranial meningioma need psychosocial support and specific information in the postoperative phase to aid recovery and improve HRQOL (Baba et al. 2020). In a known aggressive disease such as high-grade glioma (HGG), patients and their relatives have a life marked by loss and their life changes drastically (Collins et al. 2014; Piil et al. 2018; Sterckx et al. 2015). Focus on extending support and care beyond medical needs and implementing an improved multidisciplinary care model in HGG was addressed by several studies (Collins et al. 2014; Philip et al. 2015). However, in a disease with a unique cancer trajectory such as malignant meningioma, the perceived quality of daily living and unmet needs are unknown.

The question of how patients diagnosed with a malignant meningioma experience everyday living was approached by conducting semi-structured interviews with patients with malignant meningioma and their relatives. This study aims to (unprecedentedly) describe the perceived quality of daily life, disease course, and everyday life from a patient perspective in patients living with malignant meningioma.

## Material and methods

### Design

A qualitative explorative design was chosen, and 8 semi-structured interviews with patients living with a malignant meningioma were conducted followed by a thematic analysis.

### Setting

Eligible patients with malignant meningioma treated at the Department of Neurosurgery, Copenhagen University Hospital, Rigshospitalet and the Department of Neurosurgery at Odense

University Hospital, Odense, were contacted by telephone and were invited to participate in the study (combined catchment area of approximately 3.4 million people). No patients with malignant meningioma at Odense University hospital were alive at the time of inclusion. Patients were eligible if (1) they had undergone neurosurgical intervention for a malignant meningioma between March 2000 and October 2021, (2) they had the capacity to consent to participating research, and (3) their symptom status was acceptable for participating in an 1- to 2-hour long interview. Patients entering end-of-life care, younger than 18 years, and non-fluent in Danish were excluded. Patients with symptoms such as unmanageable pain, being bedridden and bed-bound, extreme somnolence, aphasia, and organic psychosis were excluded.

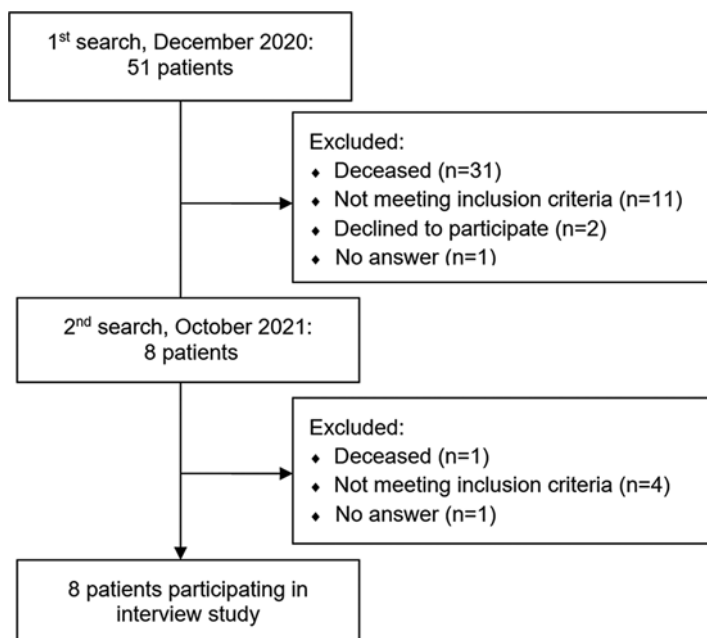
Study recruitment, inclusion, and interviews occurred from June 2021 to November 2021. Patients with malignant meningioma were identified by searching the Snomed Code M95303 in the pathology database. Two searches, 10 months apart, were performed (Fig. 1). Patients still alive were screened regarding general performance status and neurological symptoms by reviewing medical documentation. After screening, the patient's surgeon was contacted to discuss and confirm suitability and eligibility. Out of a population of 59 patients with malignant meningioma, 23 were still alive at the end of inclusion period and 12 were eligible. The patients were asked whether they would like their relatives present at the interview for support. Three patients had their relatives present at interviews.

### Interview

To ensure relevance for the study, a semi-structured interview guide was constructed based on existing literature on meningioma, clinical experience in the research team, and earlier experience conducting qualitative studies using semi-structured interviews. The interview guide was reviewed and revised by the entire research team. The topics of the interview included disease course and treatment, decision-making regarding surgery and radiotherapy, changes in social relationships, changes in everyday life, changes in subjective perception of quality of life (QOL) and thoughts of the future. We did not address HRQOL as an objective parameter but attempted to capture patient's first-person assessment of quality. During the interview, patients were asked to assess their perceived quality of daily life on a subjective scale from 1 to 10 (10 being the best and 1 being the worst) before and after being diagnosed with malignant meningioma, which prompted a discussion of changes in their QOL and everyday living. Additional examples of questions were as follows: Has your everyday life changed from before you were diagnosed with the tumor until now? Can you describe how? Can you describe what gives you quality in your everyday living? Will you tell me about the decisions made regarding your treatment? Did you feel involved in the decision-making process? – and how? Patients were asked to choose between in-person interviews, interview by telephone, or interview by video connection (due to geography and the COVID-19 pandemic). Interviews were audio recorded and transcribed verbatim by AM.

### Data analysis

The 8 transcripts were thematically analyzed by the research team. An inductive approach was used inspired by Braun and Clark's 6 steps: (1) familiarizing yourself with your data, (2) generating initial codes, (3) searching for themes, (4) reviewing themes, (5) defining and naming themes, and (6) producing the report



**Figure 1.** Inclusion of patients based on 2 searches, 10 months apart. First search in December 2020 yielded 51 patients of which 31 had died. The 20 patients still alive were screened regarding general performance status and neurological symptoms. Nine were identified as eligible and contacted by telephone. One did not answer, and 2 did not want to participate. Second search in October 2021, applying the exact same terms, yielded 8 additional patients, of which 3 were eligible and asked to participate. One did not answer, and 2 wanted to participate.

(Braun and Clarke 2006). Transcribed data were read thoroughly by all authors. Thematic codes for these themes were noted at every transcript reading at the appropriate text passage, and reoccurring themes were noted. Themes were generated throughout the reading process and no preset codes were used. Thematic codes and relevant text passages were gathered into a separate document, and overlapping themes were grouped together to form broader themes. All themes were reassessed to secure internal consistency and internal validity. This resulted in 2 themes being renamed to reflect the contents more precisely. The broader themes were again reviewed to ensure that there was no overlap in data supporting each theme.

### Methodological rigor

We used the 4 quality criteria as suggested by Lincoln and Guba (Lincoln and Guba 1985): dependability, credibility, confirmability, and transferability. Dependability was assured by having a stable research design and involving the entire research group in the research process, from conceptualization to interpretation of results. Credibility was assured by thorough analyst triangulation: discussing data and findings within the group and reaching an agreement. Confirmability was assured by checking and rechecking themes with all researchers. Moreover, confirmability was assured by assembling a diverse research team in terms of medical professionals (doctors and nurses) and at different levels of experience (senior researcher, PhD candidates, and post doc). Transferability was assured by providing contextual data about study setting.

### Ethical considerations

The Danish ethics committee were contacted, and there was no need for approval regarding this interview study (request 21012337). Participants were informed written and orally that study participation was voluntary, had no consequences for their treatment, and that they could withdraw their consent at any time. Identification of patients and data procurement was approved by the Danish Center of Regional Development (approval

R-21015041), and data storage was approved by Danish Data Protection Agency in the Capital Region of Denmark (approval P-2021-71). To meet Danish ethics regulations and ensure patient anonymity, details on disease course and demographics are limited to make identification of individual patients impossible. Typically, identified cohorts need to contain 5 or more individuals.

### Results

Out of 12 eligible patients, 8 volunteered to and were considered able to participate. Semi-structured interviews were conducted in Danish by phone ( $n = 1$ ), by video connection ( $n = 3$ ), or in person ( $n = 4$ ) by A.D.M., S.N., or R.G. as the COVID-19 pandemic prompted alternatives to in-person interviews. The interviews lasted on average 1 hour and 10 minutes (range 45 minutes to 1 hour and 35 minutes).

Among the interviewed patients, less than half were women. Most patients presented with a primary (de novo) WHO grade 3 meningioma. The average age was 71 (median 73 years). All patients had undergone at least 1 surgery for WHO grade 3 meningioma, and all had received radiation therapy ( $30 \times 2$  Gy). Symptoms included focal neurological symptoms (e.g., hemiparesis and visual impairment). Most patients had seizures managed with antiepileptic treatment, mental and physical fatigue, and cognitive impairment (concentration and memory problems). Of the few patients still in the workforce, all had to either diminish work hours or quit. Most patients had lost their driver's license for a period or permanently. No patients were bedridden with unmanageable pain or had extreme somnolence, as these later-stage disease symptoms were noncompatible with a 1- to 2-hour long interview.

### Perceived illness and cause of symptoms

This theme focused on the heterogeneity of perceived illness as revealed by very different explanations of the patients' disease, symptoms, and perceived cause. Participants perceived their illness, its gravity, and the threat to their everyday life very differently.

Their perception ranged widely from seeing tumor removal as a cure to accepting the disease as a cancer and reorientating their outlook on life because of the disease. Most participants could not describe the nature of their disease. Some were aware of the WHO grade and understood that their tumor was a grade 3 meningioma. This perception of illness influenced how the participants reflected on the changes in their perceived quality of daily life. Downplaying the seriousness of the disease and its impact seemed to hinder reflection, acceptance, and reorientation toward a new everyday life. Despite this heterogeneity, all patients identified intracranial surgery as a major threat. The patients' dominating perception of the disease was often not in line with the present knowledge about malignant meningioma:

"I shouldn't have been nervous, as I was certain that it was benign, as most of these tumors are. Right, and it was – the tumor was semi-benign. But that doesn't matter. I am completely cured today" (Patient 1).

However, others were deeply impacted by their diagnosis and reflected on their acceptance of a likely incurable disease:

"It took me many weeks before I could say that I was a cancer patient. That wasn't me. But it was. That was difficult, very difficult" (Patient 2).

All patients mentioned fatigue as a part of their disease course, especially in relation to radiotherapy, but for those patients still experiencing fatigue with the need of restructuring their everyday life (daily naps, recovering after social gatherings, recovering after moderate physical exercise), the perceived underlying cause was different. Some of the older patients were convinced that age was the main contributor to fatigue and restrictions in their everyday life. They reported not being as active as they wanted to and that the inactivity impacted their perceived quality of daily life, but they perceived aging as the main culprit. When one patient was asked why their perceived quality of daily life had gone from 8–9 before the diagnosis (7 years ago) and 4–5 at the time of interview, the answer was:

"Well, it has to be natural causes. Aging, and stuff like that..." (Patient 3).

The malignant meningioma disease was not mentioned by the patient when enquiring about this significant change. Pinpointing the reason for noticeable symptoms was often difficult. A patient, when asked about possible memory problems, was not certain what was aging, and what disease, 3 years after their diagnosis:

"I can't tell you what is what, about my memory. If it's this [points to head] or if it's my aging that makes it fail me. I can't tell you" (Patient 4).

Most patients were certain that the malignant meningioma was the main cause of their fatigue and described mental fatigue as well as physical fatigue. When asked about how fatigue manifested, mentally or physically, a patient answered:

"It's a combination, I would say. I become physically tired more quickly, but I am also brain-tired [mental fatigue]. And it comes suddenly, so that I cannot concentrate. Some things I mess up, purely intellectually, because I am simply tired" (Patient 2).

Other than age, we did not identify any common denominators in demographics or disease course regarding the perceived illness and threat to everyday life.

### **Roles, interaction, and identity**

The following theme highlighted the far-reaching impact the malignant meningioma had in patients' life, both in terms of roles (workload and responsibilities between relatives and patients), interaction (character and quality of communication with others), and identity (how patients perceived themselves).

Following surgery for malignant meningioma and subsequent radiotherapy, the patients' relatives had to manage everyday practical work and arrange transportation for hospital visits. The shifted balance in shared workload did, for most of the couples, partly normalize as time went on. However, not being able to drive impacted the relationship, and the patients were very aware of their dependency and toll on their relative. The limited freedom of movement often resulted in an ongoing shift of roles between relatives and patients. The transition from spouse to informal caregiver and practically managing the couple's daily living was strenuous on most relationships; the relative or spouse often experienced an increased workload with managing the house and daily living, including financial, medical, social, and caretaking decisions and tasks. Some patients and their relatives explicitly stated that they experienced more negative communication in their relationship, directly or indirectly coupled to the patient not being able to do what they wanted and normally did before the disease:

"I have a shorter fuse, yes, I do. It's probably because I am annoyed that I can't do this or that. Inadvertently and undeserving, it affects my spouse, and it shouldn't. I try to be conscious about it, but sometimes it can be difficult – we all know this" (Patient 1).

Both patients and relatives mentioned that the patient "snapped" more often and explicitly stated that this was a new, unrecognizable trait. At the time of surgery and radiotherapy, some couples actively sought psychotherapy to cope with the treatment and the changed life situation, whereas others did not. Patients mentioned support from friends and relatives at the time of surgery, but some were met with a misunderstanding of the lingering effects of surgery and disease:

"I quickly found out that I couldn't return to my everyday life. And everyone asked: When are you coming back, when are you coming back? Because you can't see that I am sick. I know that I am sick, and I know the things about my disease" (Patient 5).

For patients not able to return to work, or uncertain if they could in the future, their identity was impacted as they often took pride in their jobs and careers. However, most of the patients were not in the workforce anymore at the time of diagnosis. Here, the change in identity was mostly based on their role in their family and their expectations of how they would cope with retirement, as they often looked forward to an active, retired life:

"Going traveling, having my grandchild in the weekend, things like that. Having a social life together. But that was not how it turned out, no, no. But that is how it is" (Patient 3).

### **Acceptance and uncertainty of the future**

This theme revolved around the difficulty of coming to terms with the threat of the disease in combination with an unknown disease trajectory. When asked about changes in perceived quality of daily life, some patients were still struggling to accept a new reality of living with this disease. They were uncertain of how to accept this new reality and work actively with acceptance, as they did not know how



this disease usually manifested. For some, having an “invisible disease” reinforced their divided perception of their illness: still being alive, without severe neurological symptoms but living a different life having to cope with fatigue and loss of freedom of movement, either because of epilepsy or mild loss of strength. Patients reported perceived quality of daily life as poorer, overall:

“It is poorer. It is. I do not have the freedom of movement, I had. I miss that, I really miss that. But I can't really do anything about it, I can maintain my training, but it will never be good” (Patient 1).

Some patients struggled immensely during the waiting time until the operation:

Patient: “No, I did not want to live. Every time I met a doctor, I said: I don't want to wake up and be like a vegetable in a nursing home. And they all said, ah, but you won't. I had thoughts about ending this life because I did not want to live. That's how I felt”

Interviewer: And what was it, that made you feel like that?”

Patient: I think it was not knowing. And that I suddenly could not do what I usually do.” (Patient 6)

Another patient expressed a wish to talk with a neuropsychologist about experiences with the disease course until now:

“I have this thought because I need to talk to him [neuropsychologist] about how this has been until now [...] Maybe he can help me accept, how it was. Because I think, well I am brain-tired, but otherwise I don't think there is much wrong. Maybe my memory, a bit. I think it's coming more and more together now. And I have been in an enormous crisis.” (Patient 2)

The change in perceived quality of daily life and accepting possible permanent restrictions as a part of the future was difficult for most of the patients who perceived the malignant meningioma as a threat to their everyday life. A patient, who quantified change in perceived quality of daily life from 10 to 3–4, said, when asked about acceptance:

“Yes, it has been very difficult. And different people say so – “you have to accept it. And you *are* still here!” Well, yeah ....” (Patient 7)

The same patient, when asked about if they had any future dreams, answered:

“No, no. That would be getting my vision back, ha! And the balance and not being dizzy and things like that. But I think that's a closed chapter. And other than that, I do not have any dreams. Well, yes! Running a half-marathon. But that's not going to work.” (Patient 7)

Patients saw MRI scans as milestones in their disease course, but the possibility of facing a scan with a recurrence was associated with uncertainty and being emotionally overwhelmed:

“Would I have to have another surgery? They told me that that wasn't given. What will I do then? Will I have to live with it, can something be done – what will it affect?” (Patient 5)

### **Belief in authority**

Patients were often aware that their type of brain tumor was mostly benign, making the transition to having a malignant brain tumor that more difficult. Moreover, receiving radiotherapy for a tumor they understood as benign was confusing and shocking for some.

“They said it was a benign tumor, then why do I need radiotherapy? That was a surprise.”

Interviewer: “And did you get to know more...”

“No, I didn't. But I didn't ask, so.” (Patient 4)

Patients and relatives primarily received information from doctors and nurses during the disease course, and none mentioned actively seeking information on the internet. Few reported that they found out that meningiomas were a very common brain tumor, but they did not report about specific knowledge of malignant meningioma, other than it being a “category 3.” One couple explained how relieved they were, that it was not a “category 4.” Some patients had adapted a very passive approach to understanding the nature of their disease, almost giving the impression of not wanting to know.

Interviewer: “Do you know what it is?”

Patient: “No, I don't. I don't know. I do not understand it. Others [referring to healthcare professionals] understand it.” (Patient 3)

The passive approach was also reflected in answers when asked about decision-making, and one patient stated, “I leave that to science.” Most participants were convinced from the beginning of the disease course that surgery was necessary; however, a few were not convinced as they perceived the intracranial surgery as too big a risk to take. They were convinced by health-care professionals and relatives. Regarding radiotherapy, the perceived threat was not as grave as the intracranial surgery, and in most cases, they felt included in decision-making, and if not, they often stated that “there was no need.”

“There wasn't any doubt with the surgery, we agreed a hundred percent.

The radiotherapy ... I didn't feel very included in that way, but then I also didn't feel that I was kept out of it or pushed into it, I didn't. I just felt that I had to. Because I wanted to survive. Preferably in a good way, and that [the radiotherapy] was a part of surviving it.” (Patient 2)

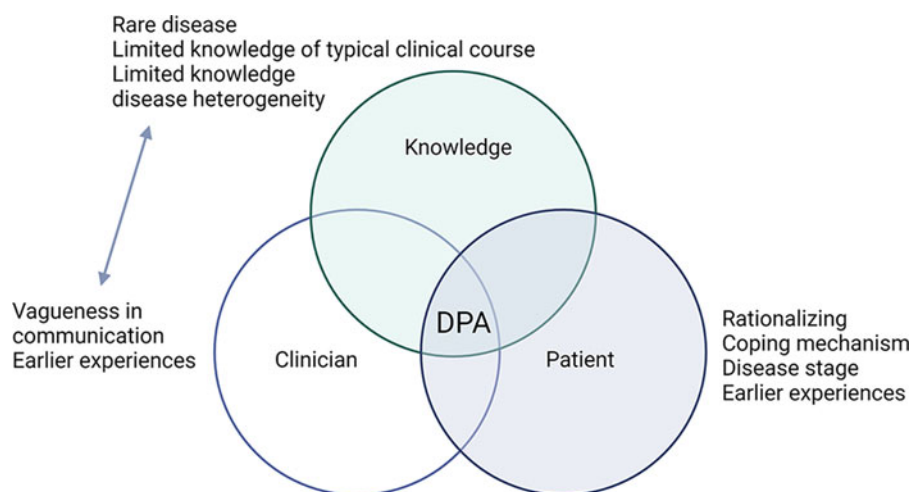
Several patients and relatives reported the immense disappointment, grief, and confusion of being told that their brain tumor most likely was benign but then not and that the transition to radiotherapy with 30 sessions was emotionally and practically difficult.

## **Discussion**

By conducting semi-structured interviews, we were able to identify specific challenges in patients living with malignant meningioma. We identified 4 overarching themes in the material: perceived illness and cause of symptoms; belief in authority; identity, roles, and interaction; acceptance and uncertainty of the future.

### **Possible discordant prognostic awareness**

We encountered 2 conflicting first-person perspectives toward prognoses. Despite being diagnosed with an objectively malignant tumor that is expected to result in death within 3–4 years, patients either identified with cancer patients or reported a very trusting and positive outlook (Achey et al. 2019; Kshetry et al. 2015; Sá-Marta et al. 2021). Some patients appeared unaware of the severity and future implications of their disease. Patients reported extensively altered roles in social interaction with limitations in everyday life and poorer perceived quality of daily life; however, some did not consider these effects as morbidity. We interpreted the latter reports to reflect discordant prognostic awareness. The phenomenon of prognostic awareness has been recently noticed and defined as awareness of a terminal prognosis or shortened life expectancy in



**Figure 2.** Possible factors contributing to discordant prognostic awareness (DPA) in malignant meningioma.

patients with advanced cancer, which is clearly applicable to our patients (Applebaum et al. 2014; Mone and Kerr 2021). The definitions of prognostic awareness, tools, and proxy parameters to investigate prognostic awareness are numerous (Applebaum et al. 2014), and consensus is lacking. Awareness is either considered as concordance between patient and health-care professionals or concordance between patients and an “objective reality.” We have only the patients’ understanding of the disease as “information received” and internalized consciously. We are unaware of the information that health-care professionals delivered. The simplest explanation of discordant prognostic awareness would be that the patients were not given the proper information after surgery (Figure 2). However, other explanations are possible. Potential patient-related factors include psychological defense-mechanisms such as denial or rationalizing a perceived threat among other coping mechanisms. After receiving the grade 3 diagnosis, patients may have rationalized and downplayed the severity because of an emotional need for normality (Yong et al. 2021), although they may have been informed that the intracranial tumor posed a major threat. Since most patients were interviewed after the initial surgery, they had no firsthand experience of recurrence or negative consequences of repeated surgeries; coping in a state of good physical health could thus easily include denial of bad prognosis. Still, it is highly likely that information given by health-care professionals was absent, imprecise, vague, or open to misinterpretation. Grade 3 meningiomas are rare, and health-care professionals may manage these patients as any meningioma patient, or they may themselves use defense-mechanisms to handle an existential threat to their patients. Vague or limited information of prognosis could thus be clinician-related in several ways. Even in benign meningioma, where knowledge on disease course is more consolidated, recent findings have shown an unmet need of information specific to post-surgical management in meningioma patients (Baba et al. 2020).

The balance of giving precise information on prognosis and being realistic, without removing hope and inflicting despair, is challenging. More adequate prognostic awareness is associated with higher existential QOL (Fisher et al. 2015) but also with depressiveness, anxiety, lower emotional, and physical QOL (El-Jawahri et al. 2015, 2014; Fisher et al. 2015; Janssens et al. 2019; Sato et al. 2018). Prognostic awareness and low QOL may be associated, although this link might reflect confounding since both parameters indicate advanced disease. However, prognostic

awareness could be prerequisite to cope, participate in treatment decisions, and maybe avoid futile surgery for repeated recurrences. Surgeries for recurrence of grade 3 meningioma seldomly improve neurological condition, and morbidity and mortality are high (Maier et al. 2022). Uncertainty and acceptance could thus cut 2 ways depending on the stage of disease, since uncertainty can foster hope while acceptance of inevitable fate at an early stage could shorten good quality life. Conversely, inappropriate hope at a late stage can lead to futile and potentially harmful attempts of rescue. Importantly, later uncertainty may be caused by the initial discordance when patients had been informed of surgery for a likely benign meningioma, which turned out to be a malignant tumor.

### **The need for personalized follow-up and shared decision-making in an ongoing personal crisis**

Patients with malignant meningioma expressed a strong belief in authority. Belief in authority is not necessarily associated with discordant prognostic awareness nor is it opposing a shared decision-making approach. Our study had a neurosurgical perspective. Surgery comprises the action of one person upon another (Wightman and Angelos 2014); a relationship of trust, where a patient trusts the surgeon for an expectation of rescue (Little 2001). Presence and proximity are prerequisites for this relationship (Little 2002). Thus, trust rather than paternalism may be reflected in belief in authority. A patient who is aware of their condition and a future non-curative stage could still have a strong belief in authority and practice shared decision-making if desired. Evidence of whether patients prefer shared decision-making for brain tumors is sparse and limited to HGG (von Essen et al. 2020). Shared decision-making is not preferred by all patients, and experience has shown that “one size does not fit all” (Halkett et al. 2010; Janda et al. 2008; Malmström et al. 2021). Prognostic awareness that was discordant between patient and health-care professional could cause distress and impact future planning of care in HGG (Halkett et al. 2010; Janda et al. 2008). In patients with HGG, discordance of prognostic awareness fluctuated during the course of disease and treatment (Sharma et al. 2021). Discordant prognostic awareness can also exist between patients with incurable cancer and relatives (Gray et al. 2021). One could expect even more fluctuation of prognostic awareness in patients with malignant

meningioma, given the lack of evidence, long-stretched disease course, and multiple recurrences in some patients. It is, however, fundamental for health-care professionals to remain sensitive in communication since need for information and participation varies with emotional distress and stage of disease (Enzinger *et al.* 2015).

Patients reported a significant, negative impact on their life and relatives' lives in terms of roles, interaction, and identity. Inability to perform as before the diagnosis, and uncertainty whether this state was permanent, put the patients and relatives in a state of limbo: an ongoing personal crisis. The situation was augmented by a lack of understanding from employers, friends, and sometimes relatives. However, tackling the crisis varied individually, reflected in statements of patients and relatives. Some expressed acceptance, while others were prognostically unaware, but still impacted, as discussed above. In essence, the patient's self-image and self-concept had changed. Self-concept was defined by Mead *et al.* (2015) as a relatively stable set of perceptions regarding physical features, roles, values, talents, emotional states, social skills, and limits that people hold about themselves. Acceptance of chronic illness was already challenging. This difficulty was further amplified by uncertainty of not knowing what the future brings in terms of clinical course and treatment options. Patients could probably be helped to understand and accept "the new normality" necessary to renew their self-concept by minimizing the cognitive dissonance of discordant prognostic awareness. A crucial element in coping could be discussions of the inherent uncertainty of the disease course with the aim of acceptance. In shared decision-making, cooperation provides for transactional communication (Turner and West 2017). Health-care professionals, patients, and relatives negotiate meaning simultaneously, participate in an active process of mutual understanding, and develop shared meaning. This transactional process is fundamental for the patient's development and evaluation of a new self-concept based on the dialogue with health-care professionals. It becomes increasingly clear that best management of patients with malignant meningioma include continuity and maintenance of dialogue at different stages of the disease, keeping in mind that the speed of disease progression can be heterogeneous. Regardless, at some point in the disease stage, management of grade 3 meningiomas must revert to palliative care (Elia *et al.* 2020). At this stage, concordant prognostic awareness and agreement on treatment goals are necessary.

### Recommendations

Traditionally, care for meningiomas is based on surgery and has a surgical perspective. In contrast to medical treatments and psychiatric treatments where long-term commitment is frequently an essential element of chronic disease management, surgery is more focused on cure and rescue (Little 2002; Wightman and Angelos 2014). This focus provides a danger in management of a chronic life-threatening disease such as grade 3 meningioma unless management is based on a commitment to continuity of care and observance of heterogeneous biology and personal values. At various stages throughout disease, neurosurgeons, oncologists, and neurologists have indispensable roles to manage the tumor, neurological symptoms, and existential challenges. Whether follow-up takes place at a neurosurgical, neurological, or neurooncological department is less important than realizing complementarity of the specialties and a need for cooperation between specialties and patient. However, a clear responsibility is important.

The interpretation of our interviews indicate that patients could benefit from a shared decision-making approach but also more broadly by approaching the communication to the patient from a transactional perspective with extensive continuity of care. Uncertainty, acceptance, and belief in authority were interpreted also to reflect a lack of continuity of care. During chemotherapeutic treatment of many cancers, a cemented connection to the outpatient clinic and oncological department is automatically established. Unfortunately, chemotherapies are mostly futile for grade 3 meningiomas (Kaley *et al.* 2014) and such a connection is not automatically established.

The key word in our recommendation is continuity. Grade 3 meningioma is a chronic disease where recurrences and final demise are likely. Especially, continuity is necessary for a shared understanding and prognostic awareness. We recommend regular follow-ups, not necessarily driven only by disease progression or MRI scans but also to manage the frequent neurological sequelae including epilepsy, headaches, and complex pain syndromes (Maier *et al.* 2022) and to maintain dialogue and establish a sharing meaning of existential values and individual paths to the patient's QOL. Follow-ups after recent diagnosis could include simply checking the patient's understanding of the nature of the disease to early disclose any potential discordant prognostic awareness. Later follow-ups could preferably include discussion and investigation of whether meaning-centered psychotherapy (Roberts and Applebaum 2022) or Acceptance and Commitment-based therapy (Fang *et al.* 2022) could be practically possible and beneficial; however, meningioma-specific evidence is lacking.

Finally, discordant awareness necessitates agreement on common body of clinical information. We have recently concluded that professional caregivers may underestimate the gravity of a grade 3 meningioma (Maier *et al.* 2022). Research to produce explicit knowledge on population-based long-term outcomes would facilitate congruent expectations from treatment and natural history and optimize treatment decisions.

### Strengths and limitations

This study is limited by the inherent uncertainty of generalization of qualitative data and a risk of pre-understanding. Yet, our findings and conclusions were at variance with expectations, which is a strength to assess internal validity. External validity is affected by limited availability of patients. To protect patient integrity, we could not disclose demographical data on an individual level; we adhered to the rule of not reporting data on groups of less than 5 patients. Our study population reflected the window of time between the WHO grade 3 diagnosis and deterioration beyond a clinical status that allowed a 1–2-hour interview. Thus, patients late in their disease course were not included. Relatives were present at some interviews for support but were not systematically included or investigated, which is an important next step in future studies into this patient population. Having only a few of the patient's relatives present limited the relative's perspective. Furthermore, the full depth of the toll on the relationship could not optimally be disclosed with the patient present as burdensome information from the relative might be held back.

Unlike benign meningioma, the malignant meningioma patient group does not have a female predominance, and the sex ratios differ in studies (Moreau *et al.* 2020; Sá-Marta *et al.* 2021). Our study included only few female patients. Moreover, we have interviewed patients treated in a single country and at a single institution, and management could be different in different departments. Trust in



social institutions is high in Scandinavian countries, while trust in health care varies across countries (Jen et al. 2010). World Value Survey (Institute for Comparative Survey Research 2023) reports the diverse distribution of several societal variables. Generally, trust in others is exceptionally high in Denmark (74% report “people in general can be trusted”) compared to the rest of the world (28%). “Trust” was equally high in all Nordic countries, while the attitude to authority differed. In Denmark, 63% report “trust in authority” to be a good thing, which is slightly higher than the rest of the world (56%) and much higher than the other Nordic countries (29–35%). Societal values thus appeared to agree with the acceptance of uncertainty, interpretation of symptoms as unrelated to disease, and the marked belief in authority we observed in our Danish patients with malignant meningioma. It may be that the semiology of disease interacts and is formed in an interplay with societal variables. Our Danish cultural context entailed strong trust in health-care providers reliance on the authority of doctors, which may to some extent reflect a unique cultural landscape. Our findings mandate surveys of the variation of values and perceptions of illness from other nations. It is probable that different strategies are warranted to empower patients for shared decision-making in different cultural contexts.

The main strength of our study is a stable research process based on a comprehensive insight into this rare and vulnerable patient population. This is the first exploratory study into the daily living and perceived quality of daily life in patients with malignant meningioma selected from a large background population. The focus on first-person perspectives for meningioma patients is novel, and a focus on first-person perspectives in malignant meningioma is unprecedented.

## Conclusion

Following diagnosis and treatment for a WHO grade 3 meningioma, patients reported a poorer perceived quality of daily living and a change of roles. They experienced a shift in their self-concept and close interactions. Malignant meningioma is a chronic and ultimately fatal disease associated with uncertainty. Patients showed belief in authority; however, we noted discordant prognostic awareness and a lack of continuity and care that may have affected coping. Uncertainty of the disease course could impact acceptance and reorientation. This rare patient population would benefit from extensive continuity of care, implementation of shared decision-making, and a strengthened but differentiated follow-up program.

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