



**HAL**  
open science

## 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.

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

3 **Albarel F<sup>1</sup>, Pellegrini I<sup>1</sup>, Rahabi H<sup>1</sup>, Baccou C<sup>1</sup>, Gonin L<sup>1</sup>, Rochette C<sup>1</sup>, Vermalle M<sup>1</sup>,**  
4 **Cuny T<sup>1,2</sup>, Castinetti F<sup>1,2</sup>, Brue T<sup>1,2</sup>**

5 *<sup>1</sup>Assistance Publique-Hôpitaux de Marseille (AP-HM), Department of Endocrinology, Hôpital*  
6 *de la Conception, Centre de Référence des Maladies Rares de l'hypophyse HYPO, 13005,*  
7 *Marseille, France and <sup>2</sup>Aix-Marseille Université, Institut National de la Santé et de la*  
8 *Recherche Médicale (INSERM), U1251, Marseille Medical Genetics (MMG), Institut Marseille*  
9 *Maladies Rares (MarMaRa), Marseille, France*

10 **Corresponding author:** Thierry Brue, MD, PhD

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

13 Telephone: +33 491 383 479, Fax: +33 491 384 131, e-mail: [thierry.brue@univ-amu.fr](mailto:thierry.brue@univ-amu.fr)

14 **Short title:** education program in pituitary disease

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

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

18 **Number of figures and tables:** 4

19 **Abstract**

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

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

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

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

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

## 40 **Introduction**

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

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

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

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

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

## 86 **Subjects and methods**

### 87 **Patients**

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

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

## 96 **Program and data collection**

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

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

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

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

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

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

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

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

### 130 **Statistical analysis**

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

## 135 **Results**

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

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

### 143 Questionnaires

144 All 53 patients answered the *satisfaction questionnaire*, with a mean satisfaction level of  $3.75$   
145  $\pm 0.29$  (with a 1 (not satisfied) to 4 (very satisfied) scale). Patients were particularly satisfied  
146 by the attentiveness of the educational team ( $3.91/4$ ) and the lowest average score ( $3.54/4$ )  
147 related to the tools and material used in the workshops. *The patients also reported that the*  
148 *program met their expectations ( $3.85 \pm 0.36$ ), the contents of the workshops was useful ( $3.83$*   
149  *$\pm 0.38$ ), the sessions improved their knowledge ( $3.81 \pm 0.44$ ), allowed them to share their*  
150 *experiences ( $3.68 \pm 0.58$ ) and will help them to manage their pituitary disease in daily life*  
151 *( $3.66 \pm 0.59$ ).*

152 Fifty-two patients completed the *follow-up evaluation questionnaire* (32 women/20 men),  $5.5$   
153  $\pm 3.0$  months after the final meeting. The mean scores for the questions on changes to the  
154 management of their pituitary disease and on increased self-efficacy were  $3.6 \pm 0.9$  and  $3.8 \pm$   
155  $0.7$  (out of 5), respectively. There were no reports of acute events requiring hospitalization in  
156 the period since completing the course. Patients' comments highlighted the need for new  
157 sessions on pituitary disease-related professional issues and the possibility of additional  
158 educational tools.

159 Thirty-five patients (23 women, 12 men) completed the *SF36* quality of life  
160 questionnaire before and after completing the program. The results at the start and end of the  
161 program (35 respondents) and at the follow-up evaluation (21 respondents) are listed in **Table**  
162 **1**. Physical limitation scores were significantly higher at the end of the program than at the start  
163 ( $P < 0.001$ ), and remained higher at the follow-up evaluation (initial vs follow-up,  $P=0.05$ ).



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

### 168 **Patients' objectives**

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

### 184 **Discussion**

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

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

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

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

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

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

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

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

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

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

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

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

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

293

#### 294 **Declaration of interest**

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

#### 297 **Funding**

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

#### 300 **Acknowledgements**

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

## 311 References

- 312 1. Hahner S, Spinnler C, Fassnacht M, Burger-Stritt S, Lang K, Milovanovic D,  
313 Beuschlein F, Willenberg HS, Quinkler M, & Allolio B. High incidence of adrenal crisis in  
314 educated patients with chronic adrenal insufficiency: a prospective study. *The Journal of*  
315 *Clinical Endocrinology and Metabolism* 2015 **100** 407–416. (doi:10.1210/jc.2014-3191)
- 316 2. Lagger G, Pataký Z, & Golay A. Efficacy of therapeutic patient education in chronic  
317 diseases and obesity. *Patient Education and Counseling* 2010 **79** 283–286.  
318 (doi:10.1016/j.pec.2010.03.015)
- 319 3. Osborn RL, Demoncada AC, & Feuerstein M. Psychosocial interventions for  
320 depression, anxiety, and quality of life in cancer survivors: meta-analyses. *International*  
321 *Journal of Psychiatry in Medicine* 2006 **36** 13–34. (doi:10.2190/EUFN-RV1K-Y3TR-FK0L)
- 322 4. Warsi A, Wang PS, LaValley MP, Avorn J, & Solomon DH. Self-management  
323 education programs in chronic disease: a systematic review and methodological critique of the  
324 literature. *Archives of Internal Medicine* 2004 **164** 1641–1649.  
325 (doi:10.1001/archinte.164.15.1641)
- 326 5. Bornstein SR, Allolio B, Arlt W, Barthel A, Don-Wauchope A, Hammer GD, Husebye  
327 ES, Merke DP, Murad MH, Stratakis CA, & Torpy DJ. Diagnosis and Treatment of Primary  
328 Adrenal Insufficiency: An Endocrine Society Clinical Practice Guideline. *The Journal of*  
329 *Clinical Endocrinology and Metabolism* 2016 **101** 364–389. (doi:10.1210/jc.2015-1710)
- 330 6. Husebye ES, Allolio B, Arlt W, Badenhoop K, Bensing S, Betterle C, Falorni A, Gan  
331 EH, Hulting AL, Kasperlik-Zaluska A *et al.* Consensus statement on the diagnosis, treatment  
332 and follow-up of patients with primary adrenal insufficiency. *Journal of Internal Medicine* 2014  
333 **275** 104–115. (doi:10.1111/joim.12162)
- 334 7. Guignat L. Therapeutic patient education in adrenal insufficiency. *Annales*  
335 *D'endocrinologie* 2018 **79** 167–173. (doi:10.1016/j.ando.2018.03.002)

- 336 8. Burman P, Mattsson AF, Johannsson G, Höybye C, Holmer H, Dahlqvist P, Berinder  
337 K, Engström BE, Ekman B, Erfurth EM, Svensson J *et al.* Deaths among adult patients with  
338 hypopituitarism: hypocortisolism during acute stress, and de novo malignant brain tumors  
339 contribute to an increased mortality. *The Journal of Clinical Endocrinology and Metabolism*  
340 2013 **98** 1466–1475. (doi:10.1210/jc.2012-4059)
- 341 9. Burger-Stritt S, Kardonski P, Pulzer A, Meyer G, Quinkler M, & Hahner S.  
342 Management of adrenal emergencies in educated patients with adrenal insufficiency-A  
343 prospective study. *Clinical Endocrinology* 2018 **89** 22–29. (doi:10.1111/cen.13608)
- 344 10. Schad R, Schneider PJ, & Nold EG. Reimbursable pharmacy teaching program for  
345 adrenalectomy patients. *American Journal of Hospital Pharmacy* 1979 **36** 1212–1214.
- 346 11. Guignat L. [Therapeutic education in adrenal insufficiency: A tool insufficiently used  
347 to prevent or treat early acute adrenal insufficiency]. *Presse Medicale (Paris, France: 1983)*  
348 2014 **43** 444–452. (doi:10.1016/j.lpm.2014.01.008)
- 349 12. Repping-Wuts HJWJ, Stikkelbroeck NMML, Noordzij A, Kerstens M, & Hermus  
350 ARMM. A glucocorticoid education group meeting: an effective strategy for improving self-  
351 management to prevent adrenal crisis. *European Journal of Endocrinology* 2013 **169** 17–22.  
352 (doi:10.1530/EJE-12-1094)
- 353 13. Vidmar AP, Weber JF, Monzavi R, Koppin CM, & Kim MS. Improved medical-alert  
354 ID ownership and utilization in youth with congenital adrenal hyperplasia following a parent  
355 educational intervention. *Journal of pediatric endocrinology & metabolism* 2018 **31** 213–219.  
356 (doi:10.1515/jpem-2017-0435)
- 357 14. Hahner S, Burger-Stritt S & Allolio B. Subcutaneous hydrocortisone administration for  
358 emergency use in adrenal insufficiency. *European Journal of Endocrinology* 2013 **169** 147-54.  
359 (doi: 10.1530/EJE-12-1057)



- 360 15. Bancos I, Hahner S, Tomlinson J, & Arlt W. Diagnosis and management of adrenal  
361 insufficiency. *The Lancet. Diabetes & Endocrinology* 2015 **3** 216–226. (doi:10.1016/S2213-  
362 8587(14)70142-1)
- 363 16. Haugland T, Veenstra M, Vatn MH, & Wahl AK. Improvement in Stress, General Self-  
364 Efficacy, and Health Related Quality of Life following Patient Education for Patients with  
365 Neuroendocrine Tumors: A Pilot Study. *Nursing Research and Practice* 2013 695820.  
366 (doi:10.1155/2013/695820)
- 367 17. De Bucy C, Guignat L, Niati T, Bertherat J, & Coste J. Health-related quality of life of  
368 patients with hypothalamic-pituitary-adrenal axis dysregulations: a cohort study. *European*  
369 *Journal of Endocrinology* 2017 **177** 1–8. (doi:10.1530/EJE-17-0048)
- 370 18. Kyriakakis N, Lynch J, Gilbey SG, Webb SM, & Murray RD. Impaired quality of life  
371 in patients with treated acromegaly despite long-term biochemically stable disease: Results  
372 from a 5-years prospective study. *Clinical Endocrinology* 2017 **86** 806–815.  
373 (doi:10.1111/cen.13331)
- 374 19. Klaauw AA van der, Kars M, Biermasz NR, Roelfsema F, Dekkers OM, Corssmit EP,  
375 Aken MO van, Havekes B, Pereira AM, Pijl H *et al.* Disease-specific impairments in quality of  
376 life during long-term follow-up of patients with different pituitary adenomas. *Clinical*  
377 *Endocrinology* 2008 **69** 775–784. (doi:10.1111/j.1365-2265.2008.03288.x)
- 378 20. Andela CD, Repping-Wuts H, Stikkelbroeck NMML, Pronk MC, Tiemensma J, Hermus  
379 AR, Kaptein AA, Pereira AM, Kamminga NGA, & Biermasz NR. Enhanced self-efficacy after  
380 a self-management programme in pituitary disease: a randomized controlled trial. *European*  
381 *Journal of Endocrinology* 2017 **177** 59–72. (doi:10.1530/EJE-16-1015)
- 382 21. Martínez-Momblán MA, Gómez C, Santos A, Porta N, Esteve J, Úbeda I, Halperin I,  
383 Campillo B, Guillaumet M, Webb SM *et al.* A specific nursing educational program in patients  
384 with Cushing's syndrome. *Endocrine* 2016 **53** 199–209. (doi:10.1007/s12020-015-0737-0)

- 385 22. Andela CD, Niemeijer ND, Scharloo M, Tiemensma J, Kanagasabapathy S, Pereira AM,  
386 Kamminga NGA, Kaptein AA, & Biermasz NR. Towards a better quality of life (QoL) for  
387 patients with pituitary diseases: results from a focus group study exploring QoL. *Pituitary* 2015  
388 **18** 86–100. (doi:10.1007/s11102-014-0561-1)
- 389 23. Andela CD, Lobatto DJ, Pereira AM, Furth WR van, & Biermasz NR. How non-  
390 functioning pituitary adenomas can affect health-related quality of life: a conceptual model and  
391 literature review. *Pituitary* 2018 **21** 208–216. (doi:10.1007/s11102-017-0860-4)
- 392 24. Andela CD, Scharloo M, Pereira AM, Kaptein AA, & Biermasz NR. Quality of life  
393 (QoL) impairments in patients with a pituitary adenoma: a systematic review of QoL studies.  
394 *Pituitary* 2015 **18** 752–776. (doi:10.1007/s11102-015-0636-7)
- 395 25. Newman S, Steed L, & Mulligan K. Self-management interventions for chronic illness.  
396 *Lancet (London, England)* 2004 **364** 1523–1537. (doi:10.1016/S0140-6736(04)17277-2)
- 397 26. Lobatto DJ, Steffens ANV, Zamanipoor Najafabadi AH, Andela CD, Pereira AM, Hout  
398 WB van den, Peul WC, Vliet Vlieland TPM, Biermasz NR, & Furth WR van. Work disability  
399 and its determinants in patients with pituitary tumor-related disease. *Pituitary* 2018 **21** 593–  
400 604. (doi:10.1007/s11102-018-0913-3)
- 401 27. Hahner S, Loeffler M, Fassnacht M, Weismann D, Koschker AC, Quinkler M, Decker  
402 O, Arlt W, & Allolio B. Impaired subjective health status in 256 patients with adrenal  
403 insufficiency on standard therapy based on cross-sectional analysis. *The Journal of Clinical*  
404 *Endocrinology and Metabolism* 2007 **92** 3912–3922. (doi:10.1210/jc.2007-0685)
- 405 28. Andela CD, Tiemensma J, Kaptein AA, Scharloo M, Pereira AM, Kamminga NG, &  
406 Biermasz NR. The partner's perspective of the impact of pituitary disease: Looking beyond the  
407 patient. *Journal of Health Psychology* 2019 **24** 1687–1697.

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

411 30. Lobatto DJ, Hout WB van den, Zamanipoor Najafabadi AH, Steffens ANV, Andela CD,  
412 Pereira AM, Peul WC, Furth WR van, Biermasz NR, & Vliet Vlieland TPM. Healthcare  
413 utilization and costs among patients with non-functioning pituitary adenomas. *Endocrine* 2019  
414 64 330–340. (doi:10.1007/s12020-019-01847-7)

415

For Review Only

## 416 **Legends to Tables and Figures**

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

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

425

### 426 **Figure 2:** Follow-up evaluation questionnaire.

427 Fifty-two patients filled in this questionnaire,  $5.5 \pm 3.0$  months (mean  $\pm$  standard deviation)  
428 after the final evaluation.

429

### 430 **Figure 3: Patients’ objectives.**

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

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

436

### 437 **Table 1: Evolution in SF36 quality of life dimensions for patients attending the education 438 program on pituitary diseases.**

439 Results are presented as mean  $\pm$  standard deviation.

440 Higher scores indicate better quality of life.

441 Significant *P*-values are shown in bold

For Review Only

# Patients with pituitary diseases

## Educational diagnosis

Individualized objectives

Initial evaluation (*consent, SF36*)

**1- Daily life with a pituitary disease**

*Psychologist, nurse*

**CHOOSE  
at least 3 sessions**

**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)

## 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**

.....

.....

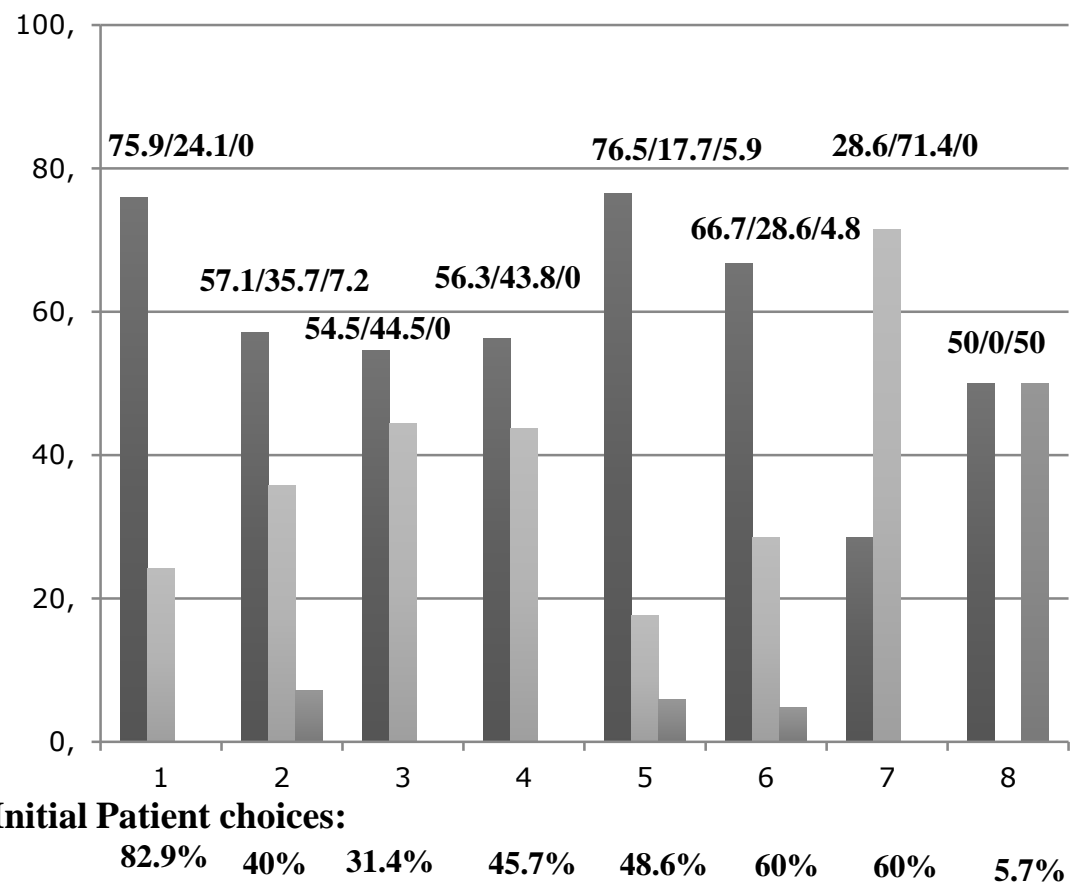
**5- Have you had any problems related to your pituitary disease since completing the program that required hospitalization? If you have, please provide details.**

.....

.....

**Comments and suggestions:**

**35 respondents: 12 M / 23 F**



- Totally achieved
- 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**
- 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)
<b>Physical functioning</b>	76.1 +/- 15.1	75.9 +/- 15.4	74.6 +/- 11.1	0.94	0.40	0.62
<b>Physical limitations</b>	31.8 +/- 6.8	62.1 +/- 14.3	42.7 +/- 18.5	<0.001	<0.001	0,005
<b>Physical pain</b>	75,0 +/- 13.7	75.2 +/- 12.3	73.4 +/- 13.9	0.92	0.39	0.06
<b>General health perceptions</b>	61.9 +/- 12.9	64.5 +/- 14.8	62.7 +/- 14.9	0.16	0.47	0.71
<b>Energy/Vitality</b>	61.3 +/- 13.8	64.6 +/- 14.7	59.5 +/- 14.3	0.09	0.1	0.52
<b>Social functioning</b>	73.1 +/- 17.5	75.7 +/- 18.8	75.7 +/- 17.8	0.88	1	0.25
<b>Mental health</b>	73.6 +/- 13.0	67.0 +/- 11.1	71.9 +/- 11.8	<0.001	0.047	0.42
<b>Psychic limitations</b>	70.5 +/- 21.8	76.6 +/- 21.1	76.6 +/- 20.7	0.046	1	0.06
<b>Physical component summary</b>	45.3 +/- 9.1	53.2 +/- 12.4	45.3 +/- 7.1	0.58	0.52	0.69
<b>Mental component summary</b>	53.3 +/- 12.7	52.5 +/- 13.9 eje@bioscientfica.com	52.6 +/- 12.3	0.95	0.60	0.66

### **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.

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.