

Evaluation of an individualized education program in pituitary diseases: a pilot study

Frédérique Albarel, I Pellegrini, H Rahabi, C Baccou, L Gonin, C Rochette, M Vermalle, Thomas Cuny, Frederic Castinetti, Thierry Brue

▶ **To cite this version:**

Frédérique Albarel, I Pellegrini, H Rahabi, C Baccou, L Gonin, et al.. Evaluation of an individualized education program in pituitary diseases: a pilot study. European Journal of Endocrinology, 2020, 183 (6), pp.551-559. 10.1530/EJE-20-0652 . hal-03223111

HAL Id: hal-03223111
https://amu.hal.science/hal-03223111

Submitted on 11 May 2021

HAL is a multi-disciplinary open access archive for the deposit and dissemination of scientific research documents, whether they are published or not. The documents may come from teaching and research institutions in France or abroad, or from public or private research centers.

L'archive ouverte pluridisciplinaire **HAL**, est destinée au dépôt et à la diffusion de documents scientifiques de niveau recherche, publiés ou non, émanant des établissements d'enseignement et de recherche français ou étrangers, des laboratoires publics ou privés.

Evaluation of an individualized education program in pituitary diseases: a pilot study

Albarel F[1], Pellegrini I[1], Rahabi H[1], Baccou C[1], Gonin L[1], Rochette C[1], Vermalle M[1], Cuny T[1, 2], Castinetti F[1, 2], Brue T[1, 2]

[1]Assistance Publique-Hôpitaux de Marseille (AP-HM), Department of Endocrinology, Hôpital de la Conception, Centre de Référence des Maladies Rares de l'hypophyse HYPO, 13005, Marseille, France and [2]Aix-Marseille Université, Institut National de la Santé et de la Recherche Médicale (INSERM), U1251, Marseille Medical Genetics (MMG), Institut Marseille Maladies Rares (MarMaRa), Marseille, France

Corresponding author: Thierry Brue, MD, PhD

Department of Endocrinology, Conception Hospital, 147 Boulevard Baille, 13005 Marseille, France

Telephone: +33 491 383 479, Fax: +33 491 384 131, e-mail: thierry.brue@univ-amu.fr

Short title: education program in pituitary disease

Keywords: education program, pituitary disease, acromegaly, corticotropin deficiency, Cushing's disease

Word count: 3232 words (3399 with declaration of interest, funding and acknowledgements)

Number of figures and tables: 4

For Review Only

Abstract

The low prevalence of pituitary diseases makes patient autonomy crucial, and self-management programs should be more common.

Objectives: To assess the efficacy of an education program for patients with pituitary diseases in terms of patients' quality of life, satisfaction and goal attainment.

Design and methods: Adult patients with pituitary disorders were recruited in a tertiary referral center and chose at least three of eight possible sessions on various topics, from disease management to psychosocial issues. Patients were included if they attended at least three sessions between 2012 and 2016 and completed the initial, final, and follow-up questionnaires. Data on quality of life (SF36), satisfaction and goal attainment were analyzed.

Results: Fifty-three patients were included (33 women; mean age, 53.5 years). There were significant quality of life improvements in terms of physical and psychic limitation scores at final assessment that persisted at follow-up evaluation. Most patients reached their objectives, especially those on sharing experiences and improving autonomy and self-confidence. More than half set new objectives at the end of the program, the most popular one being to reinforce their knowledge of their pituitary disease, its evolution and treatment (17.1% of patients). The mean overall satisfaction score was 3.75/4. At follow-up evaluation, patients reported improved self-management of pituitary disease (3.6/5) and improved self-efficacy (3.8/5).

Conclusion: Individualizing the educational objectives of patients with pituitary disease improves the way they live with their disease. If confirmed in other cohorts this approach could become the gold standard for education programs in rare endocrine diseases.

For Review Only

Introduction

Educating patients with rare diseases is crucial as this rarity makes management by non-specialized health professionals difficult. The degree of emergency of certain situations may be underestimated and their management suboptimal, which can in some cases put the patient's life at risk (1). It is therefore important for patients to acquire knowledge on their disease and its treatments, and to be able to manage their medical condition autonomously, both on daily life and in emergency situations.

Education programs have been developed for many frequent chronic diseases (such as diabetes, asthma, hypertension, dyslipidemia, obesity) and in oncology, and most have been shown to improve patient outcomes (2). One noteworthy fact is that these programs are only seldom described in detail, which makes it difficult to form a clear idea of the characteristics of the educational modules that are the most effective. Education programs can be delivered in a wide variety of forms: in single or multiple sessions, with or without handouts – with spoken explanations or not – to be read or learnt, individually or in group sessions, as practical workshops with health professionals, using an individualized or standardized program, and so on. They can also differ widely in terms of content and content type: classes based on the transmission of information versus practice, training, and/or psychological interventions. The effectiveness of an education program has been shown to depend on all these features (2–4).

Several education programs have been developed in different countries for rare endocrine diseases, notably for adrenal insufficiency. Indeed, the most recent French and international guidelines on the management of adrenal insufficiency highlight the importance of patient education, in particular for the administration of stress-dose glucocorticoids and emergency hydrocortisone injections (5–7). Most of these programs are based on learning and training and aim to allow patients to manage emergency situations by themselves and avoid potentially life-threatening adrenal crises (1,8–13). Some programs include patients with

For Review Only

secondary adrenal insufficiency as they also need to be educated on how to adapt hydrocortisone doses and perform hydrocortisone subcutaneous injections (14), and because morbidity and mortality are higher in patients both with primary and with secondary adrenal insufficiency (1,12,15). An educational program has also recently been developed for patients with neuroendocrine tumours (NET) (16), based on individual and group learning and discussion sessions focusing on self-efficacy, showing significant improvement in patients' general self-efficacy, physical component scores of HRQoL and stress (16).

Patients with pituitary diseases could be included in programs such as these, but their specific hormonal imbalances – deficiencies or, in some conditions such as acromegaly or Cushing disease, hormone hypersecretion – and their corresponding needs and complaints must be taken into account. It is now well established that patients with pituitary diseases can have poorer quality of life even when their disease is considered in remission (17–19). It is therefore essential to provide these patients with individualized multidimensional education programs. Very few education programs have been developed for patients with pituitary diseases. To our knowledge, the only examples in the literature are a recently published program focusing on psychosocial issues in patients with pituitary disease (20) and an educational nursing intervention program specifically designed for patients with Cushing syndrome (21). Detailed data on pituitary-oriented education programs are currently lacking. We have developed a new type of individualized education program for patients with pituitary diseases and in this study, we evaluated its efficacy in terms of quality of life outcomes, adaptability to patient needs, patient goal attainment and patient satisfaction.

Subjects and methods

Patients

Between 2012 and 2016, 171 patients were included in the pituitary education program and attended an "educational diagnosis". The first 53 patients who finished the education program

For Review Only

in pituitary diseases held at the Conception hospital in Marseille, France, and completed the follow-up evaluation were included. All patients with pituitary diseases followed up in the department had the option of taking part in the "pituitary disease education program" presented to them by their referring endocrinologist. Written informed consent was obtained from all patients who agreed to participate during the first meeting, before any involvement in group sessions. The ethics committee of Aix-Marseille University specifically approved this study.

Program and data collection

The therapeutic education program was developed by a team of health professionals (physicians, nurses, dieticians, psychologists) from the French Reference Centre for Rare Pituitary Diseases in the Department of Endocrinology of the Conception hospital, Marseille, France, in collaboration with five French support groups for patients with pituitary diseases (growth disorders, diabetes insipidus, adrenal diseases, McCune Albright syndrome, craniopharyngioma), listed in the Acknowledgements. The education program was approved by the regional health agency (*Agence Régionale de Santé Provence-Alpes-Côte d'Azur*) at the end of 2012 and was implemented at the beginning of 2013.

The main objective of this self-management education program is to improve the quality of life of patients with pituitary diseases. The program was designed to empower patients and increase self-efficacy in the treatment of their pituitary disease by improving their knowledge of management measures and treatments. It also aims to facilitate interactions with other patients suffering from related pathologies and to improve self-confidence and skills, especially in dealing with stressful and emergency situations.

Participating patients with pituitary diseases had an initial ~1 h appointment with a health care provider for an "**educational diagnosis**". At the end of this meeting, patients set "individualized education objectives" based on their knowledge and perceived strengths and

For Review Only

weaknesses in dealing with their pituitary disease, and were asked to choose at least three out of eight possible sessions, designed to help them achieve these goals.

Eight different types of group sessions were organized regularly. These workshops involved three to eight patients, accompanied if they wished by one or two relatives and lasted about 2.5 h. The instructors were either nurses, psychologists, medical doctors or dieticians, depending on the type of session. Members of the partnering patient support groups with knowledge of the disease and who had received training in education programs and methods also participated as "expert patients" (***Figure 1***).

Details of the eight sessions are reported in ***Appendix 1.***

Once patients had completed the set of sessions they had selected, they had a "**final appointment**"". The patient's progress in the program was reviewed with a focus on whether and to what extent the initial goals had been achieved. Additional abilities or objectives that they would have liked to achieve were also discussed. Further workshops (for reinforcement, consolidation) were proposed in some cases to achieve these new goals.

Follow-up evaluation questionnaires (*SF36* and a *follow-up evaluation questionnaire*, ***Figure 2***) were sent to patients between three and six months after the final appointment.

Statistical analysis

Quantitative variables were expressed as mean ± standard deviation (SD) and were compared between groups using unpaired two-tailed Student's t-tests. Categorical variables were expressed as frequencies and compared using two-tailed Pearson's chi-square tests. Results were considered statistically significant at $P < 0.05$.

Results

Among the 53 patients included in the study (33 women; mean age 53.5 ± 17.0 years), 14 had Cushing disease, 3 had acromegaly, 2 had suprasellar germinoma, 10 had isolated and 1 multiple pituitary deficiency, and 3 had isolated central diabetes insipidus. There were also

eje@bioscientfica.com

For Review Only

cases of pituitary sarcoidosis (n = 1), pituitary apoplexy (n = 4), Sheehan syndrome (n = 3), non-secreting adenoma (n = 5), craniopharyngioma (n = 4) or other parasellar lesions with pituitary deficiencies (n = 3; one case each of meningioma, choristoma and glioma). The mean time between educational diagnosis and the final meeting was 11.0 ± 7.6 months.

Questionnaires

All 53 patients answered the *satisfaction questionnaire*, with a mean satisfaction level of 3.75 ± 0.29 (with a 1 (not satisfied) to 4 (very satisfied) scale). Patients were particularly satisfied by the attentiveness of the educational team (3.91/4) and the lowest average score (3.54/4) related to the tools and material used in the workshops. ***The patients also reported that the program met their expectations (3.85 ± 0.36), the contents of the workshops was useful (3.83 ± 0.38), the sessions improved their knowledge (3.81 +/- 0.44), allowed them to share their experiences (3.68 ± 0.58) and will help them to manage their pituitary disease in daily life (3.66 ± 0.59).***

Fifty-two patients completed the *follow-up evaluation questionnaire* (32 women/20 men), 5.5 ± 3.0 months after the final meeting. The mean scores for the questions on changes to the management of their pituitary disease and on increased self-efficacy were 3.6 ± 0.9 and 3.8 ± 0.7 (out of 5), respectively. There were no reports of acute events requiring hospitalization in the period since completing the course. Patients 'comments highlighted the need for new sessions on pituitary disease-related professional issues and the possibility of additional educational tools.

Thirty-five patients (23 women, 12 men) completed the *SF36* quality of life questionnaire before and after completing the program. The results at the start and end of the program (35 respondents) and at the follow-up evaluation (21 respondents) are listed in ***Table 1.*** Physical limitation scores were significantly higher at the end of the program than at the start ($P < 0.001$), and remained higher at the follow-up evaluation (initial vs follow-up, $P=0.05$).

eje@bioscientfica.com

For Review Only

Patients also reported improved psychic limitation scores (initial vs final, p =0.046) that persisted at follow-up (final vs follow-up, $P = 1$). Mental health scores were significantly lower at the end of the program than on initial evaluation ($P < 0.001$), but increased significantly between the end of the program and the follow-up evaluation ($P = 0.047$).

Patients' objectives

We collected answers from 35 patients concerning their objectives at the start (educational diagnosis) and at the end (final appointment) of the program. The patients' objectives were classified in one of eight general categories and were considered totally, partially, or not achieved, and requiring strengthening or not (***Figure 3).*** The most widely chosen type of objective, by 29/35 patients (82.9%) was interacting and sharing experiences with other patients (first category). Learning how to manage emergency situations (sixth category) and improving their knowledge on their pituitary disease and its treatment and evolution (seventh category) were also priority objectives for more than half of the patients. Objectives classified as sharing experiences (first category) and improving self-confidence and autonomy in their condition (fifth category) were achieved by more than three quarters of patients and those for managing emergency situations (sixth category) were achieved by two thirds of patients. At the final meeting, 18/35 patients (51.4%) chose new objectives to achieve or reinforce. The objectives most commonly chosen for this consolidation phase concerned the patients' knowledge of their condition and treatments (seventh category, 6 patients), how to manage and adapt treatments (fourth category, 4 patients) and dietary management (eighth category, 4 patients).

Discussion

This education program was designed to develop the self-management skills of patients with a pituitary disease. To our knowledge, this is the first program for patients with endocrine diseases whose content can be customized to such an extent, focusing on patients' individual

eje@bioscientfica.com

For Review Only

needs and objectives, and covering a wide variety of subjects, from knowledge of pituitary diseases, hormones and healthcare, to psychosocial and practical aspects of everyday life.

Two other education programs for patients with pituitary diseases have been published in the last few years. Andela *et al*. recently implemented a self-management program in pituitary diseases targeting psychosocial issues and including patients' partners. They report promising results from a randomized controlled trial involving 174 patients in two groups, with improved self-efficacy persisting up to 6 months after the end of the program (20). Patients attended 8 weekly sessions of 90 min in groups of 5–7 participants stratified by disease. At the end of the program, patients stated that they would have liked to receive more medical information about their disease (20). Since patients with pituitary disease have disease specific needs (22,23) and are known to have an impaired quality of life that can be improved but not normalized by surgical and pharmacological interventions (24), we also chose to include workshops on psychosocial issues in our program (sessions 1, 7 and 8, led by a psychologist), in addition to the informal exchanges that took place during each group learning session. The second recently published educational program, developed for patients with Cushing's syndrome, was based on five visits over 9 months (including four 2 h educational sessions), and focused on the disease and its comorbidities, treatment and management, with no workshop devoted to its psychosocial aspects (21). In a randomized controlled study of 61 patients, those in the intervention group had improved physical activity, a more healthy lifestyle, better sleep patterns and lower pain than those in the control group at the end of the program, leading to better health related quality of life and reduced health resources consumption (21). Our patients reported a decrease in physical role limitations at the end of the program that persisted until the follow-up evaluation. Psychic limitations were also significantly reduced at the end of the program, while the mental health score worsened

For Review Only

and then returned to the baseline level, ***maybe because patients had to get used to the change in management of their pituitary disease***.

We did not observe significant changes in terms of physical functioning or pain, general health perceptions, energy/vitality or social functioning. This may be because patients enrolled had a variety of pituitary pathologies and were re-assessed several months after the end of the program, while in Martinez-Momblan *et al*.'s study, the patients all had Cushing's syndrome and were evaluated at the end of the study only, with no follow-up questionnaire (21). Indeed, Andela *et al.* found that the reported reduction in bother from mood problems at the end of the program had disappeared at 6 months' follow-up evaluation (20). Furthermore, since only 35 of the 53 patients in our study completed the SF36 questionnaire both at the beginning and at the end of the program and only 21 at follow-up may explain why some of the differences did not reach the significance threshold due to a lack of statistical power. Finally, mirroring the reduction in health resources consumption observed by Martinez-Momblan *et al*.**,** no patient in our study reported any acute event requiring hospitalization in the few months between the end of the program and the follow-up evaluation.

The personalization and adaptability of the program to individual needs and requirements is also an important factor in promoting patient involvement and adherence, which are essential for learning and for changing behaviours. The time requirements of such programs can lead to low participation and high rates of attrition, so adapting contents to the specific needs and pathologies of participants is crucial to generate and maintain interest (25). In our program, patients initially choose at least three sessions to achieve their individual goals with a wide variety of combinations of sessions. We hypothesized that making this education program customizable would limit the attrition rate, which can be as high as 50% for some programs (25). While we were unable to test this hypothesis because the education sessions were scheduled on demand and there was no time limit for participants to complete their chosen set,

eje@bioscientfica.com

For Review Only

patient satisfaction rates, especially regarding their expectations, the content of the sessions and learning outcomes, indicate that the design of the program and its adaptability to their needs was well received by patients and met their expectations.

In a recent study by Burger-Stritt *et al*. (9), 95% of the patients with adrenal insufficiency participated in an education program where they received general information on the disease and practical training on self-injection and how to adjust daily doses of hydrocortisone. Despite this education program, glucocorticoid injections were only performed by patients or their relatives in 26 of the 59 emergency situations, and at least 15% of patients failed to adjust their oral glucocorticoid dose. The main reasons reported for not performing self-injection were an impaired health condition and a too high inhibition threshold. The authors recommend that patients should be educated repeatedly on dose adjustment and hydrocortisone self-injection (9). Andela et al. (20) also emphasize the importance of additional refreshing sessions in the first year after completing the program or later, and on adjusting the sessions to patients' needs for consolidation, which is necessary in the self-management of chronic diseases in general (25). One of the strengths of our program is the final appointment in which patients reassess their situation in light of their newly acquired knowledge and skills and consider any new needs related to their pituitary disease. Indeed, more than half of our patients chose to set new objectives or to strengthen skills.

The biases and limitations of this study are typical of any retrospective study based on self-reported questionnaire assessments. Further limitations are the small study size, due to the rarity of the pituitary diseases involved, and the length of the program, which lasted 11 months on average and varied between patients. As education is an integrative process, the patient's family and social environment – and the possible changes in these during the program – should also be taken into account to fully understand the results of the program. The low incidence of pituitary diseases meant that we chose not to divide the study population into separate disease-

For Review Only

specific groups (acromegaly, Cushing disease and so on), but on the contrary to make the program inclusive of all pituitary disease subtypes. Patients mostly commented positively on this diversity, reporting that they found it enriched their exchanges.

The originality and richness of this pituitary education program lies in the ability of patients to personalize the set of sessions they attend and to make contents evolve with the evolution of their needs. The program is set to evolve in the future with the inclusion of new topics and learning tools, new types of health professionals, based on recurring remarks. A new workshop on the professional issues caused by pituitary diseases is thus being developed in collaboration with a social worker, as pituitary diseases are known to affect social functioning and health–related absenteeism (26,27). We would like to involve more partners in general to offer the best possible support to pituitary disease patients, particularly in terms of their psychosocial well-being (28). ***We are developing new e-learning tools to complement the already available educational videos on hydrocortisone injections. This will include videoconference sessions allowing disabled or mobility-impaired patients to take part in virtual workshops and education sessions and development of a smartphone application specific for pituitary disorders providing with customized information and allowing follow-up of some clinical, treatment-related or biological parameters. These new tools will aim at optimizing self-management, autonomy, compliance and distant follow-up of pituitary patients.*** Lastly, we are developing a specific questionnaire on the management of hydrocortisone and desmopressin treatments to more objectively evaluate how this changes in our patients after completing the course.

This first evaluation of our individualized self-management education program for patients with pituitary diseases revealed promising results in terms of patient satisfaction, quality of life, skills progression, self-confidence, knowledge of their pituitary disease, its treatment and management. A wider roll-out is now required to evaluate its effect in a larger number of patients.

eje@bioscientfica.com

For Review Only

To conclude, this study supports our view that alongside surgical and pharmacological treatments, education programs such as the one considered here are essential for the well-being of patients with pituitary diseases. The development and dissemination of such programs will have a positive impact on public health, as educated patients manage their symptoms more effectively, and thereby also potentially reduce overall healthcare costs (21,29,30).

Declaration of interest

The authors declare that there is no conflict of interest that could be perceived as prejudicing the impartiality of this study.

Funding

This research did not receive any specific grant from any funding agency in the public, commercial or not-for-profit sector.

Acknowledgements

The authors thank all the patients who took part in the education program, in particular those who collaborated as expert patients. They are grateful for the input and support received from the following patients' associations, especially in the development of the program and for their active participation in the workshops: "GRANDIR" (the French association for patients with growth disorders); "AFDI" (the French association for patients with diabetes insipidus); "Association surrénales" (the French adrenal association); "ASSYMCAL" (the French McCune Albright syndrome association) and "craniopharyngiome-solidarité" (the French association for patients with craniopharyngioma). The authors also thank the members of the AP-HM's healthcare quality department for their help in designing this program and the endocrinologists who have referred their patients to the pituitary education program.

For Review Only

References

1. Hahner S, Spinnler C, Fassnacht M, Burger-Stritt S, Lang K, Milovanovic D, Beuschlein F, Willenberg HS, Quinkler M, & Allolio B. High incidence of adrenal crisis in educated patients with chronic adrenal insufficiency: a prospective study. *The Journal of Clinical Endocrinology and Metabolism* 2015 **100** 407–416. (doi:10.1210/jc.2014-3191)

2. Lagger G, Pataky Z, & Golay A. Efficacy of therapeutic patient education in chronic diseases and obesity. *Patient Education and Counseling* 2010 **79** 283–286. (doi:10.1016/j.pec.2010.03.015)

3. Osborn RL, Demoncada AC, & Feuerstein M. Psychosocial interventions for depression, anxiety, and quality of life in cancer survivors: meta-analyses. *International Journal of Psychiatry in Medicine* 2006 **36** 13–34. (doi:10.2190/EUFN-RV1K-Y3TR-FK0L)

4. Warsi A, Wang PS, LaValley MP, Avorn J, & Solomon DH. Self-management education programs in chronic disease: a systematic review and methodological critique of the literature. *Archives of Internal Medicine* 2004 **164** 1641–1649. (doi:10.1001/archinte.164.15.1641)

5. Bornstein SR, Allolio B, Arlt W, Barthel A, Don-Wauchope A, Hammer GD, Husebye ES, Merke DP, Murad MH, Stratakis CA, & Torpy DJ. Diagnosis and Treatment of Primary Adrenal Insufficiency: An Endocrine Society Clinical Practice Guideline. *The Journal of Clinical Endocrinology and Metabolism* 2016 **101** 364–389. (doi:10.1210/jc.2015-1710)

6. Husebye ES, Allolio B, Arlt W, Badenhoop K, Bensing S, Betterle C, Falorni A, Gan EH, Hulting AL, Kasperlik-Zaluska A *et al.* Consensus statement on the diagnosis, treatment and follow-up of patients with primary adrenal insufficiency. *Journal of Internal Medicine* 2014 **275** 104–115. (doi:10.1111/joim.12162)

7. Guignat L. Therapeutic patient education in adrenal insufficiency. *Annales D'endocrinologie* 2018 **79** 167–173. (doi:10.1016/j.ando.2018.03.002)

For Review Only

8. Burman P, Mattsson AF, Johannsson G, Höybye C, Holmer H, Dahlqvist P, Berinder K, Engström BE, Ekman B, Erfurth EM, Svensson J *et al.* Deaths among adult patients with hypopituitarism: hypocortisolism during acute stress, and de novo malignant brain tumors contribute to an increased mortality. *The Journal of Clinical Endocrinology and Metabolism* 2013 **98** 1466–1475. (doi:10.1210/jc.2012-4059)

9. Burger-Stritt S, Kardonski P, Pulzer A, Meyer G, Quinkler M, & Hahner S. Management of adrenal emergencies in educated patients with adrenal insufficiency-A prospective study. *Clinical Endocrinology* 2018 **89** 22–29. (doi:10.1111/cen.13608)

10. Schad R, Schneider PJ, & Nold EG. Reimbursable pharmacy teaching program for adrenalectomy patients. *American Journal of Hospital Pharmacy* 1979 **36** 1212–1214.

11. Guignat L. [Therapeutic education in adrenal insufficiency: A tool insufficiently used to prevent or treat early acute adrenal insufficiency]. *Presse Medicale (Paris, France: 1983)* 2014 **43** 444–452. (doi:10.1016/j.lpm.2014.01.008)

12. Repping-Wuts HJWJ, Stikkelbroeck NMML, Noordzij A, Kerstens M, & Hermus ARMM. A glucocorticoid education group meeting: an effective strategy for improving self-management to prevent adrenal crisis. *European Journal of Endocrinology* 2013 **169** 17–22. (doi:10.1530/EJE-12-1094)

13. Vidmar AP, Weber JF, Monzavi R, Koppin CM, & Kim MS. Improved medical-alert ID ownership and utilization in youth with congenital adrenal hyperplasia following a parent educational intervention. *Journal of pediatric endocrinology & metabolism* 2018 **31** 213–219. (doi:10.1515/jpem-2017-0435)

14. Hahner S, Burger-Stritt S & Allolio B. Subcutaneous hydrocortisone administration for emergency use in adrenal insufficiency. *European Journal of Endocrinology* 2013 **169** 147-54. (doi: 10.1530/EJE-12-1057)

For Review Only

15. Bancos I, Hahner S, Tomlinson J, & Arlt W. Diagnosis and management of adrenal insufficiency. *The Lancet. Diabetes & Endocrinology* 2015 **3** 216–226. (doi:10.1016/S2213-8587(14)70142-1)

16. Haugland T, Veenstra M, Vatn MH, & Wahl AK. Improvement in Stress, General Self-Efficacy, and Health Related Quality of Life following Patient Education for Patients with Neuroendocrine Tumors: A Pilot Study. *Nursing Research and Practice* 2013 695820. (doi:10.1155/2013/695820)

17. De Bucy C, Guignat L, Niati T, Bertherat J, & Coste J. Health-related quality of life of patients with hypothalamic-pituitary-adrenal axis dysregulations: a cohort study. *European Journal of Endocrinology* 2017 **177** 1–8. (doi:10.1530/EJE-17-0048)

18. Kyriakakis N, Lynch J, Gilbey SG, Webb SM, & Murray RD. Impaired quality of life in patients with treated acromegaly despite long-term biochemically stable disease: Results from a 5-years prospective study. *Clinical Endocrinology* 2017 **86** 806–815. (doi:10.1111/cen.13331)

19. Klaauw AA van der, Kars M, Biermasz NR, Roelfsema F, Dekkers OM, Corssmit EP, Aken MO van, Havekes B, Pereira AM, Pijl H *et al.* Disease-specific impairments in quality of life during long-term follow-up of patients with different pituitary adenomas. *Clinical Endocrinology* 2008 **69** 775–784. (doi:10.1111/j.1365-2265.2008.03288.x)

20. Andela CD, Repping-Wuts H, Stikkelbroeck NMML, Pronk MC, Tiemensma J, Hermus AR, Kaptein AA, Pereira AM, Kamminga NGA, & Biermasz NR. Enhanced self-efficacy after a self-management programme in pituitary disease: a randomized controlled trial. *European Journal of Endocrinology* 2017 **177** 59–72. (doi:10.1530/EJE-16-1015)

21. Martínez-Momblán MA, Gómez C, Santos A, Porta N, Esteve J, Úbeda I, Halperin I, Campillo B, Guillaumet M, Webb SM *et al.* A specific nursing educational program in patients with Cushing's syndrome. *Endocrine* 2016 **53** 199–209. (doi:10.1007/s12020-015-0737-0)

For Review Only

22. Andela CD, Niemeijer ND, Scharloo M, Tiemensma J, Kanagasabapathy S, Pereira AM, Kamminga NGA, Kaptein AA, & Biermasz NR. Towards a better quality of life (QoL) for patients with pituitary diseases: results from a focus group study exploring QoL. *Pituitary* 2015 **18** 86–100. (doi:10.1007/s11102-014-0561-1)

23. Andela CD, Lobatto DJ, Pereira AM, Furth WR van, & Biermasz NR. How non-functioning pituitary adenomas can affect health-related quality of life: a conceptual model and literature review. *Pituitary* 2018 **21** 208–216. (doi:10.1007/s11102-017-0860-4)

24. Andela CD, Scharloo M, Pereira AM, Kaptein AA, & Biermasz NR. Quality of life (QoL) impairments in patients with a pituitary adenoma: a systematic review of QoL studies. *Pituitary* 2015 **18** 752–776. (doi:10.1007/s11102-015-0636-7)

25. Newman S, Steed L, & Mulligan K. Self-management interventions for chronic illness. *Lancet (London, England)* 2004 **364** 1523–1537. (doi:10.1016/S0140-6736(04)17277-2)

26. Lobatto DJ, Steffens ANV, Zamanipoor Najafabadi AH, Andela CD, Pereira AM, Hout WB van den, Peul WC, Vliet Vlieland TPM, Biermasz NR, & Furth WR van. Work disability and its determinants in patients with pituitary tumor-related disease. *Pituitary* 2018 **21** 593–604. (doi:10.1007/s11102-018-0913-3)

27. Hahner S, Loeffler M, Fassnacht M, Weismann D, Koschker AC, Quinkler M, Decker O, Arlt W, & Allolio B. Impaired subjective health status in 256 patients with adrenal insufficiency on standard therapy based on cross-sectional analysis. *The Journal of Clinical Endocrinology and Metabolism* 2007 **92** 3912–3922. (doi:10.1210/jc.2007-0685)

28. Andela CD, Tiemensma J, Kaptein AA, Scharloo M, Pereira AM, Kamminga NG, & Biermasz NR. The partner's perspective of the impact of pituitary disease: Looking beyond the patient. *Journal of Health Psychology* 2019 **24** 1687–1697.

For Review Only

29. Dibley L, Norton C, & Jones R. Informing a patient education programme for GORD: A literature review. *Gastrointestinal Nursing* 2009 **7** 28–37. (doi:10.12968/gasn.2009.7.5.42907)

30. Lobatto DJ, Hout WB van den, Zamanipoor Najafabadi AH, Steffens ANV, Andela CD, Pereira AM, Peul WC, Furth WR van, Biermasz NR, & Vliet Vlieland TPM. Healthcare utilization and costs among patients with non-functioning pituitary adenomas. *Endocrine* 2019 **64** 330–340. (doi:10.1007/s12020-019-01847-7)

For Review Only

Legends to Tables and Figures

Figure 1: Design of an individualized education program in pituitary diseases

Flow diagram of the individualized education program for patients with pituitary diseases. Patients with pituitary diseases have an initial "**educational diagnosis**" appointment where they choose at least three of the eight proposed sessions (described on the right and left of the central arrow with the instructors involved in italics), based on their chosen goals. After attending the chosen sessions, patients have a final appointment and can choose to follow other workshops to achieve new objectives. The questionnaires given to the patients at the different appointments are named in italics.

Figure 2: Follow-up evaluation questionnaire.

Fifty-two patients filled in this questionnaire, 5.5 ± 3.0 months (mean ± standard deviation) after the final evaluation.

Figure 3: Patients' objectives.

The proportions of patients who reported completely, partially and not reaching their objectives are shown in black, light grey and grey, respectively, for each objective (numbered 1–8) with the corresponding percentages shown above each group of bars.

The proportions of patients who initially chose each objective are shown below the plot (objectives listed on the right side from 1 to 8)

Table 1: Evolution in SF36 quality of life dimensions for patients attending the education program on pituitary diseases.

Results are presented as mean ± standard deviation.

Higher scores indicate better quality of life.

eje@bioscientfica.com

For Review Only

441 Significant *P*-values are shown in bold

For Review Only

Manuscript submitted for review to Journal of Endocrinology

Patients with pituitary diseases

Educational diagnosis

Individualized objectives
Initial evaluation *(consent, SF36)*

CHOOSE at least 3 sessions

1- Daily life with a pituitary disease
Psychologist, nurse

2- How to manage metabolic and bone complications of pituitary diseases
Doctor, dietetician

3- Corticotropin deficiency and diabetes insipidus in "real life"
Doctor, nurse

4- Weight increase and obesity, how to manage food and a satiety problem
Dietetician, nurse

5- How to manage your pituitary treatment, knowledge and practice
Doctor, nurse

6- Corticotropin deficiency: treatment and emergency situations
Doctor, nurse

7- Sexuality and fertility with a pituitary disease
Psychologist, doctor

8- Long-term treatments complications of pituitary diseases
Psychologist, doctor

Final meeting

Have objectives been achieved? New objectives?
Wish for consolidation?
Final evaluation *(satisfaction questionnaire, SF36)*

Further sessions if desired
Follow-up evaluation (follow-up *questionnaire, SF36*)

eje@bioscientfica.com

Follow-up evaluation questionnaire

Name: **Date:**

1- Having completed this program, would you say the management of your pituitary disease has been modified:

Radically A lot A little Very little Not at all

2- Having completed this program, would you say that your self-assurance in managing your pituitary disease has increased (regarding your knowledge, your treatment(s))

Radically A lot A little Very little Not at all

3- Can you list each of the treatments for your pituitary condition and their roles?

Treatment	Role

4- Describe your pituitary disease and its complications

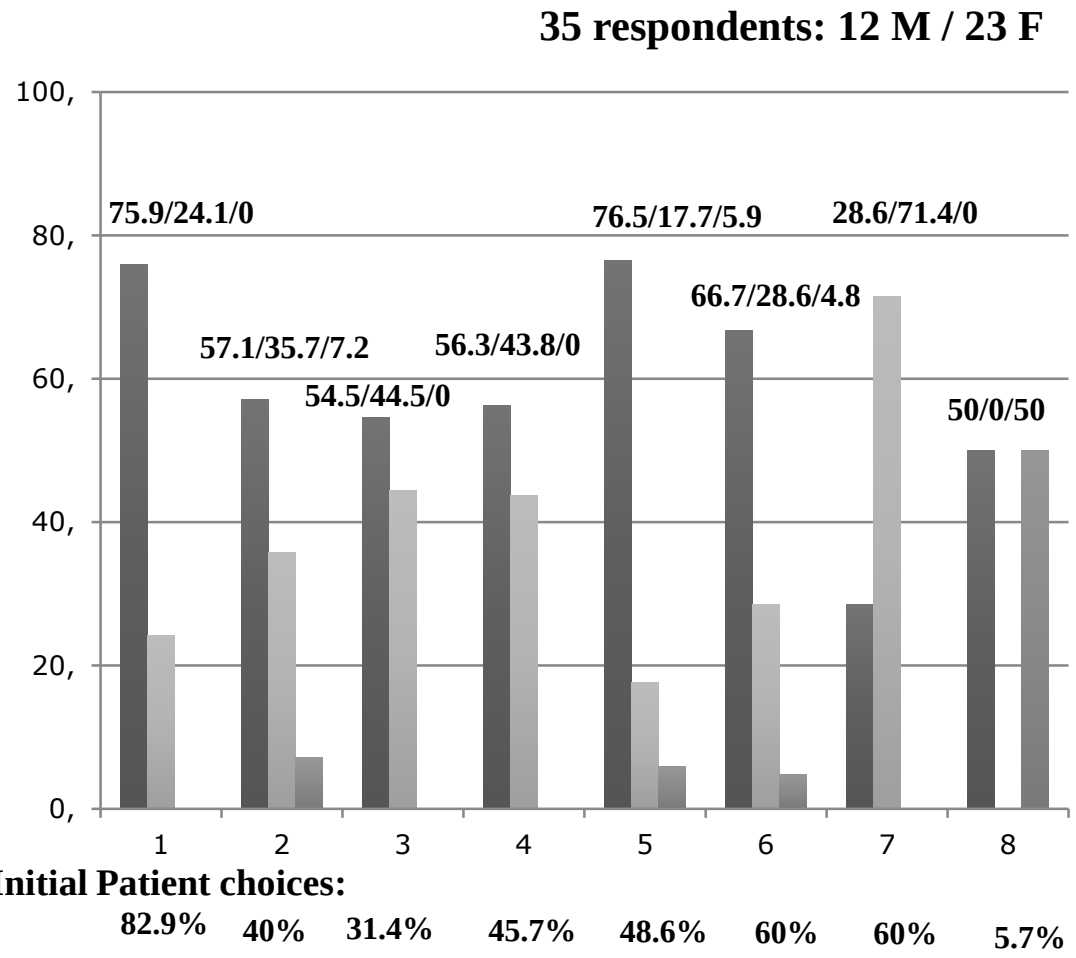
……
……

5- Have you had any problems related to your pituitary disease since completing the program that required hospitalization? If you have, please provide details.

……
……

Comments and suggestions:

For Review Only



- Totally achieve
- Partially achieved
- Not achieved

Patients' objectives

1 Share experiences, daily life and feelings of isolation

2 Share everyday experiences

3 Share long-term experiences

4 Learn how to manage and adapt their treatment

5 Improve their autonomy and self-confidence in their pituitary disease

6 Learn how to manage emergency situations

7 Improve knowledge on their pituitary disease, its evolution and hormone treatments

eje@bioscientifica.com

8 Learn to better manage food and dietary issues

	Initial evaluation score	Final evaluation score	Follow-up evaluation score	P (initial vs final)	P (final vs follow-up)	P (initial vs follow-up)
Physical functioning	76.1 +/- 15.1	75.9 +/- 15.4	74. 6 +/- 11.1	0.94	0.40	0.62
Physical limitations	31.8 +/- 6.8	62.1 +/- 14.3	42.7 +/-18.5	<0.001	<0.001	0,005
Physical pain	75,0 +/- 13.7	75.2 +/- 12.3	73.4 +/- 13.9	0.92	0.39	0.06
General health perceptions	61.9 +/- 12.9	64.5 +/- 14.8	62.7 +/- 14.9	0.16	0.47	0.71
Energy/Vitality	61.3 +/- 13.8	64.6 +/- 14.7	59.5 +/- 14.3	0.09	0.1	0.52
Social functioning	73.1 +/- 17.5	75.7 +/- 18.8	75.7 +/-17.8	0.88	1	0.25
Mental health	73.6 +/- 13.0	67.0 +/- 11.1	71.9 +/- 11.8	<0.001	0.047	0.42
Psychic limitations	70.5 +/- 21.8	76.6 +/- 21.1	76.6 +/- 20.7	0.046	1	0.06
Physical component summary	45.3 +/- 9.1	53.2 +/- 12.4	45.3 +/- 7.1	0.58	0.52	0.69
Mental component summary	53.3 +/- 12.7	52.5 +/- 13.9	52.6 +/- 12.3	0.95	0.60	0.66

eje@bioscientfica.com

Appendix 1: details of the eight sessions offered to participants in the education program on pituitary diseases

The first session is on "**Daily life with a pituitary disease**". This is a psychosocial workshop, led by a psychologist and a nurse, based on photos that lead to discussions and a sharing of experiences. The workshop focuses first on the difficulties patients face in daily life and then on the solutions found by each participant to improve their everyday life.

The second session is entitled "**How to manage the metabolic and bone complications of pituitary diseases**". Workshops are led by a doctor and dietician using the metaplan technique and are based on patients' knowledge with guidance from the instructors.

The third session, specifically for patients with corticotropin deficiency or diabetes insipidus, is entitled "**Corticotropin deficiency or diabetes insipidus in real life**". In these workshops, led by a doctor and a nurse, patients talk with one another, under the doctor's supervision, about everyday situations in which their treatment (hydrocortisone or desmopressin) should or should not be adapted. They chose cards representing different everyday activities and learn when and how to adapt their treatments and when to perform hydrocortisone injections. At the end of the session, they practice and self-evaluate their ability to perform subcutaneous hydrocortisone injections with the nurse. A video of one of the nurses in the department demonstrating how to perform a subcutaneous hydrocortisone injection is available on the reference centre's website and patients are provided with an illustrated guide.

The fourth session is a dietary workshop entitled "**Weight gain and obesity; how to manage food and satiety problems**". This class is led by a dietician and a nurse; it focuses on how to maintain a balanced diet with a pituitary disease, physical activity, nutritionally balanced meals, tasting, enjoying food, shopping at the supermarket, and so on, and involves patients preparing balanced meals with play food.

The fifth class, "**How to manage your pituitary treatment, in theory and in practice**" focusses on pituitary disease and hormone knowledge. The instructor, a medical doctor explains the physiology of the pituitary gland and of pituitary hormones based on participants' knowledge and questions. Then patients give their medicine package, and are asked to explain at what level their treatments act and how. Finally, they share their experiences and understanding of their treatments and their direct and side effects.

For Review Only

The topic of the sixth session, "**Corticotropin deficiency: treatment and emergency situations**", is focused on the emergency management of adrenal crises. Patients choose pictures representing acute events or everyday situations that could justify an increase in their hydrocortisone treatment or a subcutaneous hydrocortisone injection. A role-playing game is then led by the instructors (a medical doctor and a nurse) involving these situations and the patients decide what the best way to manage their hydrocortisone treatment is in these events of stress, pain, illness etc. The decisions are then discussed and evaluated constructively in the group. In the final part of the workshop, patients learn how to explain their corticotropin deficiency and their hydrocortisone treatment to emergency department doctors, and then practice subcutaneous hydrocortisone injections with the nurse, and self-evaluate their ability to perform these injections.

The seventh session is a discussion group on "**Sexuality and fertility with a pituitary disease**" led by a doctor and a psychologist using the metaplan method and patients' knowledge and questions.

The eighth session is on "**Long-term treatment complications of pituitary diseases**" and deals with the long term evolution of pituitary pathologies and how patients imagine their future with their pathology. Patients first describe and share experiences about their pathology and the treatments they have received (surgery, radiotherapy, medical) and then describe the future they envisage with their pituitary disease, based on photos of different pathways.

For Review Only