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Endo-ERN Pituitary Transition of Care Study Group

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RESEARCH

Mapping of the current transition of care practice for patients with pituitary disease at Endo-ERN reference centers

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Abstract

Rare endocrine conditions present specific diagnostic and management challenges for healthcare providers, one of which is the understudied transition of care. Despite the need for guidance regarding transition, consensus on structured and protocolled approaches is lacking. Therefore, we aimed to map the current clinical practice and identify unmet needs regarding transition of care for patients with pituitary disease in the reference centers (RCs) of the European Reference Network on Rare Endocrine Conditions (Endo-ERN). A survey-based, cross-sectional study using the EU Survey tool was performed and completed by 46 physicians (*n*) from 30 RCs (*N*). Transition is a common practice among RCs ($n = 44/46$), usually accomplished by a multidisciplinary team meeting ($N = 20/30$). Criteria for start and end of transition were defined in half of the RCs, with 16.7% of centers providing dissimilar answers. Transition readiness was assessed by >75% of the RCs, mostly by unvalidated means (e.g. subjective opinions, informal consultations). Pituitary-specific transition assessment tool was applied in one RC only. Transition protocols were present in only 9% of RCs, while in many RCs, transition decisions were taken in combined adult-pediatric meetings or based on clinicians' personal judgment. A minority of physicians evaluated the effectiveness of transition-related interventions ($n = 11/46$) or medical outcomes ($n = 8/46$). Patient-reported outcome measures were infrequently used ($n = 4/46$). Identified unmet needs included the development of guidelines ($n = 5/46$) and EU-wide approach ($n = 2/46$). This study exemplifies the unmet needs for a structural definition of the transition period and transition management for patients with rare hypothalamic and pituitary conditions from healthcare providers' perspective.

Key Words
- transition of care
- pituitary disease
- Endo-ERN
- survey
- quality of care

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Background

Disorders of the pituitary gland can affect individuals of any age or sex, depending on the type of the disease (1, 2). Diagnoses include neoplasms of the pituitary fossa, for example, craniopharyngiomas, Rathke's cleft cysts, or functioning and non-functioning pituitary adenomas (1, 2, 3), and congenital or acquired hypopituitarism (defined by either a partial or a complete deficiency of pituitary hormone secretion) (4). Other ultra-rare manifestations of pituitary diseases include pituitary carcinomas, autoimmune disorders, and vascular events (e.g. apoplexy) (3). Due to the differences in age-related prevalence, the adult endocrinologist has generally more expertise in pituitary adenoma and the pediatric endocrinologist in congenital forms and craniopharyngiomas, so shared care is relevant to ensure optimal expertise.

The management of these conditions requires special precautions during the most delicate phases of patients' lives, for example, in the transition from childhood to adulthood. Despite the importance of this period, agreement on how to define (timing of) transition is nonexistent. Previously, physicians have defined transition as the transitional phase between the end of puberty and reaching peak bone mass, involving a wide range of physical, psychological, and sociocultural changes (5). For healthcare systems, however, transition is defined as the planned movement of adolescents and young adults with chronic physical and medical conditions with associated needs from child-centered to adult-oriented assistance (6). Several authors have defined the start of transition by physical development, with Tanner stage 5 being the starting point (5, 7), whereas other authors preferred to consider chronological age as a cut-off value (8). Optimal expertise for the disease may also affect the decision about transition. Nevertheless, a uniform definition, specifically for patients affected by pituitary diseases, is needed to improve uniformity and (quality) standards of care.

Currently, a well-structured and evidence-based transition of care protocol for patients with pituitary diseases is lacking worldwide. The commitment of the European Reference Network on Rare Endocrine Conditions (Endo-ERN) (9) is to lay the foundations for standardization of care through guideline conformity. Therefore, the aim of this study was to map the current clinical practice regarding transition of care across reference centers (RCs) of Endo-ERN, which have specifically been endorsed for their expertise on hypothalamic and pituitary conditions, and to identify the needs regarding transition of care for these patients. The need to consider the general transition aspects alongside the pituitary-specific ones is highlighted throughout the current work. However, general transition aspects will not be discussed at length, as respondents are primarily pituitary experts.

Methods

Study setting

European Reference Networks (ERNs) have been established in 2017 as virtual networks of RCs with specific expertise in a subgroup of rare and complex conditions across the European Union with the primary aim to reduce present health care inequalities through cross-border expert consultation. At the time of the survey (November 2021), Endo-ERN (https://endo-ern.eu/) included 78 RCs in 19 EU member states. Forty-three of these 78 RCs had been endorsed both nationally and subsequently at the European level for specific expertise for Hypothalamic and Pituitary Diseases – Main Thematic Group 6 (MTG6).

Study design

For this survey-based cross-sectional study, a survey was developed using the online EU Survey tool, as published previously (10, 11, 12). The representatives of the RCs within MTG6 were approached by email with a link to the survey. Both adult and pediatric endocrinologists listed as Endo-ERN MTG6 representatives for the respective expert center were eligible for this survey. At the start of the survey, each participant was asked whether they agreed to continue after an initial explanation about the survey's aim and scope.

After an initial email, two reminders were sent to non-responders, the first reminder approximately 4 weeks and the second reminder approximately 8 weeks following the initial email. RCs that did not respond following both reminder emails were considered non-respondents. A response rate of 60% of the RCs was *a priori* considered as satisfactory for analysis (12), which meant that a cut-off of ≥26 RCs filling out the survey was required for the present study. For each RC, multiple respondents were accepted, whereas solely one response was accepted per physician. In the case of duplicate surveys by one physician, the study team communicated with the responding physician regarding the final answers to be used for data analysis. Partial completions of the survey by one respondent were included in the study analysis due to the independent nature of the survey questions.

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Survey content

The survey questions were aimed at establishing the clinical expertise of the center (volume of patients and diseases managed), characteristics of the transition process, and principles behind its organization and conduct. The survey, therefore, included 31 questions presented in open-ended and closed format (both binary and multiple choice). The survey was divided into two sections: (a) general aspects about the practice of the physicians and (b) transition of care aspects (e.g. highlighted definitions, care-set management, protocols, and outcomes). More specifically, questions targeted transition-specific information, including which professionals were involved into the process of transition; how often combined meetings were held; which criteria were used to define the initiation and the end of the transition; which methods and outcomes were used to assess its success. Open-ended questions were aimed at investigating what should have been the definition of transition, the purpose of a well-structured transition program, as well as giving the possibility to add personal suggestions. A Likert scale was used to assess the perceived need for potential intervention strategies. The full version of the survey is available in Supplementary Table 1 (see section on supplementary materials given at the end of this article).

Statistical analyses

Descriptive statistics were used to present data, with categorical variables being presented as numbers (*N* or *n*) with percentages. Capital '*N*' was used to denote the number of RCs and lower case '*n*' for the number of physicians. Data were reported for RCs and for individual respondents, in some cases, stratified by adult and pediatric endocrinologists. Differing answers to binary questions describing transition practices among RC representatives were considered as 'dissimilar;' different answers to multiple choice questions describing transition practices among RC representatives were considered as 'complementary'. Total scores were derived by summing up individual responses.

Results

Response rate and demographics

In total, 30 out of 43 MTG6 RCs responded to the survey, resulting in a response rate of 70%. From those 30 RCs, 46 individual physicians filled out the survey. Among the 46 responders, 30 (65.2%) were adult endocrinologists and 16 (34.8%) were pediatric endocrinologists. Most of the RCs (17/30 or 56.7%) provided 1 responder (11 adult and 6 pediatric endocrinologists) or 2 responders (4 RCs (13.3%) represented by 2 adult endocrinologists, and 6 RCs (20%) represented by 1 adult and 1 pediatric endocrinologist). Furthermore, 3 RCs (10%) provided 3 responses (all represented by 2 adult and 1 pediatric endocrinologist). Thus, the total number of physicians was 46 from 30 expert centers.

Characteristics of responding RCs

Virtually, all centers (*N* = 28/30, 93.3%) treated a large contingency of patients with pituitary diseases – over 100 patients visiting the outpatient clinic annually. The distribution of conditions managed, as shown in Table 1, indicated that hypopituitarism was managed by all RCs and physicians, while acromegaly was managed by the fewest RCs (*N* = 15/30, 50%) and physicians (*n* = 18/46, 39%). In 13 (43%) of the responding RCs and by 10 (22%) of the responding physicians, all conditions were managed.

Definitions of transition

The first section of the survey was directed at defining transition, and all responders were asked to provide an appropriate definition of transition. The response rate for this specific question was 78.3% (*n* = 36/46). All characteristics of the definitions are summarized in Table 2. Twelve of the definitions given included one of the keywords from Table 2, while 22 out of 36 (61.1%)

Table 1 Distribution of expertise and total number of patients. Distribution of RCs (middle column) and number of physicians (right column) involved in the treatment of certain pituitary disease ordered by the frequency of answers. Most common is hypopituitarism treated in all centers, one of the less common pituitary conditions is acromegaly – managed in 50% of reference centers. Thirteen RCs are involved in managing all the mentioned conditions. Data are shown as numbers (*N* or *n*) with percentages (%).

Conditions	**RCs** (*N* = 30)	**Physician** (*n* = 46)
All	13 (43%)	10 (22%)
Hypopituitarism	30 (100%)	46 (100%)
Craniopharyngioma	27 (90%)	40 (87%)
Cushing's syndrome	20 (67%)	28 (61%)
Prolactinoma	22 (73%)	31 (67%)
NFPA	17 (57%)	19 (41%)
Acromegaly	15 (50%)	18 (39%)
Prader–Willi syndrome		3 (7%)

NFPA, nonfunctioning pituitary adenoma; RCs, reference centers.

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Table 2 Definitions of transition. Length and keywords included in the definitions of transition given by respondents. Each participant was asked to define the transition process in the form of an open answer. Keywords from all definitions were combined in groups according to their meaning. The number of times each term was used is displayed and also what percentage of the definitions it was included in.

		Physicians (n = 46)
Elements included in the definition of transition of care	No definition	10 (21.7%)
	Pediatric to adult transfer	31 (67.4%)
	Planning	9 (19.6%)
	Joint clinics/efforts	6 (13%)
	Autonomy	4 (8.7%)
	Patient expectations/needs	3 (6.5%)
	Parent involvement	3 (6.5%)
	Pituitary	3 (6.5%)
	Writing a summary/letter	4 (8.7%)
	Administrative burden	1 (2.2%)
	Building physician expertise	1 (2.2%)

were longer and included 2 or more keywords. Three of the definitions resembled closely the definition given by the Got Transition program: '*The process of moving from a child/family-centered model of care to an adult/patient-centered model of care*' (https://www.gottransition.org). The majority of physicians (n = 31/46, 67.4%) acknowledged the fact that transition involved the transfer from pediatric health care settings to adult health care settings. The need for planning (n = 9/46, 19.6%), combined pediatric-adult clinics (n = 6/46, 13%), patient autonomy (n = 4/46, 8.7%) and parent involvement (n = 4/46, 8.7%) were respectively the second, third, and fourth most common keywords seen in the definitions. In two instances, transition was viewed as administrative burden or as opportunity to build physicians expertise. In four of the definitions, chronological age was mentioned as a factor in the transition process, while in one, puberty was the landmark of the time for transition. Solely in 3 out of the 36 responses (8.3%), pituitary conditions were included in the definition.

Transition protocol

For the presence of transition protocol, consistent answers were given by the majority of RCs (N = 22/30, 73.3%). A protocol was available in two RCs (9%), while in the other RCs, (90.9%) decisions were made at combined meetings or based on transition readiness, as reported in one of the RCs. In five of the RCs (20%), dissimilar answers were given by their representatives to whether such protocols were available. Around a fifth of responders (N = 9/46, 19.6%) reported that a protocol was under development. Virtually, all responders (n = 44/46, 95.7%) pointed to the fact that adolescents with pituitary disease underwent transition from pediatric to adult health care setting in their RC, except for one of the RCs where both school-age pediatric and adult patients were managed by a medical specialist with expertise in the specific condition.

Transition of care practices

Start and end of transition of care

The criteria for start and end of the transition period were the topic of the following section of the survey. In one-third of the RCs, criteria were not available (N = 10/30, 30%), whereas in half of the RCs (N = 15/30, 50%), criteria were available. This tendency was valid for both subgroups of pediatric and adult endocrinologist: 9 out of 16 pediatric (56.3%) and 18 out of 30 adult endocrinologists (60%) confirmed the presence of criteria (Supplementary Table 1).

Determinants for the start of transition of care

The most influential factors for the start of transition were age (n = 42/46, 91.3%), personal judgement of the clinician (n = 25/46, 54.3%), skills of the patient (n = 22/46, 47.8%), wish of the patient (n = 21/46, 45.7%), and outcomes (n = 14/46, 30.4%), as shown in Fig. 1. When asked about the chronological age of start of transition, the most common answer was 16–18 years old (n = 22/40, 55%), and the least common answer was <14 years (n = 1/40, 2.5%).

Transition readiness

Readiness for transition was routinely considered and assessed by >80% of the responders (N = 26/30, 86.7%). Mostly, conclusions regarding readiness were based on

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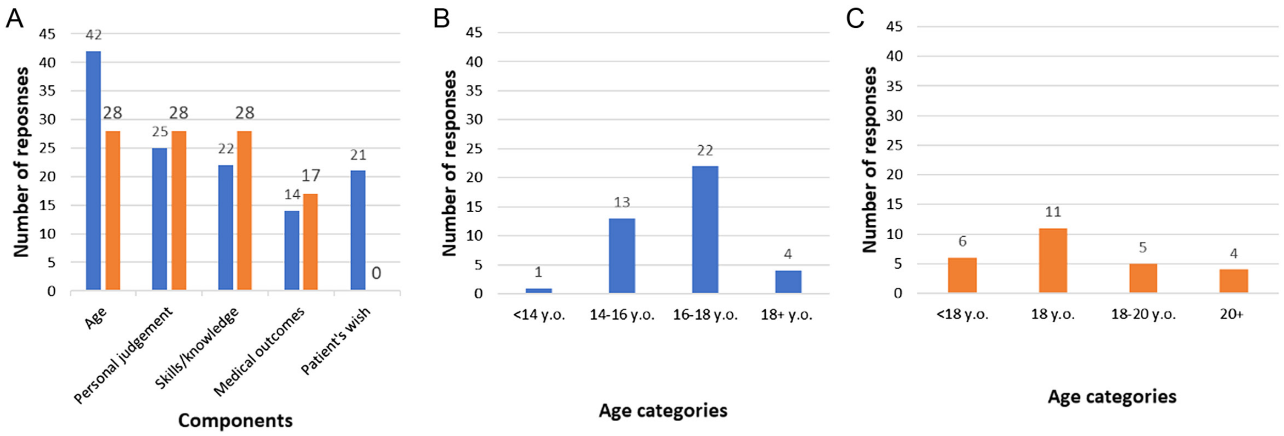

Figure 1
Criteria defining borders of transition period. (A) The bar chart demonstrates the number of respondents that consider each of the factors shown as important for the beginning (blue) and the end (orange) of the transition process. (B) Number of physicians considering the age at transition initiation. (C) Number of physicians considering the age at end of transition. (B) and (C) show the average age at which the respondents usually initiate and end the transition process. Forty of the respondents gave information for the age at start and 20, for the age at end.

subjective opinions, including informal consultation with a colleague (*n* = 14/26, 53.9%) or asking parents' opinion (*n* = 13/26, 50%). Other mentioned methods included talking to the patient alone and inquiring about their expectations. Three adult endocrinologists reported using validated tools: Transition Readiness Assessment Questionnaire (TRAQ) (13), ReadySteadyGo (14), and Endocrine Society transition readiness assessment tool for patients with pituitary hormone deficiencies, (https://www.endocrine.org/improving-practice/transitions), of which only the latter was tailored to patients with pituitary diseases.

Multidisciplinary team meetings

In most of the RCs, multidisciplinary meetings regarding transition occurred regularly (*N* = 13/20, 65%), whereas in 7 of the 30 RCs (20%), they were conducted only for difficult cases, as summarized in Table 3. Apart from pediatric and adult endocrinologists, the multidisciplinary team included a specialized nurse (*N* = 26/30, 86.7%), a psychologist (*N* = 15/30, 50%), a gynecologist, and a geneticist (both *N* = 14/30 46.7%). The distribution of other medical and non-medical specialists is shown in Table 3.

Transfer documentation

Out of the 26 RCs and 36 physicians that reported the presence of written systematic practices in the transfer process, documents used were health summary (*N* = 18/26, 69.2%), and transfer plan (*N* = 15/26, 57.6%). Three of the responders were not involved at all in the transfer process. The distribution of these characteristics per center and per physician is shown in Table 4. The most important characteristics included in the patient summary were comorbidities (*n* = 36/36, 100%), medical history (*n* = 36/36, 100%), and current medication (*n* = 36/36, 100%). Least reported were the patient-reported outcome measures (PROMs; by 8.3% of RCs (*N* = 2/24) and 5.6% of clinicians (*n* = 2/36)).

Determinants for the end of transition of care

The end of the transition period was influenced equally by the judgment of the clinician, patients' skills, and age (*n* = 28/46, 60.9%). Medical outcomes played a role in the end of transition according to 17 participants (37%). The age at which transition ended was specified by 26 of physicians (56.6%) and was considered 18 years by 11 responders (23.9%) (Fig. 1).

Outcomes

Only a minority of physicians evaluated the effectiveness of transition-related interventions (*n* = 11/46, 23.9%) or medical outcomes following transition (*n* = 8/46, 17.4%), which was observed in both pediatric and adult endocrinologists, as shown in Table 5. The medical outcomes specified by the eight responders were loss to follow-up (*n* = 8/8, 100%), adherence to therapy (*n* = 7/8, 87.5%), emergency admissions (*n* = 5/8, 62.5%), and rebound to pediatric care (*n* = 4/8, 50%). PROMs were least often used in the evaluation of transition (*n* = 4/46, 8.7%).

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Table 3 Multidisciplinary teams and transition meetings. The table shows the coverage of transition per endocrinologist, the members of the multidisciplinary team, and the frequency of transition meetings.

		RCs (*N* = 30)
Specialists health care professionals involved	Nurse	26 (86.7%)
	Psychologist	15 (50.0%)
	Gynecologist	14 (46.7%)
	Geneticist	14 (46.7%)
	Social worker	3 (10.0%)
	Urologist	1 (3.3%)
	Neurosurgeon	1 (3.3%)
	Andrologist	1 (3.3%)
	Cardiologist	1 (3.3%)
Frequency of transition meetings[a]	Regular	13/20 (65.0%)
	Irregular[b]	7/20 (35.0%)

[a]For the last indicator, the maximum number of RCs is 20 – this equals the number of RCs for which information was shared; [b]Some of the responders did not specify the frequency of meetings but reported having them only for difficult cases – they are included in the ‘irregular’ group.

Unmet needs and future directions

Results from the survey questions regarding the necessary changes in the current transition practices (using a 5-point Likert scale) are represented in Fig. 2. Key priorities reported were interventions that would enhance patients’ self-management and adherence to treatment and improvement of the collaboration between pediatric and adult endocrinologists. Twelve of the participants pointed out the unfulfilled needs in the transition of care, which included development of a guideline ($n = 5$), development of an EU-wide approach ($n = 2$), further mapping the needs of the practitioners ($n = 1$), sharing currently existing protocols ($n = 1$), and definition of successful transition ($n = 1$).

Discussion

The present survey-based study investigated the current practices of transition of care for adolescents with pituitary diseases across Endo-ERN RCs, with the aim to assess the differences in transition management and to identify gaps and unmet needs in the current practices. Major findings were the widespread lack of pituitary-specific transition protocols, heterogeneity of practices, and lack of affirmed measurable outcomes. These issues are acknowledged by clinicians and are perceived as major unmet needs for optimal quality of care for patients with pituitary diseases.

The life phase of transition is difficult to manage because of the many challenges that occur in that period of time, especially when having a chronic endocrine disease

Table 4 Documentation used and information included in the transfer of patients. The use of different document types and information included in them for transfer of pituitary patients. Numbers displayed are counts and percentages for RCs (*N*) and physicians (*n*), respectively.

Question		**RCs** (*N* = 26)	**Physician** (*n* = 46)
Documentation at patient transfer	Health summary	18 (69.3%)	31 (67.4%)
	Transfer plan	15 (57.7%)	25 (54.3%)
	Not involved	3 (11.5%)	7 (15.2%)
Question		**RCs** (*N* = 24)	**Physician** (*n* = 36)
Information included in the transfer documentation	Age	23 (95.8%)	33 (91.7%)
	Comorbidities	24 (100%)	36 (100%)
	Past illness	24 (100%)	36 (100%)
	Current medication	24 (100%)	36 (100%)
	Adherence	23 (95.8%)	34 (94.4%)
	Future appointment	19 (79.1%)	25 (69.4%)
	Planned test	21 (87.5%)	27 (75%)
	Planned interventions	17 (70.8%)	20 (55.6%)
	PROMs	2 (8.3%)	2 (5.6%)

PROMs, patient reported outcomes.

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Table 5 Evaluation of transition effectiveness and clinical outcomes. Distribution of outcome measures among pediatric, adult endocrinologists, and in RCs.

Question		**Physicians** (*n* = 46)	**Pediatric** (*n* = 16)	**Adult** (*n* = 30)
Perform effectiveness evaluation	No	35 (76.1%)	11 (68.75%)	24 (80%)
	Yes	11 (23.9%)	5 (31.3%)	6 (20%)
Use outcomes to describe transition success	No	38 (82.7%)	14 (87.5%)	24 (80%)
	Yes	8 (17.4%)	2 (12.5%)	6 (20%)
Use of PROMs	No	42 (91.3%)		
	Yes	4 (8.7%)		

(1). Many additional aspects of a patient's personal life are complicated by the preexisting diseases or its sequels and comorbidities and need special attention to prevent detrimental disruption (15). Therefore, there is a high risk in these patients' special needs being unmet.

Clearly, the first and most difficult challenge to overcome is the lack of a uniform definition for transition of care. In the present literature, differing definitions reflect many perspectives (*vide supra*). Based on the survey results, definitions most commonly emphasized the process of transferring care to adult services, the need for planning, and the interdisciplinary approach required to perform the transferring. Interestingly, two of the responders view transition of care as either an administrative burden or as a possibility to improve physicians' expertise, responses crucial to identifying facilitators and barriers toward a successful transition process (16). Transition of care is certainly complicated by the administrative tasks required for the referral of a patient. Simultaneously, expertise building is an issue for adult endocrinologists, who had to manage pediatric conditions without being accustomed to these age-related conditions and challenges.

Only a few RCs reported that a written transition protocol or 'guideline' was currently available, which might be explained by the lack of literature on this topic or lack of uniformity in clinical practice, highlighting the need for transition of care guidelines. In the absence of protocols, responding RCs and healthcare providers have managed transition of care differently. Based on the survey results, transition was most commonly managed by pediatric-adult combined meetings, although these meetings varied in number and frequency.

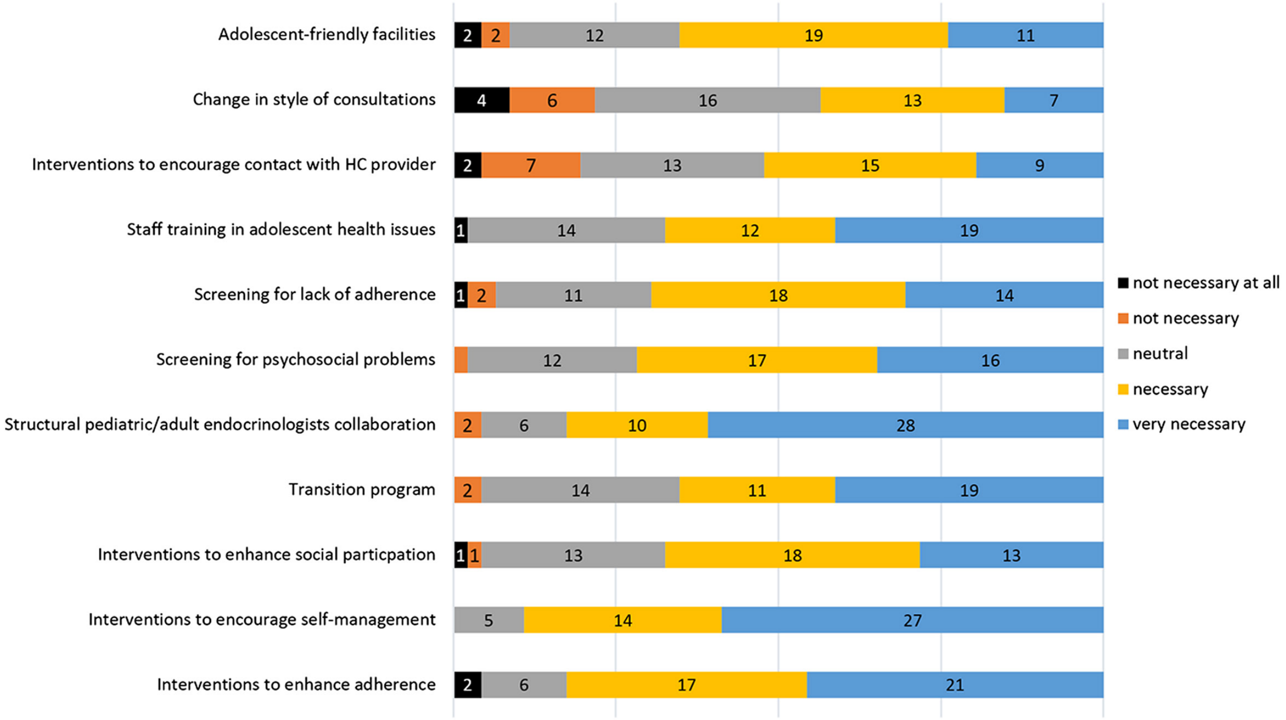

Figure 2
Likert scale results. Most needed changes in the current transition practices according to the respondents.

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Moreover, the course and timing of transition are mostly based on expert opinion rather than an algorithm. This flexible, personalized approach demonstrated the lack of coherent guidelines for transition of care for pituitary patients, with a clinical practice guideline being a current unmet need. The fact that protocols are under development at a number of RCs indicates that the problem is acknowledged. Ideally, aligned multicenter efforts on a European level would benefit patient care.

At present, specific tools (e.g. questionnaires) to assess the readiness for transition of care are seldom used in the responding RCs, although there are multiple tools available, albeit mostly untailored to pituitary disease (e.g. Ready Steady Go and TRAQ (13, 14)). The only pituitary-specific tool used was the transition readiness checklists and questionnaires by the Endocrine Society, but this tool is not incorporated in a structured transition program, unfortunately for unknown reasons. The overlooking of developed tools is an important observation, as it may be one of the barriers to implementing a transition protocol. The use of generic tools has the advantage of uniformity and comparability between conditions, as well as many psychosocial aspects are in common for chronic illnesses (17, 18). By contrast, a disease-specific tool would offer the possibility to track disease progression and (more) detailed measures of medical management. In one of the RCs where a transition of care protocol is under development, a complete transition of care strategy is applied, combining TRAQ with multidisciplinary transition clinics, electronic reminders, and assessment of PROMs. This integrated approach has been proven beneficial in a recent systematic review (19). Whether the generic, the disease-specific, or a mixed approach is preferable for patients with pituitary diseases should be addressed in the future. Regardless, transition intervention appeared to be cost-effective (20).

Despite the lack of transition protocols, factors influencing the timing of transition are well-known and appreciated by clinicians. Age was the most dominant factor influencing the start of transition. By contrast, age is considered less important for the end of transition. Moreover, medical outcomes and skills of the patient take on a more important role at the end of transition. The average age of transition onset from our survey is higher than that usually cited in the literature (https://www.gottransition.org) (21, 22).

This discrepancy between the theoretical framework and practical application could be explained by administrative requirements or specificity of pituitary population, or societal changes with time, although these questions cannot be answered using the present survey data. Age alone is not a sufficient criterion for appropriate timing. Therefore, establishing a transition 'window' is vital prior to the development of protocols or guidelines.

Another prerequisite for successful management of rare diseases – the multidisciplinary team – is an integral part of the transition process. The present observations confirm that the circumstances required for structured transition program may already be present at most of the RCs. Multidisciplinary team meetings combined with pediatric-adult clinics, transfer documentation, and assessment of readiness (with or without a specific tool) are already applied in large proportion of the centers. Despite the fact that most of the prerequisites for structured transition may already be present, multiple decisions were taken based solely on the specialists' opinion. Beyond the healthcare providers involved, innovative transition models could include alternative clinical environments and methods that may suit young people more such as phone/video appointments, text message communication, or flexible evening consultations (23).

The success of transition programs and interventions in pituitary disease should be explored further, despite the fact that they are proven valuable in other chronic diseases, based on criteria like loss to follow-up or adherence (24, 25). Taking hypopituitarism as an example, certain biochemical, auxological, and psychosocial parameters are widely accepted goals of treatment. In view of the transition process, few of the respondents view medical outcomes as markers for its success, demonstrating lack of understanding that the transition process directly affects medical outcomes and the need for unified criteria for assessment of pituitary transition interventions.

Another important observation is that large proportions of the clinicians do not routinely measure the effects of transition-related interventions, with PROMs being the least employed method of evaluation. PROMs enable putting the patients experience in a context of value and thereby provide additional information on the results and cost-effectiveness of interventions (26). In addition to the traditional medical outcomes, PROMs – and perhaps even patient-reported experience measures (PREMs) – should be implemented in a tailored pituitary transition protocol (27). As there is no unanimity concerning the set of medical (physician-reported) and PROMs, the success of the transition process is not defined, which requires further research as it is of great practical importance.

An alternative to the transition clinics and combined meetings mentioned by one of the RCs is

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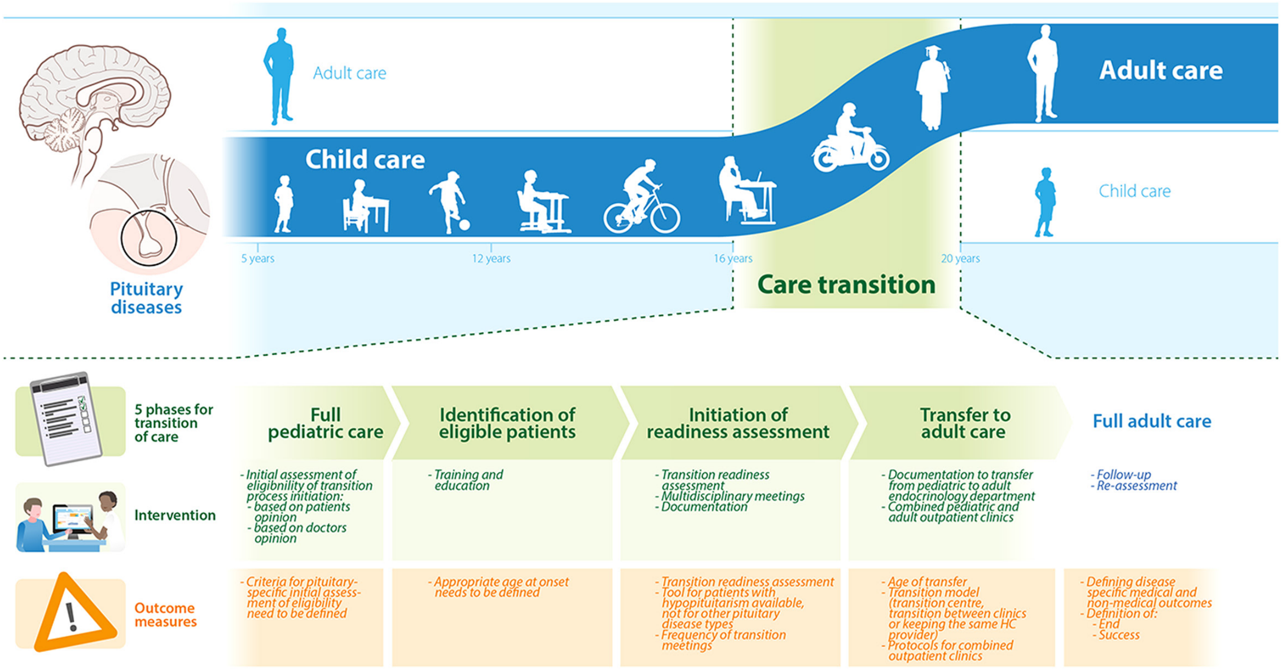
that endocrinologists with expertise in specific diseases take care of the patients independent of age. This is a model in which certain problems of transition (e.g. the fragmentation of services and transfer to new settings and team) are avoided, whereas other medical and organizational problems (e.g. larger administrative staff required) arise. A similar non-transitional model has been described in diabetes care as well (28). Notably, this model of uninterrupted care has not been compared to transition of care model yet. Moreover, this model of care does not reflect the specific expertise required to manage either pediatric or adult patients. Therefore, the current level of knowledge and medical practice indicates that transition of care model might be preferable to tailor to the patients' needs with the physicians' expertise.

Following the present exploration of the practice and organization of transition of care in Endo-ERN RCs, multiple important facets have become evident: (a) transition of care is commonplace, mostly planned, and most often done at combined clinics; (b) generic and pituitary-specific (focused on hypopituitarism) transition readiness assessment tools are available but rarely used; (c) transition of care for pituitary disease guidelines are needed; (d) methods to assess success and outcomes of transition are needed, including patient experiences. Translating these findings and clinical expertise, we propose the following model for transition of care in pituitary disease and highlight the most important unmet needs in Fig. 3. Transition of care can be described in five phases: (a) full pediatric care, (b) identification of eligible patients, (c) initiation of readiness assessments, (d) transfer from pediatric to adult care, and (e) full adult care. For all phases, both known and unknown aspects (i.e. unmet needs) were identified, which require further analysis of the landscape and possibilities. Future research should focus on the identification of barriers to the transition process, critical outcomes (both general and pituitary-specific), and the most appropriate measures to measure these outcomes. These are some of the necessary steps that precede any future work on a guideline in the topic. Ideally, such a guideline should be developed by a multinational board taking in consideration evidence-supported patient and expert opinion. In light of the poor standardization of transition practices, such efforts should be a priority for European pituitary experts.

Several limitations need to be addressed. Multiple biases possibly influence the results of this survey. Selection bias is likely, as not all European countries are represented, and the geographical location of RCs was not distributed equally among countries. Moreover, all



5 phases for transition of care	Full pediatric care	Identification of eligible patients	Initiation of readiness assessment	Transfer to adult care	Full adult care
Intervention	- Initial assessment of eligibnility of transition process initiation: - based on patients opinion - based on doctors opinion	- Training and education	- Transition readiness assessment - Multidisciplinary meetings - Documentation	- Documentation to transfer from pediatric to adult endocrinology department - Combined pediatric and adult outpatient clinics	- Follow-up - Re-assessment
Outcome measures	- Criteria for pituitary-specific initial assessment of eligibility need to be defined	- Appropriate age at onset needs to be defined	- Transition readiness assessment - Tool for patients with hypopituitarism available, not for other pituitary disease types - Frequency of transition meetings	- Age of transfer - Transition model (transition centre, transition between clinics or keeping the same HC provider) - Protocols for combined outpatient clinics	- Defining disease specific medical and non-medical outcomes - Definition of: - End - Success

Figure 3

Transition timeline. An optimal timeline of transition based on the survey results. Five major phases are displayed: pediatric care, identification of eligible patients, readiness assessment, transfer to adult care, and full adult care. For each of these phases, appropriate interventions and unmet needs are also shown in the figure.

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respondents are pituitary experts and might therefore overvalue pituitary-specific tools and questionnaires. Finally, twice as many adult endocrinologists participated in the survey, which may have biased the results.

Conclusion

To date, the clinical practice of transition of care for patients with hypothalamic and pituitary conditions varies greatly, even in expert RCs endorsed by Endo-ERN for the diagnosis, treatment, and management of these specific rare conditions. Definitions of successful transition of care and factors facilitating the successful transition should be identified. The proposed model of transition of care for patients with pituitary disease needs to be developed further and systematically assessed in the future.

Supplementary materials

This is linked to the online version of the paper at https://doi.org/10.1530/EC-22-0308.

Declaration of interest

All authors listed have made substantial, direct, and intellectual contribution to the work and approved it for publication.

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Ethics approval and consent to participate

Since no patient data were collected during this study, approval of the Medical Ethical Committee was not required.

Author contribution statement

All authors listed have made substantial, direct, and intellectual contribution to the work, and approved it for publication. The Endo-ERN respondents of Reference centers for MTG6 which are part of the Endo-ERN pituitary transition of care study group. All authors evaluated the study.

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