

Acromegaly in remission: a view from the partner

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Abstract

Objective: A relative can be an asset in dealing with chronic illnesses, such as acromegaly, where quality of life (QoL) is altered even after remission. However, it has been shown that quality of life of caregivers can also be impacted. Our main objective was to compare the perception of acromegaly in remission in the patient–relative dyad.

Methods: In this observational study, 27 patients in remission and relatives were first asked to complete QoL, anxiety/depression and coping strategy questionnaires. Then, the patient's body image and self-esteem were evaluated from both the patient's and the relative's point of view using the same questionnaires with modified instructions.

Results: Relatives had overall an accurate estimation of patient body image using the Figure Rating Scale by Stunkard. However, there were wide variations between the patient's and the relative's responses regarding self-esteem and body perception. The QoL of relatives was not altered and was significantly higher in the social domain than for the patient.

Conclusions: Our results show that relatives require education concerning all the steps involved in the management of acromegaly, as they likely do not fully understand the sequelae of acromegaly.

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Introduction

In chronic diseases, health-related quality of life (QoL) has become an increasingly important aspect of patient care (1). For example, patients with active as well as controlled acromegaly report an impaired QoL, linked to the duration of the disease, to the necessity of prolonged medical treatment, to a history of radiotherapy or to persistent symptoms such as joint complaints (2, 3, 4). The partner (family member or close friend) can also be impacted by the patient's chronic disease (5). Indeed, this has been thoroughly described in both dementia and cancer patients (6, 7, 8).

The patient–relative dyad in pituitary disease has to date only been examined in two qualitative studies

(9, 10). Andela *et al.* conducted focus group interviews to explore the partners' perspective. They found that partners had negative beliefs about medication and felt that they were required to make adaptations in their own behaviour to accommodate their ill partner (10). Interestingly, partners requested more information about the disease and additional guidance. However, the consequences of acromegaly on the dyad have never been specifically studied.

The main objective of this study was to determine the partner's perception of acromegaly in remission via an original approach in which we asked him/her to imagine what the patient responses had been. Secondary

objectives were to determine the QoL of the partner, as well as anxiety and depression (comparing these with patient characteristics), and the coping strategies of the dyad.

Patients and methods

Patients aged 18–80 years, in remission and/or controlled by pharmacological treatment for 1–10 years, were recruited in the Reference Center for Rare Pituitary Diseases in the course of their follow-up at our department between June 2019 and August 2020. Remission was defined as follows: normal age- and sex-matched insulin-like growth factor (IGF-1) levels, random growth hormone (GH) <1 ng/mL and/or a GH level nadir after an oral glucose tolerance test (OGTT) of <0.4 ng/mL. Patients were considered controlled if they had normal IGF-1 levels and random GH <1 ng/mL on medical treatment. Patients with a known cognitive deficiency were excluded. The partner and/or family member was chosen by the patient. Informed written consent to participate in the study was obtained from all patients, and the study was approved by the ethics committee of Aix Marseille University.

Patients and relatives were asked to evaluate their QoL, anxiety, and depression by filling in the following questionnaires, from their own perspective:

- The French version of the generic scale, World Health Organization Quality of Life-BREF (WHOQOL-BREF) (11, 12).
- The acromegaly-specific scale, Acromegaly Quality of Life (AcroQoL) (13) (for patients only).
- The Hospital Anxiety and Depression Scale (HADS) (14, 15).

For self-esteem and body-image perception, the following questionnaires were first answered by the patient. We then studied the relative's perception of the patient's answers using the same questionnaires with modified instructions, and without knowing the patient's answers:

- Rosenberg's Self-Esteem Scale (RSES) (16, 17).
- Body-Image Questionnaire (BIQ) (18) and the Figure Rating Scale (FRS) (19).

This technique has already been used in the literature to study the perception of the patient's QoL by a relative in other chronic diseases (20), particularly in cancer and neurological diseases (21, 22).

Lastly, we explored the coping strategies of the patient–relative dyad using the Brief COPE situational

scale (23). We used a French version, regrouping these dimensions into four coping strategies: seeking social support, positive thinking, avoidance and problem solving (24).

Statistical analysis

Comparisons between the patient and the relative, in terms of QoL, were performed by univariate analyses, which were carried out on all variables of interest (including each QoL score, the four QoL scores of the WHOQOL-BREF (physical, psychological, social and environmental scores) and the total score for the AcroQoL) using the following tests: non-parametric test (Mann–Whitney *U* test), Spearman's Rho test and Kruskal–Wallis test (used for multimodal variables). BIQ, FRS and RSES evaluations from the relative were correlated with evaluations of the patient.

Data analyses were performed using Prism version 8.4.3 (GraphPad Software, LLC). *P* values < 0.05 were considered significant, and all statistical tests were two-tailed.

Results

Thirty-five patients were contacted; 27 patients (15 women and 12 men) agreed to participate and were enrolled in this study. Regarding the partner and/or family member, 77.7% of the relatives were partners and 22.2% were children.

For the socio-cognitive characteristics of the relatives, the median quality of life scores are detailed and compared to the patient scores in Table 1. No significant differences were observed except for one parameter, social relationships, for which the median score was 19 points higher in the relative questionnaire ($P=0.0435$). Likewise, we found no significant difference between the patient and family member HADS scores ($P=0.247$). Finally, the adaptation strategy which was preferentially adopted by the patient–relative dyad was positive thinking. Problem solving was in second position, and seeking social support in third. Coping strategy scores for the dyad are detailed in Table 1.

Regarding self-esteem and body image, there was no significant difference between the patient and relative views. However, there was a wide variation when taken from an individual viewpoint: with the exception of the FRS scores ($r=0.792$, $P < 0.001$), both RSES and BIQ scores showed no correlation between the patient and relative scores ($r=0.203$ and $r=0.080$, respectively) (Fig. 1).

Table 1 Quality of life and psychological characteristics of the patient and relative. Values are presented as median and Interquartile Range (IQR).

	Patient	Family	P
AcroQoL ^a	61 (20–98)		
WHOQOL-BREF ^b			
Social	56 (25–100)	75 (44–88)	0.0435
Physical	63 (13–94)	69 (31–100)	0.212
Psychological	69 (38–88)	69 (25–88)	0.886
Environmental	69 (50–88)	75 (44–88)	0.185
HADS anxiety ^c	6 (1–11)	5 (1–13)	0.247
HADS depression ^c	4 (1–11)	3 (1–12)	0.247
Rosenberg Self-Esteem Scale (RSES) ^d	33 (31–37.7)	33 (30–36)	0.983
Body-Image Questionnaire (BIQ) ^e	56 (51–64)	56 (42–66)	0.549
Figure Rating Scale (Stunkard)	5 (3–9)	5 (1–8)	0.370
Coping strategies (Brief-COPE) ^f			
Seeking social support		46.9 (35.9–53.1)	
Problem solving		50 (40.6–71.9)	
Avoidance		32.5 (28.7–37.5)	
Positive thinking		58.3 (50–66.7)	

^aScore from 22 to 110 converted to 0 (worst QoL) to 100 (best QoL); ^b26 items. Responses to each item are coded from 1 to 5, summed, and transformed to a scale from 0 (worst QoL) to 100 (best QoL); ^c14 items rated from 0 to 3. 7 items for each anxiety and depression component. Score from 0 to 21: > 8 possible symptomatology, > 11 certain symptomatology; ^d10 items: 5 for positive self-esteem (score from 0 to 4), 5 for negative self-esteem (score from 4 to 0). Score from 10 to 40: <25 very low, 25–31: low, 31–34: in the average range, 34–39: high, >39: very high; ^eScore from 0 to 9; ^fResults of the four factors Brief-COPE with transformed score from 0 to 100. *Giatus, volent aliquis di ne nis velit, si ilit harum libus voluptas id entiam quiam quis doluptia*

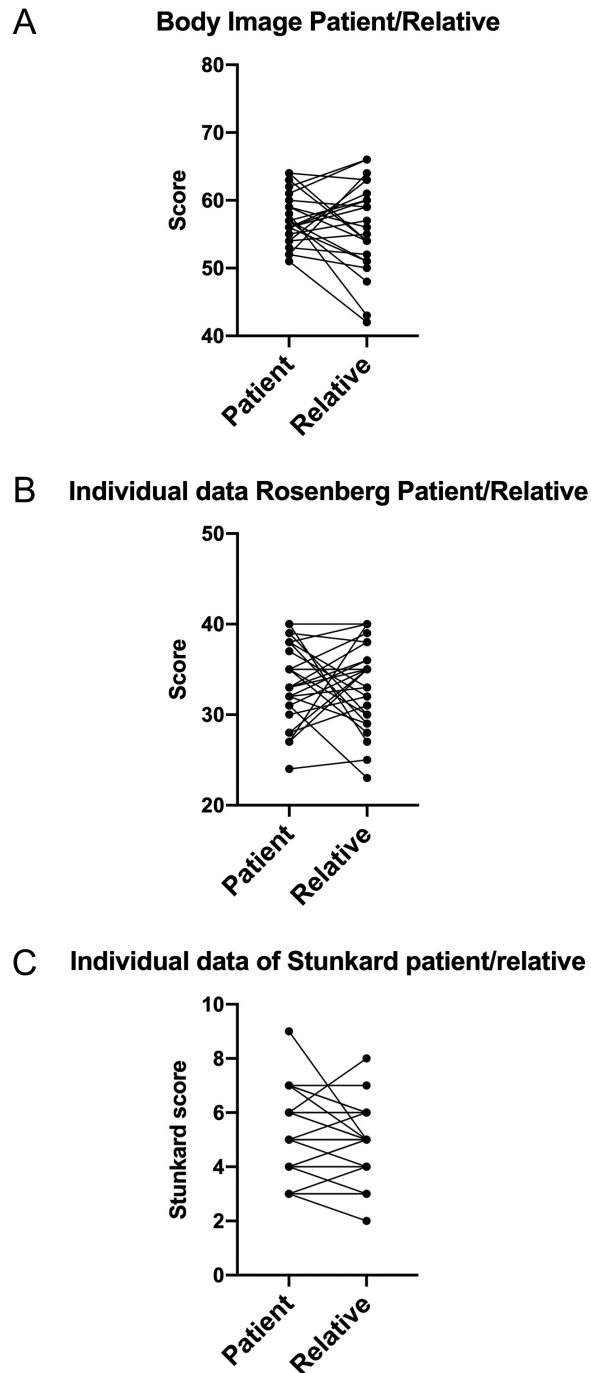
Discussion

Perceptions of illness can differ between the patient suffering from a chronic disease and his/her partner or relative. Our study showed that the patient relatives had a globally accurate estimation of that patient body image. However, even though there was no statistical difference between the patient BIQ and RSES scores and their relative's scores, major differences were observed when taken individually, as shown in Fig. 1. Family members of acromegaly patients may not be fully aware of the consequences of body changes on the self-esteem of their relative. In the study conducted by Andela *et al.*, partners reported viewing their relative differently but for other reasons such as changes in their relationship (partner becoming the counsellor or carer) or differences in coping mechanisms. Both patients and partners reported difficulties in communicating about the disease, both with doctors and their social network, resulting in a decrease in social interactions (10). Accordingly, seeking social support was only placed third in our results for the Brief COPE scale. Dunning *et al.* explored experiences of patients with pituitary disease and their partners by monitoring chat room discussions. They found that patients more openly discussed physical changes, appearance and feelings among their peers than they did with family or friends (9). Taken together, this suggests that we should use body image as a starting point to discuss the particularity of the patient's

disease with their relative/partner, either in consultations or in education programmes (25).

From our results, the relatives of acromegaly patients do not appear to have an altered QoL. We did find a difference of 19 points between the patient's and the relative's social WHOQOL scores, in contrast to previous findings (5). However, this should be interpreted with caution considering the high variability of answers and the small size of our cohort. A review of the literature identified difficulties experienced by family caregivers of cancer patients, including fatigue, sleep disturbance and anxiety or mood disturbances, highlighting the burden of caregiving responsibilities (7). Using specific scales such as the Caregiver Burden Scale and the Caregivers Quality of Life Index-Cancer, another study reported that the caregiver burden increased with anxiety, depression and poorer QoL, and that these results were similar to those reported in other chronic conditions (8). However, the majority of pituitary diseases are not malignant tumours, making a formal comparison difficult. Moreover, we cannot fully relate these observations to pituitary disease, although we could hypothesize that QoL scores might be different during active acromegaly when the burden is higher for the relative.

The main limitation of our study is that we were unable to analyse the medical history of the patients' relatives, as they were not inpatients of our institution. We thus cannot exclude that other diseases may have

**Figure 1**

Correlation scores between the patient and the relative for Body-Image Questionnaire (A), Rosenberg Self-Esteem Scale (B) and Figure Rating Scale by Stunkard's figurines (C). The relative was asked to complete the questionnaire as if he/she was the patient. Overestimation by the relative: 50% for the Rosenberg, 45.8% for the IBQ and 19.2% for Stunkard's figurines. Underestimation: 42.3, 54.1 and 38.4%, respectively. Accurate estimation: 7.7, 0 and 42.3%, respectively.

biased their QoL scores. However, our main criterion was not biased by this medical history. We also decided to include patients who were controlled by medical treatments or cured by surgery. Though we acknowledge that QoL may likely be different in these two groups of patients, this should not modify the partner results from an individual point of view. Finally, our results may be biased by the low number of patients in this pilot study. A large multicentric study will be necessary in the future to draw firm conclusions. Such a study would preferably use more robust methodological analyses, such as the Actor-Partner Interdependence Model, Mutual Influence Model, Common Fate Model and structural equation modelling, as these would allow variable interdependency to be taken into account.

In conclusion, our study is the first to analyse the consequences of acromegaly on the patient–relative dyad in terms of QoL and self-esteem. Our results support the development of education programmes that are centred not only on the patient but also on the familial environment. In a disease such as acromegaly, in which physical and psychological sequelae can likely persist for years after remission, the overall acceptance of the disease will likely be improved by endocrinologists involving the partner/relative of the patient.

Declaration of interest

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Data sharing statement

Data collected for the study, including individual participant data and a data dictionary defining each field in the set, will be made available upon reasonable request to the corresponding author.

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