



HAL
open science

Transition of young adults with endocrine and metabolic diseases: the TRANSEND cohort

Enora Le Roux, Florence Menesguen, Isabelle Tejedor, Marc Popelier, Marine Halbron, Pauline Faucher, Sabine Malivoir, Graziella Pinto, Juliane Léger, Stéphane N Hatem, et al.

► To cite this version:

Enora Le Roux, Florence Menesguen, Isabelle Tejedor, Marc Popelier, Marine Halbron, et al.. Transition of young adults with endocrine and metabolic diseases: the TRANSEND cohort. *Endocrine Connections*, 2020, 10.1530/EC-20-0520 . hal-03104007

HAL Id: hal-03104007

<https://hal.sorbonne-universite.fr/hal-03104007>

Submitted on 8 Jan 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.

Title**Transition of young adults with endocrine and metabolic diseases: The "TRANSEND" cohort****All authors names and full addresses**

Le Roux Enora ^a, Menesguen Florence ^b, Tejedor Isabelle ^b, Popelier Marc ^c, Halbron Marine ^c, Faucher Pauline ^d, Malivoir Sabine ^b, Pinto Graziella ^e, Léger Juliane ^f, Hatem Stephane ^e, Polak Michel ^g, Poitou Christine ^d, Touraine Philippe ^{*b}

^a Université de Paris, ECEVE UMR 1123, Inserm, Paris, France, Paris, France ; AP-HP.Nord-Université de Paris, Hôpital Universitaire Robert Debré, Unité d'épidémiologie clinique, Inserm, CIC 1426, F-75019 Paris, France; ^b AP-HP. Sorbonne Université, Hôpital Universitaire Pitié Salpêtrière-Charles Foix, Service d'Endocrinologie et médecine de la reproduction ; Centre de maladies endocriniennes rares de la croissance et du développement; Centre de pathologies gynécologiques rares, Paris, France ; ^c AP-HP. Sorbonne Université, Hôpital Universitaire Pitié Salpêtrière- Charles Foix, Service de diabétologie, Paris, France ; ^d AP-HP. Sorbonne Université, Hôpital Universitaire Pitié Salpêtrière- Charles Foix, Service de nutrition, Centre du Syndrome de Prader-Willi et autres obésités rares, Paris, France ; ^e AP-HP.Centre-Université de Paris, Hôpital Universitaire Necker enfants malades, Département d'endocrinologie, diabétologie et gynécologie pédiatrique, Paris, France ; ^f AP-HP.Nord-Université de Paris, , Hôpital Universitaire Robert Debré, Service d'Endocrinologie Diabétologie Pédiatrique, Centre de Référence des Maladies Endocriniennes de la Croissance et du développement, Paris, France ; ^g Director of the Institute of Cardiology and Nutrition, Paris, France.

Corresponding author's postal and email address

Pr Philippe Touraine (email: philippe.touraine@aphp.fr)

Postal adress : Service d'Endocrinologie et médecine de la reproduction

Hôpital Universitaire Pitié Salpêtrière- Charles Foix

47-83 Boulevard de l'Hôpital,

75013 Paris, FRANCE

Short title : Transition cohort in adult endocrinology care

Keywords Transition to adult care ; Endocrinology ; Case Management; Cohort; Care pathway; Rare Diseases

Word count of the full article, excluding references and figure legends : 2896

ABSTRACT

Objective. The transition from paediatric to adult medicine involves risks of poor patient outcomes and of significant losses of patients to follow up. The research aimed to analyse the implementation in an initial cohort of patients of a new program of transition to adult care based on a case management approach.

Design. A longitudinal study of the case management approach to transition, initiated in a university hospital in France in September 2016.

Methods. Patients with endocrine or metabolic disease diagnosed during childhood and transferred to adult care were included. The transition program includes 3 steps based on case management: liaising with paediatric services, personalising care pathways, and liaising with structures outside the hospital (general practitioners, agencies in the educational and social sector).

Results. The cohort included 500 patients, with malignant brain tumour (n=56 (11%)), obesity (n=55 (11%)), type 1 diabetes (n=54 (11%)), or other disease (n=335 (67%)). Their median age at transfer was 19, and the sex ratio was 0.5. At median 21 months of follow-up, 439 (88%) had regular follow-up in or outside the hospital, 47 (9%) had irregular follow-up (absence at the last appointment or no appointment scheduled within the time recommended), 4 had stopped care on doctor's advice, 4 had died, 3 had moved, and 3 had refused care. The program involved 9,615 case management actions; 7% of patients required more than 50 actions. Patients requiring most support were usually those affected by a rare genetic form of obesity.

Conclusions. Case managers successfully addressed the complex needs of patients. Over time, the cohort will provide unprecedented long-term outcome results for patients with various conditions who experienced this form of transition.

INTRODUCTION

Metabolic and endocrine diseases in children and adolescents include more than 500 different diseases with an overall incidence of between 1: 2000 and 1: 4000 (1). The most common condition is type 1 diabetes, which has an incidence of about 15 cases per 100,000 children under the age of 15 (2), while the rarest have reporting incidences of less than one in every 1,000,000 children. These diseases differ according to the type of involvement (isolated or multisystemic), the organs affected, the age of disclosure and the need for paramedical or social care and support.

In recent decades, the survival rate of these paediatric patients has improved, and their care pathway now involves a transition to adult care around their age of majority. In the literature, this period is associated with poor outcomes that have various consequences for the health of young people: poor clinical results, disruptions of care, visits to emergency departments, hospitalisations and intensive care admissions, especially in endocrine diseases(3,4). In addition, patients and families report low satisfaction with care and increased psychological distress during the transition (5).

For patients, in particular young people with various endocrine and metabolic diseases (6), the transfer to adult care represents a major challenge. In paediatric care, young people generally have a long-standing relationship and are comfortable with a team familiar with their illness and with their personal and social history; however, consultations do not always adapt to meet the needs of adolescents and to encourage their empowerment (7). Patients in adult care, who are considered autonomous, may consult a different caregiver at each visit, while consultations focus mainly on clinical outcomes and do not always address the psychosocial or developmental concerns of young people. Poorly prepared for the management of these paediatric patients, adult specialists may neglect the pathological past of the child and its consequences, both physical and psycho-social (8).

To deal with this situation, recommendations have been developed internationally to support successful transition from paediatric to adult care (9–11). However, most of the content of these recommendations focuses on paediatric preparation. They advocate ongoing information about

transition throughout the care pathway, inclusion of the family, consideration of developmental aspects, patient education and specific resources, coordination with primary care, and flexible adaptation of the moment of transfer to the individual case. Several programs have been developed and published, showing that a prepared and coordinated transition can have a positive impact on patients' health, their experiences of care and the use of care. In a systematic review published in 2017, only 6 of the 39 (12) identified transition programs were delivered in adult services (13), and 5 were dedicated exclusively to young people with type 1 diabetes. The same review showed that programs developed for a specific pathology may be appropriate for young people with various diseases. The published programs are generally accessible only to some individuals on the active patient list: 20% of programs exclude young people with an intellectual disability (13). Finally, the timing of data collection after patient transfer enables assessment of the short- and medium-term outcomes of the interventions, but rarely of longer-term ones (13).

It is in this context that a new program of transition based on case management has been developed in a French adult hospital for young people with various endocrine and metabolic diseases. The development project was based on a survey, conducted by the team involved, of the difficulties faced during transition by young adults with congenital adrenal hyperplasia, hypogonadotropic hypogonadism and growth hormone deficiency (14). The need for development of such a program was reinforced by a second survey showing an association between successful transition to specialist adult care and better health-related quality of life and regular medical follow-up in adulthood (15). The very large number of patients shown to be lost to follow up in this study motivated the development of the TRANSEND project to reconsider the organisation of transition to adult care. Although these surveys focused on specific pathologies, the conclusions and implications that could be drawn concerned a larger group of patients; the TRANSEND project was therefore developed to meet the needs of all chronically ill patients during the transition rather than the specific requirements of a particular disease.

In the light of these observations, the objective was twofold: firstly to implement a transition program in an adult care department that meets the needs of patients and fulfils the recommendations, and secondly to collect data on a large number of patients in transition to study their long-term outcomes.

The TRANSEND program takes place in an adult hospital (hospital de la Pitié-Salpêtrière, Paris, France) in the endocrinology, nutrition and diabetology departments. The steering committee (made up of doctors from the three departments involved) defined the TRANSEND program as a dedicated care pathway for patients in transition, where a case manager has a central role in adapting and coordinating for each patient, from their discharge from paediatric services to their strong attachment in adult services.

The specific objective of the present study was to describe the initial cohort of patients and analyse their experience of a transition managed under the new program based on a case management approach.

MATERIALS AND METHODS

Patients

All the patients cared for by hospital-based paediatricians in transition to adult care, aged less than 25 at transfer and referred to services of nutrition, diabetology or endocrinology (metabolism, reproduction, thyroid and endocrine tumours) were included in the transition program and constitute the study cohort. The study is reported according to the STROBE Statement (16) (Supplementary material 1).

Description of the transition program

A medical steering committee for the TRANSEND program development was created comprising the doctors from the nutrition, diabetology, and endocrinology services. Meetings and informal interviews were carried out with all the professionals of these services involved in the transition (nurses, social workers, psychologists, dieticians, medical secretaries). The information and opinions collected have been combined with the findings of surveys carried out in the department, to define a

care pathway for patients in transition. This development phase was done in parallel with the recruitment of the case manager. The developed program involved three steps based on case management (Figure 1).

Step 1 is dedicated to liaising with paediatric services and patients to facilitate the patient's first visit to adult care (transfer of the file, call-up of the patient, file presentation by transition staff).

Step 2 defines the care pathway in adult service. Upon arrival in the adult service, patients coming from paediatrics systematically meet the case manager and their new doctors. The meeting takes place in a dedicated TRANSEND room, youth-friendly and adapted to all types of mobility and bodily need. The case manager assesses the needs of each patient using interview and questionnaires (on social needs, patients' expectations and patients' autonomy) to offer them a personalised pathway in the hospital involving the different available resources and professionals (medical, paramedical, social). Throughout the follow-up, care pathways are supervised by the case manager, who is identified as the key contact by the patients, and who is reachable by text, phone or mail.

Step 3 focuses on liaising with structures outside the hospital (general practitioner, agencies in the educational and social sectors), to improve the long-term follow-up of patients, and to enhance wide-scale coordination of the life project (ensuring that the patient has appropriate training and employment, a suitable place to live, social activities). (See all the details of the care approach in TidiER checklist (17) –Supplementary material 2)

The program was launched in September 2016, with a full time case manager and a dedicated place in the adult hospital concerned. The program is dynamic and has evolved each year since its launch, the evolutions are discussed and decided with the steering committee. Additional elements based on the observation of patients' needs have been added over time: additional time dedicated to coordination, creation of explanatory leaflets about patients' diseases intended for professionals outside the hospital, dedicated therapeutic education sessions, questionnaires that allow to complete the tools available for the patients' needs' assessment (questionnaire to define patient expectations developed specifically for the program, questionnaire to assess patients' autonomy using the Good2Go (18)).

Data collection and statistical analysis

The data used were collected as part of the case manager's routine activity of monitoring and filing activity reports. The sources of data are the patient's medical file and the case report form used by the case manager in routine collection of all the individual data of the patients related to their arrival and follow-up in the TRANSEND program prospectively. These data are summarised and not identifiable, and their collation does not require Institutional Review Board approval according to European or French regulations (Regulation (Eu) 2016/679 Of the European Parliament and of the Council of 27 April 2016 and repealing Directive 95/46 / EC (General Data Protection Regulation)).

A descriptive analysis was conducted on the data collected between the launch of the program in September 2016 and the first interim analysis realised in March 2020. Results were reported as median (Q1, Q3) for continuous variables and as frequency counts and percentages (%) for categorical variables.

RESULTS

Since its launch, 500 patients have experienced the TRANSEND program (patients with at least one visit). The distribution of the first visits in the program per year has been consistent in time: 144 took place in the first year of the launch, 134 in the second year, 140 in third year, and 82 during the first half of the fourth year. The mean follow up time in the program is 21 months (min : 0 ; max : 42). The majority of patients make their transition from a hospital at a regional level (471/500, 94%), 14 from a hospital outside the regional level, 11 from a private practitioner, and 4 from the social or voluntary sector. Patients included in the program had more than 15 different medical conditions; the main diagnoses and patients' health characteristics are presented in Table 1. Patients had diverse socio-demographic profiles (Table 2) resulting in complex and diverse needs for support. Over the period, the case manager performed 9615 coordination actions, from 1 to 461 per patient. These involved meetings, telephone conversations and emails, with the patient, the family, and intra- and extra-hospital partners. Actions relating to the TRANSEND program (call before first visit, plan and deliver the first meeting between patient and case manager) and to the link with paediatrics (request for medical records), represent 2734 (28%) of coordination actions. Management of hospital appointments (consultations with a doctor or other professional, hospitalisations, delay or postponement of

appointments, reminders in case of appointments not scheduled or not attended, requests for transportation vouchers, admission procedures, information on hospital and services, etc.) represent 3389 (35%) of coordination actions. Follow-up of patients (medical issues, social issues, links with community / private practice, other hospitals, medical-social establishments, paramedical, organisation of neuropsychological assessments) represent 3492 (36%) of coordination actions.

For the 33 (7%) patients whose transition required more than 50 actions, a median of 82 actions were performed. Of these patients, 20 (61%) have obesity (the majority having a rare genetic form of obesity).

The case manager planned care with a dietician for 148 (30%) patients, with a psychologist for 95 (20%) patients, and with a social worker for 44 (9%) patients. Patient education, in groups, was delivered to 34 (8%) patients. A third of patients (n=156) had a second individual facilitation meeting with the case manager, at the request of the doctor, especially during hospitalisation. Of the 500 patients in the cohort, 416 (83%) have regular follow-up in the hospital (next consultation already scheduled and / or within the recommended time limit for the next consultation), 23 (5%) are being followed in another hospital or in a private practice, 47 (9%) have irregular follow-up (absence at the last appointment or no appointment scheduled within the time recommended by the doctor) , 4 have stopped care in a hospital setting on the doctor's advice, 4 have died (3 fatal outcomes of a brain tumour, 1 accident related to an epilepsy seizure), 3 have moved, and 3 have refused care. Among those with regular or irregular follow-up, 149 (30%) were called once or several times to unattended and / or unplanned appointments. Among these patients, 86 (58%) scheduled a new appointment. The reasons given for the irregularity of the follow-up were resistance to medical follow-up (asymptomatic or sceptical patients), concerns linked to life projects (studies, work, their sociology-medical support agency) or to other medical care (oncology, psychiatry, neurology, etc.) and social and family difficulties (no parental support, lack of autonomy). No diagnosis is clearly overrepresented in these patients.

DISCUSSION

In 3 years of activity, the TRANSEND program has demonstrated the possibility of sharing of resources between different services and the acceptance by all professionals in an adult care department of a new organisation and a new worker (the case manager) to improve transition care by providing coordination of care and adapting support to the needs of the patients. The program acceptance is supported by the fact that all patients arriving in this department have been referred to the case manager since the launch of the program, regardless of the doctor in charge. This acceptance is probably enhanced by the fact that the TRANSEND program has developed in response to a locally identified need: that of young patients lost to follow-up during transition (14,15). The acceptability has also been probably strengthened by the involvement of the professionals of the department since the beginning in the steering committee, which developed and made the TRANSEND program evolve.. The TRANSEND program was developed based on the needs of young patients identified in the international literature, which found a lack of awareness of psychosocial and developmental aspects and coordination in adult care (19) but also based on the local needs of young people, relying on local surveys. The program is based on a patient-centred care approach. The case manager and overall organisation enable provision of care that is respectful of, and responsive to, individual patient preferences, needs and values (20). It follows international recommendations on transition by providing information about transition, including to patients' families, considering the developmental aspects, allowing access to patient education and specific resources, and coordinating with primary care; these are particularly valuable and rare in routine adult medical care.

To our knowledge, the TRANSEND cohort is the largest and the most diversified in terms of patient profile in the field of transition. This cohort, whose data are collected routinely, reflects the real life of the program outside any interventional framework. The cohort study will be enriched over the years with other data (care use, self-management, and satisfaction) and new patients. It will result in longer-term study of young people with multiple diseases in post-transition, and identification of factors linked to long term outcomes and unmet needs to inform the evolution of TRANSEND. This continuous improvement of the program is already in progress, based on questionnaire completion by

patients who report their needs to the case manager. lastly it also enables the identification of the young patients' need for more exchange between them.

The TRANSEND program includes patients with various diseases, with or without intellectual disability or in socially complex situations, which will ensure a high external validity for its results. The choice to build a cohort allows for equity in access to quality care. In clinical trials one of the constraints is to select a homogeneous population to gain in precision (often at the expense of patients with complex profiles); this cohort study makes it possible to systematically include the patients followed in the department and subsequently to study all patient profiles, their different needs in terms of accompaniment during transition, and their long term outcomes. Although the transition program offered to patients in this cohort is intended to be universal, a limit is observed. A subgroup of patients in the cohort requires a large number of time consuming actions from the case manager, which was not planned at the outset: a variation of the program to better adapt to the different patient profiles could be considered.

According to a systematic review published in 2016, there is a lack of data from prospective studies on adult care attendance by patients in transition, and few studies have reported attendance in adult care at more than one year after transfer (21). In a retrospective study of congenital adrenal hyperplasia patients, 50% were lost to follow-up after transfer to adult care, with no difference between those who transitioned through a young person clinic and those who did not (22). The same rate was reported in our department before the implementation of the TRANSEND program in patients with this same disease (15). In another study evaluating the effect of an endocrinology transition clinic for patients with diverse diseases, adherence to adult care was 83% (23). In diabetes care, a recent literature review reported that in the absence of a formal transition process for patients with type 1 diabetes, the proportion of patients lost to follow up is reported as between 11% and 62% (24). Although data collection methods, post-transfer evaluation times, and definitions of "patient lost to follow up" are heterogeneous and make comparison difficult, the continued attendance rate reported in the

TRANSEND cohort is much higher than those reported in the literature on young adults with endocrine and metabolic diseases.

A key issue in transition programs is their financial sustainability (25). With 500 active patients the case manager works full-time. The timing of patient exit from the program, initially planned when the patient is 25 years old, seems to have to adapt to the patient's needs. Prioritisation of actions and tasks delivered by the case manager becomes needed with the growth in numbers of patients in the cohort. For example, the necessary coordination with professionals outside TRANSEND remains a challenge: the involvement of private practice medicine, the handover in hospital services and links with paramedics remain fragile after several years of existence of the program. The upcoming evaluation of the costs associated with the program and also of savings (including savings of costs of avoidable care) will help to better inform the relevance and cost-benefit analysis of the program.

The principle of equivalence would not have been respected if the impact of TRANSEND was evaluated in a randomised trial against usual forms of care. Previous usual care practice did not adapt to the situation of transition or to young adults' specific needs. Given the reported data on morbidity and mortality associated with interruptions of care during transition (26), it did not seem ethical to expose the patients of a control group to additional risks of serious or irreversible damage from not receiving an intervention based on recommendations.

However, a comparative non-randomised evaluation of TRANSEND is currently underway. It compares the post-transition outcomes in patients who transferred to adult care before TRANSEND to those who transferred after its implementation and benefited from it. The evaluation is based on the set of successful transition consensual indicators (27) as well as a medico-economic assessment. To conclude, the TRANSEND cohort shows that case-management upon arrival in adult care enhances satisfactory results with regard to follow-up after transfer from paediatrics: 88 % have regular follow-up and only 4 patients in 500 are confirmed as lost to follow-up (0.6%). Achieving these results

involved significant deployment of resources: nearly 10,000 actions of case-management have been performed for the entire cohort.

DECLARATION OF INTEREST

There is no conflict of interest that could be perceived as prejudicing the impartiality of the research reported.

FUNDING

The transition program (dedicated room and case-manager funding) was supported by the "Hôpitaux de Paris – Hôpitaux de France" Foundation ("Transition program" Grant, 2015). The realisation and analysis of the cohort did not receive any specific grant from any funding agency in the public, commercial or not-for-profit sector.

ACKNOWLEDGEMENTS

We would like to thank all the nurses, social workers, dieticians, psychologists, and medical secretaries from the departments involved in the TransEND cohort for participating in the elaboration of the program. We would also like to thank Mrs E Benmansour, Mr S Morel, previous directors of Pitié Salpêtrière Hospital for their support for our proposal to set up such a program. Finally, we would like to thank Pr A Basdevant, Mr F Le Magny and Referis consultants for their advice in building up this program.

The Transend group includes all those doctors who refer their patients to the program and/or follow them in adult care: Prs and Drs. Afenjar, Aigrain, Amoura, Amouyal, Amsellem Jager, An, Andreelli, Azar, Bachelot, Belien Pallet, Beltrand, Bibal, Bidet, Bismuth, Blanc, Bodemer, Bougneres, Bourron, Bouvattier, Brauner, Brioude, Bruckert, Brugieres, Brune, Brunel, Burggraeve, Cabrol, Cacoub, Carel, Carlier, Carreau, Cassuto, Cessans, Chakhtoura, Cheikhelard, Chevignard, Ciangura, Consoli, Couderc, Coupaye, Courtillot, Da Costa, Dabbas, Dalla Vale, Dassa, De Beaufort, De Kerdanet, Delanoe, Delcroix, Diene, Dierick-Gallet, Donadieu, Dormoy, Doummar, Doz, Dubern, Dufour,

Duranteau, Esteva, Fayeche, Fiot, Flandrin, Flechtner, Fresneau, Gajdos, Gallo, Gaspar, Gelwane, Ghander, Giabicani, Gonzalez, Grill, Guilmin-Crepon, Grouthier, Hartemann, Haye, Heide, Houang, Ibrahim, Idbah, Istanbulu, Jacqueminet, Jacquin, Jeandidier, Jeannin, Karsenty, Kenigsberg, Kipnis, Le Bastard, Le Poullenech, Leenhardt, Lefevre, Loison, Lorenzini, Lucidarme, Ly, Manh, Martinerie, Masson, Meneret, Mignot, Milcent, Mircher, Mochel, Moretti, Mosbah, Mourre, Moutafoff-Borie, Muller, Nasser, Navarro, Pasqualini, Paulsen, Pauwels, Personnier, Phan, Piquard Mercier, Porquet-Bordes, Puget, Ribeiro Parenti, Ricour, Rigaux, Robert, Robin, Rougeoreille Cretier, Samara Boustani, Sarnacki, Sauvion, Semeraro, Simon, Storey, Thalassinou, Tounian, Trang, Tubiana-Rufi, Villanueva, Whalen, Zaarour, Zenaty.

REFERENCES

1. Saudubray J-M, Sedel F. Les maladies héréditaires du métabolisme à l'âge adulte. *Ann Endocrinol*. 2009 Mar;70(1):14–24.
2. Patterson CC, Dahlquist GG, Gyürüs E, Green A, Soltész G. Incidence trends for childhood type 1 diabetes in Europe during 1989–2003 and predicted new cases 2005–20: a multicentre prospective registration study. *The Lancet*. 2009 Jun;373(9680):2027–33.
3. Lotstein DS, Seid M, Klingensmith G, Case D, Lawrence JM, Pihoker C, Dabelea D, Mayer-Davis EJ, Gilliam LK, Corathers S, et al. Transition From Pediatric to Adult Care for Youth Diagnosed With Type 1 Diabetes in Adolescence. *Pediatrics*. 2013 Apr 1;131(4):e1062–70.
4. Cadario F, Prodam F, Bellone S, Trada M, Binotti M, Trada M, Allochis G, Baldelli R, Esposito S, Bona G, et al. Transition process of patients with type 1 diabetes (T1DM) from paediatric to the adult health care service: a hospital-based approach. *Clin Endocrinol (Oxf)*. 2009 Sep;71(3):346–50.
5. Hepburn CM, Cohen E, Bhawra J, Weiser N, Hayeems RZ, Guttmann A. Health system strategies supporting transition to adult care. *Arch Dis Child*. 2015 Jun;100(6):559–64.
6. Polak M, Touraine P, editors. *Transition of Care: From Childhood to Adulthood in Endocrinology, Gynecology, and Diabetes* [Internet]. S. Karger AG; 2018 [cited 2020 Jun 16]. (Endocrine Development; vol. 33). Available from: <https://www.karger.com/Book/Home/277185>
7. van Staa AL, Jedeloo S, van Meeteren J, Latour JM. Crossing the transition chasm: experiences and recommendations for improving transitional care of young adults,

- parents and providers: Crossing the transition chasm. *Child Care Health Dev.* 2011 Nov;37(6):821–32.
8. Monaghan M, Hilliard M, Sweenie R, Riekert K. Transition Readiness in Adolescents and Emerging Adults with Diabetes: The Role of Patient-Provider Communication. *Curr Diab Rep.* 2013 Dec;13(6):900–8.
 9. American Academy of Pediatrics, American Academy of Family Physicians, American College of Physicians-American Society of Internal Medicine. A consensus statement on health care transitions for young adults with special health care needs. *Pediatrics.* 2002 Dec;110(6 Pt 2):1304–6.
 10. Canadian Paediatric Society , Adolescent Health Committee. Transition to adult care for youth with special health care needs. *Paediatr Child Health.* 2007 Nov;12(9):785–93.
 11. Singh SP, Anderson B, Liabo K, Ganeshamoorthy T. Supporting young people in their transition to adults' services: summary of NICE guidance. *BMJ.* 2016 May 11;i2225.
 12. Gabriel P, McManus M, Rogers K, White P. Outcome Evidence for Structured Pediatric to Adult Health Care Transition Interventions: A Systematic Review. *J Pediatr.* 2017 Sep;188:263-269.e15.
 13. Le Roux E, Mellerio H, Guilmin-Crépon S, Gottot S, Jacquin P, Boukdedid R, Alberti C. Methodology used in comparative studies assessing programmes of transition from paediatrics to adult care programmes: a systematic review. *BMJ Open.* 2017 Jan 27;7(1):e012338.
 14. Godbout A, Tejedor I, Malivoir S, Polak M, Touraine P. Transition from Pediatric to Adult Healthcare: Assessment of Specific Needs of Patients with Chronic Endocrine Conditions. *Horm Res Paediatr.* 2012;78(4):247–55.
 15. Bachelot A, Vialon M, Baptiste A, Tejedor I, Elie C, Polak M, Touraine P, the CRMERC study group. Impact of transition on quality of life in patients with congenital adrenal hyperplasia diagnosed during childhood. *Endocr Connect.* 2017 Oct;6(7):422–9.
 16. von Elm E, Altman DG, Egger M, Pocock SJ, Gøtzsche PC, Vandenbroucke JP for the STROBE Initiative. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) statement: guidelines for reporting observational studies. *Lancet Lond Engl.* 2007 Oct 20;370(9596):1453–7.
 17. Hoffmann TC, Glasziou PP, Boutron I, Milne R, Perera R, Moher D, Altman DG, Barbour V, Macdonald H, Johnston M, Lamb SE, Dixon-Woods M, McCulloch P, Wyatt JC, Chan AW, Michie S. Better reporting of interventions: template for intervention description and replication (TIDieR) checklist and guide. *BMJ.* 2014 Mar 7;348(mar07 3):g1687–g1687.
 18. Mellerio H, Jacquin P, Trelles N, Le Roux E, Belanger R, Alberti C, Tubiana-Rufi N, Stheneur C, Guilmin-Crépon S, Devilliers H. Validation of the “Good2Go”: the first French-language transition readiness questionnaire. *Eur J Pediatr.* 2020 Jan;179(1):61–71.

19. Peters A, Laffel L, the American Diabetes Association Transitions Working Group. Diabetes Care for Emerging Adults: Recommendations for Transition From Pediatric to Adult Diabetes Care Systems: A position statement of the American Diabetes Association, with representation by the American College of Osteopathic Family Physicians, the American Academy of Pediatrics, the American Association of Clinical Endocrinologists, the American Osteopathic Association, the Centers for Disease Control and Prevention, Children with Diabetes, The Endocrine Society, the International Society for Pediatric and Adolescent Diabetes, Juvenile Diabetes Research Foundation International, the National Diabetes Education Program, and the Pediatric Endocrine Society (formerly Lawson Wilkins Pediatric Endocrine Society). *Diabetes Care*. 2011 Nov 1;34(11):2477–85.
20. Crossing the Quality Chasm: A New Health System for the 21st Century [Internet]. Washington, D.C.: National Academies Press; 2001 [cited 2020 May 20]. Available from: <http://www.nap.edu/catalog/10027>
21. Rachas A, Lefeuvre D, Meyer L, Faye A, Mahlaoui N, de La Rochebrochard E, Warszawski J, Durieux P. Evaluating Continuity During Transfer to Adult Care: A Systematic Review. *Pediatrics*. 2016 Jul;138(1):e20160256.
22. Gleeson H, Davis J, Jones J, O’Shea E, Clayton PE. The challenge of delivering endocrine care and successful transition to adult services in adolescents with congenital adrenal hyperplasia: experience in a single centre over 18 years. *Clin Endocrinol (Oxf)*. 2013 Jan;78(1):23–8.
23. Twito O, Shatzman-Steuerman R, Dror N, Nabriski D, Eliakim A. The “combined team” transition clinic model in endocrinology results in high adherence rates and patient satisfaction. *J Pediatr Endocrinol Metab*. 2019 May 27;32(5):505–11.
24. White M, O’Connell MA, Cameron FJ. Transition to adult endocrine services: What is achievable? The diabetes perspective. *Best Pract Res Clin Endocrinol Metab*. 2015 Jun;29(3):497–504.
25. Hobart CB, Phan H. Pediatric-to-adult healthcare transitions: Current challenges and recommended practices. *Am J Health Syst Pharm*. 2019 Sep 16;76(19):1544–54.
26. Sheehan AM, While AE, Coyne I. The experiences and impact of transition from child to adult healthcare services for young people with Type 1 diabetes: a systematic review. *Diabet Med*. 2015 Apr;32(4):440–58.
27. Suris J-C, Akre C. Key Elements for, and Indicators of, a Successful Transition: An International Delphi Study. *J Adolesc Health*. 2015 Jun;56(6):612–8.

FIGURE LEGENDS

Figure 1 : Schematic representation of the TRANSEND care pathways

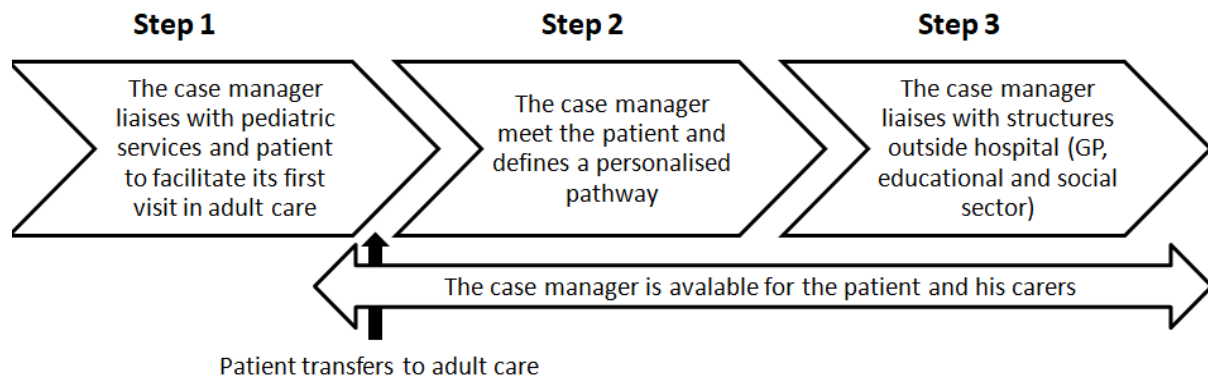


Figure 1: Schematic representation of the TRANSEND care pathway.

Table 1 : Cohort medical characteristics

Diseases	N (%)
Malignant brain tumour (except craniopharyngioma)	56 (11)
Obesity	55 (11)
Type 1 diabetes	54 (11)
Congenital hypopituitarism	43 (9)
Disorders of pubertal development	41 (8)
Thyroid diseases	38 (8)
Prader Willi syndrome	35 (7)
Congenital adrenal hyperplasia	27 (5)
Turner syndrome	22 (4)
Growth delay	18 (4)
Non congenital hypopituitarism	17 (3)
Craniopharyngioma	16 (3)
Familial dyslipidemia	8 (2)
Other conditions (rare diseases, pituitary benign tumour, patients at risk following disease or treatment)	70 (14)
Pre-transfer referral	
Hospital pediatrician	474 (95)
Hospital adult specialist	11 (2)
Private practice pediatrician	7 (1)
General practitioner	4 (1)
Social or associative sectors professional	4 (1)

Table 2 : Cohort socio-demographic characteristics

	Med (Q1-Q3)
Age the first appointment	19 (17-20)
Sex	N (%)
Male	250 (50)
Female	250 (50)
Place of residence	
Parents' home	423 (85)
Self home	52 (10)
Medico-welfare establishment	22 (4)
Other	3 (<1)
Scholarship or professional status (MD=13)	
Post graduate studies	199 (40)
High school or vocational school	117 (23)
Medico-pedagogic institute	91 (18)
No activity	39 (8)
Ordinary work	33 (7)
Adapted to disability work	4 (<1)

*MD: missing data

Supplementary material 1: STROBE Statement—Checklist of items that should be included in reports of cohort studies

	Item No	Recommendation	Page No
Title and abstract	1	(a) Indicate the study's design with a commonly used term in the title or the abstract	1
		(b) Provide in the abstract an informative and balanced summary of what was done and what was found	2
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	3
Objectives	3	State specific objectives, including any prespecified hypotheses	3
Methods			
Study design	4	Present key elements of study design early in the paper	5-6
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	5-6, supplementary material 2
Participants	6	Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up	5

Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	5-6, Sup material 2
Data sources/ measurement	8	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	6
Bias	9	Describe any efforts to address potential sources of bias	6
Study size	10	Explain how the study size was arrived at	N/A
Quantitative variables	11	Explain how quantitative variables were handled in the analyses.	6
Statistical methods	12	(a) Describe all statistical methods, including those used to control for confounding	6
		(b) Describe any methods used to examine subgroups and interactions	N/A
		(c) Explain how missing data were addressed	N/A
		(d) If applicable, explain how loss to follow-up was addressed	N/A
		(e) Describe any sensitivity analyses	N/A
Results			
Participants	13	(a) Report numbers of individuals at each stage	7

		of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	
		(b) Give reasons for non-participation at each stage	N/A
		(c) Consider use of a flow diagram	N/A
Descriptive data	14	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	Tables 1 and 2
		(b) Indicate number of participants with missing data for each variable of interest	Tables 1 and 2
		(c) Summarise follow-up time (eg, average and total amount)	7-8
Outcome data	15	Report numbers of outcome events or summary measures over time	7-8
Main results	16	(a) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	N/A
		(b) Report category boundaries when continuous variables were categorized	Tables 1 and 2
		(c) If relevant, consider translating estimates of	N/A

		relative risk into absolute risk for a meaningful time period	
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	N/A
Discussion			
Key results	18	Summarise key results with reference to study objectives	8
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	8-9
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	8-9
Generalisability	21	Discuss the generalisability (external validity) of the study results	8-9
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	10

Supplementary material 2 : The TIDieR (Template for Intervention Description and Replication)

Checklist:

Item n°	Item	Page	Other † (details)
	BRIEF NAME		
1.	Provide the name or a phrase that describes the intervention.	P3	TRANSEND A support program for patients in transition to adult endocrinology services after discharge from the paediatric services.
	WHY		
2.	Describe any rationale, theory, or goal of the elements essential to the intervention.	P3-4	TRANSEND was created to facilitate the healthcare pathway of young adults in transition from paediatric to adult care in the objective to avoid rupture in care during this pivotal period.
	WHAT		
3.	Materials: Describe any physical or informational materials used in the intervention, including those provided to	P5-6, Sup. Materia 1 2	Patients are welcomed in a dedicated room designed for young adults, in which flyers and information brochures are arranged (social initiatives, associations, etc.). Specific materials were developed for the program and are used during patient interviews with the case manager: interview

	<p>participants or used in intervention delivery or in training of intervention providers. Provide information on where the materials can be accessed (e.g. online appendix, URL).</p>		<p>scripts (including a list of actions to be carried out and free notes), social questionnaire (designed by social workers) and corresponding doctor's cards. "Rare diseases" practical sheets (produced by doctors) inform on the specific surveillance measures of these diseases. They are distributed to the patient and/or sent to their general practitioner. A numeric version of these files is available on the hospital's intranet. Transition patient education tools and projection equipment and tablets (intended for meetings between professionals and patient groups) are stored in the TRANSEND room and offices.</p>
4.	<p>Procedures: Describe each of the procedures, activities, and/or processes used in the intervention, including any enabling or support activities.</p>	P5-6	<p>The steering committee (made up of doctors from the three departments involved) defined a care pathway for patients in transition, divided into three steps. Step 1 is dedicated in liaising with paediatric services and patient to facilitate its first visit in adult care (transfer of the file, call of the patient, file presentation in transition staff). The inclusion of patients in transition in TRANSEND is systematic for adolescents</p>

			<p>and young adults having a chronic endocrine pathology, referred by paediatrics, and having a consultation in one of the adult departments of endocrinology-reproductive medicine, nutrition, diabetology, endocrinology-metabolic diseases.</p> <p>Step 2 defines the care pathway in adult service. Upon arrival in adult service, the patients coming from paediatrics systematically meet the coordinator and their new doctors. The coordinator assesses the needs of each patient to offer them a personalised pathway in the hospital involving the different available resources and professionals (medical, paramedical, social). Thorough the follow-up, care pathways are supervised by the coordinator, who is identified as the key contact by the patients, who is reachable by text, phone or mail.</p> <p>Step 3 focuses in liaising with structures outside hospital (GP, educational and social sector), to improve the long-term follow-up of patients, and to enhance large scale coordination of the project of life.</p>
--	--	--	--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------

	WHO PROVIDED		
5.	For each category of intervention provider (e.g. psychologist, nursing assistant), describe their expertise, background and any specific training given.	P5-6 and Sup. Materia 1 2	<p>At the beginning, a medical steering committee was built with the doctors from the services of Nutrition, Diabetology, and Endocrinology for the TRANSEND program development.</p> <p>For the regular monitoring of the program since its launch, a TRANSEND project committee was created and is composed of the medical project manager (head of the endocrinology department), the psychologist coordinating the transition patient education program, the responsible of the transition database and the case manager.</p> <p>The case manager is full-time on the TRANSEND program. She holds a state diploma in psychomotricity and a master's degree in rehabilitation and medical engineering, and a specialty in neurological disability in adults. This position requires clinical and professional network knowledge.</p>
	HOW		
6.	Describe the modes of delivery (e.g. face-to-face or by some other mechanism, such as internet or telephone) of the intervention and whether it was provided	P5-6 and Sup. Materia 1 2	<p>A telephone interview with the patient takes place before his first meeting with the coordinator in the TRANSEND dedicated room. A second meeting with case manager may be organised during an hospitalisation. These are individual interviews, but the patient may be accompanied by his parents. Patient education sessions are provided to few patients, in individual meeting or in little groups.</p>

	individually or in a group.		The patient care pathway and hospital appointments are monitored by the case manager thanks to the hospital intranet and result (if necessary) in calls of patients (consultation reminder or reschedule)
	WHERE		
7.	Describe the type(s) of location(s) where the intervention occurred, including any necessary infrastructure or relevant features.	P5-6 and Sup. Materia 1 2	<p>The TRANSEND program takes place in an adult hospital (hospital de la Pitié-Salpêtrière, Paris, France) in the endocrinology, nutrition and diabetology departments.</p> <p>The TRANSEND room is equipped for individual interviews and for groups. It is accessible to people with reduced mobility. The furniture is youth-friendly and suitable for all body types.</p> <p>Patient monitoring takes place in the case manager office with computer and phone.</p>
	WHEN and HOW MUCH		
8.	Describe the number of times the intervention was delivered and over what period of time including the number of sessions, their schedule, and their duration, intensity or dose.	P8	<p>The TRANSEND program started in September 2016. Two and a half years later, the cohort of patients in transition is 500.</p> <p>Details are in the results part of the article.</p>
	TAILORING		
9.	If the intervention was planned to be personalised, titrated	P8	The first meetings with the case manager are carried out systematically at the start of care in the adult department.

	or adapted, then describe what, why, when, and how.		On the other hand, consultations with the doctors of the service, planning of social worker appointment or proposition of patient education are determined individually according to the needs of the patient, by the case manager in consultation with the healthcare team.
	MODIFICATIONS		
10.†	If the intervention was modified during the course of the study, describe the changes (what, why, when, and how).	Sup. Materia 1 2.	The patient education program was offered to patients from June 2018.
	HOW WELL		
11.	Planned: If intervention adherence or fidelity was assessed, describe how and by whom, and if any strategies were used to maintain or improve fidelity, describe them.	Sup. Materia 1 2	TRANSEND has been the subject of activity reports every six months since September 2016, written by the case manager. The project committee meets every month to review program data and discuss optimisation and / or evaluation.
12.	Actual: If intervention adherence or fidelity was assessed, describe the extent to which the intervention was delivered as planned.	N/A	N/A

CPP - Ile-de-France VI

Groupe Hospitalier Pitié-Salpêtrière

Président : Nathalie BRION

Vice-Président : Marie-Cécile MASURE

Audrey BELLESOEUR - Kevin BIHAN - Laurent CAPELLE - Christophe DEMONFAUCON - Micheline DENANCE - Jacqueline DUNO - Marie-Hélène FIEVET - Marie GICQUEL-BENADE - Cloé GIQUEL - Clarisse GOUDIN - Gilles HUBERFELD - Etienne KIMMEL - Annie LE FRANC - Esther LELLOUCHE - Christiane LOOTENS - Michèle MEUNIER-ROTIVAL - Marie-Caroline MEYOHAS - Anne-Laure MORIN - Thang NGUYEN - Sabine PLANCOULAIN - Sophie TEZENAS DU MONTCEL - Martyna TOMCZYK - Dominique VARIN

Madame Enora LE ROUX
Monsieur le Professeur Ph. TOURAINE
Unité d'Epidémiologie Clinique/Inserm
CIC-EC 1426 - UMR123 ECEVE
Hôpital Universitaire Robert-Debré

Paris, le 7 juillet 2020

Dear Colleagues,

We confirm that your study *Transition of young adults with endocrine and metabolic diseases : The "TRANSEND" cohort* is not relevant to the French law (loi sur les Recherches Impliquant la Personne Humaine (RIPH)) and you don't need an authorization of CPP. The CPP sees no objection in its realization.

Sincerely Yours,



Le Président du CPP
Professeur Nathalie BRION