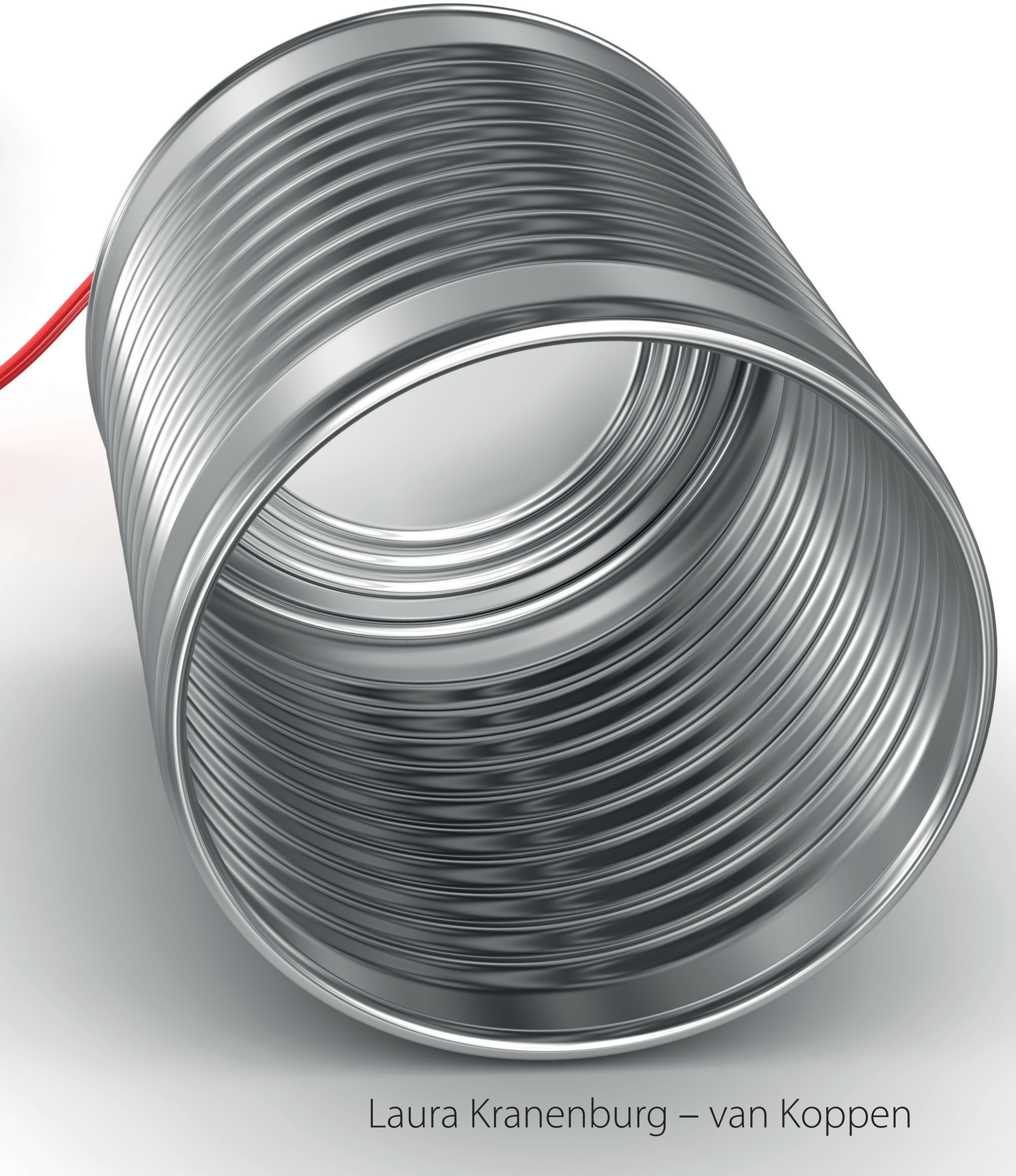
Times change:

How to train future medical specialists to become skilled communicators



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Laura Joanne Christine Kranenburg - van Koppen

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How to train future medical specialists to become skilled communicators

Tijden veranderen:
Hoe toekomstige medisch specialisten
op te leiden tot vaardige communicatoren

Thesis

to obtain the degree of Doctor from the
Erasmus University Rotterdam
by command of the
rector magnificus

Prof. dr. A.L. Bredenoord

and in accordance with the decision of the Doctorate Board.
The public defence shall be held on

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by

Laura Joanne Christine Kranenburg - van Koppen
born in Rotterdam, Netherlands.

Erasmus University Rotterdam

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CHAPTER 1

Introduction

Mr. and Mrs. de Bruin visit a rheumatologist in the out-patient clinic for the first time. They are referred because Mrs. De Bruin suffers from pain in her shoulder and hip. They are a bit nervous, because another medical specialist was skeptical about the appointment. They describe him as 'the professor', who only looks at his computer and tells them what to do. It was already clear to 'the professor' that she did not have any rheumatic disease. Mr. and Mrs. De Bruin are surprised that the rheumatologist really listens and asks what matters to them most. Mrs. De Bruin is severely invalidated by the pain and avoids long walks, what she loved to do in the past. The diagnosis severe inflammatory arthrosis is made, which improved substantially by a low dose of steroids. Mrs. De Bruin experiences a splendid effect: she was able to walk around the shopping mall, what she had not done in years. Benefits and risks of long-term steroids, such as risk for osteoporosis, possible elevated glucoses, weight gain and bruising are carefully discussed and compared with Mrs. De Bruins values and personal goals. In the end, risks are acceptable and Mrs. De Bruin is relieved that she is possible to participate in daily life again. She wonders what 'the professor' will think of it…

This example illustrates the importance of effective doctor-patient communication, which implies active listening, searching for patients' values and personalized needs and jointly making decisions based on evidence, benefits and risks. Effective doctor-patient communication forms the heart and art of modern medicine and results in patient centered care[1-4]. Much patient dissatisfaction and many complaints are due to breakdown in the doctor-patient relationship[5-7]. Despite good intentions, doctors tend to overestimate their communication skills[5 8]. Furthermore, doctors seem to overestimate their abilities of understanding of patients' health beliefs and needs[9 10]. Studies on doctor-patient communication have demonstrated patient discontent even when many doctors considered the communication adequate or even excellent[5].

Aims and outline of this thesis

Communication skills for medical specialists of the future are formed during postgraduate medical training. Main question of this thesis is how we can support postgraduate training of communication skills for future medical specialists. Two aspects are considered. First aspect is the postgraduate training of communication skills in daily practice in chronic diseases, as good doctor-patient communication forms the backbone of treatment in chronic diseases. Secondly, specific attention is needed for communication skills concerning rare diseases. In rare diseases evidence of treatment is often lacking due to small numbers in studies, which stresses the importance of shared decision making and involving patients' values and goals in medical decision making.

This introduction will discuss **postgraduate training of communication skills**. The backbone of postgraduate learning is **learning in practice, formative assessment and feedback**, which will be discussed in order to understand the educational background of learning and teaching. Finally specific aspects will be discussed regarding **communication skills in chronic diseases** and **communication skills in rare diseases**. We conclude with an **outline of the chapters**.

Skills training for doctor-patient communication

The undergraduate medical curriculum contains several communication courses dispersed throughout the curriculum. Medical students acquire a satisfactory level of communication competency early in their undergraduate training[11]. During postgraduate medical specialist training these basic skills should be further developed to expert levels.

However, communication skills achieved by medical students early in their studies do not increase substantially in later years[11]. Furthermore, clinical experience has a limited effect on the communication competence of physicians.[11] Ideally, increasing medical knowledge and clinical practice go hand in hand with increasing communication skills, resulting in expert levels of doctor-patient communication skills. At postgraduate level, doctors report that they are aware of the importance of adequate doctor—patient communication, however they face difficulties when trying to apply those communication skills in their actual workplace. Continuous formal training in communication is mostly absent and when it is indeed offered, effectiveness studies demonstrate moderate to little effect in the long run.[12]

Extensive research has been performed regarding training of communication skills. Doctor-patient communication skills can be taught in courses, are learnt, but are easily forgotten if not maintained by practice[13]. Effective training strategies include active, practice-oriented strategies.[14] Evidence for effectiveness of communication training programs varies.[1 12 14-22] This might be explained by the way effects are measured and how the outcome of these programs is perceived[23]. The acquisition of communication skills is an ongoing process.[23] Checklists are perceived as an inhibiting factor in achieving communication skills, as these do not do justice to the qualities medical specialist in training want to improve.[24] The most important enhancing factor in post-graduate communication training is encouragement to deliberately practice in an environment in which the value of communication skills is recognized and support is institutionalized with appropriate feedback from role models.[24-26]

Deliberate practice is known effective to improve performance.[27-29] It involves the provision of immediate feedback, time for problem-solving and evaluation, and opportunities for repeated performance.[27] The transformation towards skilled communicator is also a process of deliberate practice that starts with confrontation with (un)desired behavior, raising awareness, searching for alternative approaches, personalization of new techniques and finally internalization of renewed communication techniques.[25] Safety and cognitive & emotional space are labelled as overall conditions influencing this learning process.[25]

All in all, communication is a competency that should be longitudinally learned across the whole training continuum in an authentic setting with a lot of practice in many different contexts with regular meaningful feedback[26]. Instead of learning a set of skills, such a learning process will promote skilled communication that is clinically usable in a versatile goal-oriented way[26].

Knowing this, the question arises to what extent these ingredients for communication training are represented in postgraduate medical specialist training. Particularly given the poor skills reported in communication studies[11 18 30].

Learning in practice, formative assessment and feedback

In the last decades, thoughts on the role of assessment in education have changed. This is best characterized as a shift from assessment of learning to assessment for learning. Traditionally, examinations take place at the end of the instruction and are separated from the educational process. This is referred to as *summative assessment*. The purpose of such an assessment is to determine whether the student has acquired sufficient knowledge, skills, etc.

Assessment for learning, however, is an approach that embeds the assessment process within the educational process, is maximally information-rich, and serves to steer and foster each student's learning to the maximum of his/her ability. *Formative assessment* usually is an important component of this process.

From an educational perspective, communication skills are complex behavioral skills[26]. Prior research showed that assessment of communication skills should not be reduced to check-lists, but should be context-rich.[24] Repetitive, meaningful feedback by patients, educators, medical specialists and other colleagues will enable the future medical specialist to develop their own communication style build on a rich collection of experiences.[26]

Feedback is essential for learning any complex skill.[31 32] The credibility of the source of feedback relates to the use of feedback and a lot of feedback is poorly given therefore not credible[33-36]. Feedback should not be a unidirectional stream of information to the learner, but rather a reflective dialogue in which relationships and culture are very important mediators[26].

Knowing this, the question arises how assessment for learning and feedback can be used to improve communication skills in postgraduate medical specialist training.

Communication aspects in chronic diseases

Extensive research on postgraduate training of communication skills gives us an idea of the most important elements. Prior research was performed in various fields, such as general practice, surgery and oncology.[24 37 38] Question arises to what extent these elements are implemented in the daily practice of postgraduate training in chronic diseases. The field of rheumatology is highly suitable to look into this question.

Chronic rheumatic diseases influence patients in many ways on functioning, work, social contacts and sexuality.[39-42] Variation in disease activity and life events ask for ongoing attention and adjustment of treatment.[39 40 43 44] Furthermore, most rheumatology patients want to be more actively involved in medical decision-making.[45-47] Finally, patient involvement is associated with better outcomes on health status, self-management, adherence, coping behavior and satisfaction with care.[45-47]

The question arises how communication skills are trained in postgraduate training in the field of rheumatology and how this relates to evidence based principles of communication training.

Besides general communication skills specific attention goes out to Shared Decision Making (SDM), supporting patients and healthcare professionals to jointly make a decision based on the best available evidence for treatment options while respecting patient's values and preferences.[48-50] Patients, professionals and all international guidelines plea for a central role of SDM in rheumatology. [43 51-56]

Although highly valued, the majority (>50%) of healthcare professionals experienced problems with the application of SDM in clinical practice, mostly related to time constraints.[57] Other important barriers were the incompatibility of SDM with clinical practice guidelines and beliefs that patients do not prefer to be involved in decision making or are not able to take an active role.[57] Various barriers in the implementation of SDM are known.[58-60] Main barriers for the health care provider are a potential negative attitude towards SDM, lack of knowledge on how to apply SDM, lack of ability and skills in relation to applying SDM, misjudgment of patient preferences and no recognition that SDM is not applied ('we are already doing it').[58]

Question arises how rheumatologists in training think about Shared Decision Making, whether they use it in daily practice and which barriers they perceive.

Communication aspects in rare diseases

The backbone of postgraduate training of communication skills is workplace learning. As stated before repetitive practice of skills, reflection and refinement are important aspects of learning. This is challenging in dealing with rare diseases for several reasons. First of all there is less exposure to rare disease as these are less frequently presented in daily practice. Furthermore, the low number of patients makes it difficult to build experience on how these diseases develop and are optimal treated. Due to small numbers evidence about treatment options is limited and mostly based on case reports. Finally, patients are mostly referred to specialized clinics, which might implicate long-distance communication, especially in rural countries.

The question arises on how to improve doctor-patient communication in rare diseases given these challenges. In order to answer these questions we look into the field of disorders of sex development (DSD), which is a subspecialty of the paediatric endocrinologist. This field of expertise addresses various disorders resulting in a large variation of clinical cases. Disorders of sex development might be diagnosed prenatal, this might come as a surprise at birth or come to light during childhood or puberty. Online learning has been proven equally effective as classroom learning and can be a solution in postgraduate training in rare diseases.[61 62]

Care for individuals with DSD is characterized by varying short- and long-term challenges that may emerge throughout one's lifespan. This ranges from urogenital variation to concerns about fertility, gonadal malignancy, adrenal complications, hormone replacement, bone health and psychosocial support, among others. These complex challenges extend to patients and families who seek to understand DSD in biological, medical and psychosocial contexts. Clinicians widely acknowledge that family and patient support is needed as patients face various challenges throughout the lifespan. This requires, among others, good clinical reasoning skills, but above all good doctor-patient communication skills.

Knowing this, the question arises how to improve doctor-patient communication in rare diseases given these challenges and how online learning can be used to support this.

The first question is how online learning, e-consultation and online patient information can be applied to improve doctor-patient communication. The second question is to what extent e-learning can be used to improve postgraduate training in rare diseases. The third question is how e-learning can be used to train communication competences in rare diseases by the use of formative assessment and feedback. The fourth question is whether formative feedback on communication skills can be optimized by using a rubric format addressing various aspects of communication.

Outline of chapters in this thesis

Chapter 2 provides an overview of the techniques used to train communication skills among rheumatologist in training, how these are perceived by rheumatologists in training and by educators and how these relate to evidence based training of communication skills. This study is based on a survey, semi-structured interviews among rheumatologists in training and educators and a scoping literature search on evidence based principles of postgraduate communication training.

Chapter 3 looks into perceived competences, needs and barriers related to Shared Decision Making among rheumatologists in training. This is done by means of a survey, the outcomes are related to findings in literature. This is necessary to define possibilities and requirements for further postgraduate training in Shared Decision Making skills in chronic diseases.

Chapter 4 gives an overview of possibilities of e-learning, e-consultation and e-information in the field of disorders of sex development. The main questions are how e-learning can be used to train clinical skills in rare diseases, how e-consultation between colleagues/experts can be facilitated and how online information for patients can be optimized. This is done by an inventory of relevant literature and best practices.

Chapter 5 looks into the question of how online learning can be used in teaching on rare diseases by the use of assessment and feedback. In this study we analysed examples of the use of an interactive E-Learning Portal in Paediatric Endocrinology for various groups, such as medical students, medical specialist in training for paediatric endocrinology and paediatricians. Time spent, responses to the questions and user experience were analysed.

Chapter 6 addresses the question on how distance learning can be used in postgraduate training of doctors in rare diseases. Specific attention is paid to the value of multi-source feedback in the training of communication skills on a global level, connected to the role of cultural and religious aspects. Medical specialist in training worldwide studied two online cases and received personal feedback on communication skills from three different international experts. Responses and experiences from medical specialist in training and experts were analysed.

Chapter 7 looks into the question of whether structured assessment and feedback can help to optimize feedback on communication skills in rare diseases. We developed a rubric to assess communication skills of doctors in the field of disorders of sex development on a global level. Experts in DSD, patient representatives and communication experts were involved in the development. The Rubric was evaluated by a group of medical specialists in the field of DSD and adjusted. Differences in the use of the model are analysed. The second objective is to evaluate the quality of communication skills in postgraduate training by means of the developed rubric.

Chapter 8 gives a general discussion of the findings on the research questions in this thesis and puts them into a broader perspective. It concludes with recommendations for future postgraduate training of communication skills and research.

Chapter 9 contains a summary of the thesis.

References

1. Maatouk-Bürmann B, Ringel N, Spang J, et al. Improving patient-centered communication: Results of a randomized controlled trial. Patient Educ Couns 2016;99(1):117-24.
2. Griffin SJ, Kinmonth A-L, Veltman MWM, et al. Effect on Health-Related Outcomes of Interventions to Alter the Interaction Between Patients and Practitioners: A Systematic Review of Trials. The Annals of Family Medicine 2004;**2**(6):595-608.
3. Street RL, Jr., Makoul G, Arora NK, et al. How does communication heal? Pathways linking clinician-patient communication to health outcomes. Patient Educ Couns 2009;**74**(3):295-301.
4. Laidlaw A, Hart J. Communication skills: an essential component of medical curricula. Part I: Assessment of clinical communication: AMEE Guide No. 51. Med Teach 2011;**33**(1):6-8.
5. Ha JF, Longnecker N. Doctor-patient communication: a review. The Ochsner journal 2010;**10**(1):38-43.
6. Halperin EC. Grievances against physicians: 11 years' experience of a medical society grievance committee. West J Med 2000;**173**(4):235-8.
7. Tamblyn R, Abrahamowicz M, Dauphinee D, et al. Physician scores on a national clinical skills examination as predictors of complaints to medical regulatory authorities. Jama 2007;**298**(9):993-1001.
8. Stewart MA. Effective physician-patient communication and health outcomes: a review. Cmaj 1995;**152**(9):1423-33.
9. Street RL, Jr., Haidet P. How well do doctors know their patients? Factors affecting physician understanding of patients' health beliefs. J Gen Intern Med 2011;**26**(1):21-7.
10. Blalock SJ, Slota C, Devellis BM, et al. Patient-rheumatologist communication concerning prescription medications: getting to the gist. Arthritis Care Res (Hoboken) 2014;**66**(4):542-50.
11. Wouda JC, van de Wiel HB. The communication competency of medical students, residents and consultants. Patient Educ Couns 2012;**86**(1):57-62.
12. Kramer AW, Düsman H, Tan LH, et al. Acquisition of communication skills in postgraduate training for general practice. Med Educ 2004;**38**(2):158-67.
13. Aspegren K. BEME Guide No. 2: Teaching and learning communication skills in medicine-a review with quality grading of articles. Medical Teacher 1999;**21**(6):563-70.
14. Berkhof M, van Rijssen HJ, Schellart AJ, et al. Effective training strategies for teaching communication skills to physicians: an overview of systematic reviews. Patient Educ Couns 2011;**84**(2):152-62.
15. Ditton-Phare P, Loughland C, Duvivier R, et al. Communication skills in the training of psychiatrists: A systematic review of current approaches. Aust N Z J Psychiatry 2017;**51**(7):675-92.
16. Kieran K, Jensen NM, Rosenbaum M. See, Do, Teach? A Review of Contemporary Literature and Call to Action for Communication Skills Teaching in Urology. Urology 2018;**114**:33-40.

17. Weiland A, Blankenstein AH, Willems MH, et al. Post-graduate education for medical specialists focused on patients with medically unexplained physical symptoms; development of a communication skills training programme. Patient Educ Couns 2013;**92**(3):355-60.
18. Aspegren K, Lønberg-Madsen P. Which basic communication skills in medicine are learnt spontaneously and which need to be taught and trained? Med Teach 2005;**27**(6):539-43.
19. Makoul G. The SEGUE Framework for teaching and assessing communication skills. Patient Educ Couns 2001;**45**(1):23-34.
20. Delvaux N, Merckaert I, Marchal S, et al. Physicians' communication with a cancer patient and a relative: a randomized study assessing the efficacy of consolidation workshops. Cancer 2005;**103**(11):2397-411.
21. van Dalen J, Kerkhofs E, van Knippenberg-Van Den Berg BW, et al. Longitudinal and concentrated communication skills programmes: two dutch medical schools compared. Adv Health Sci Educ Theory Pract 2002;**7**(1):29-40.
22. Rao JK, Anderson LA, Inui TS, et al. Communication interventions make a difference in conversations between physicians and patients: a systematic review of the evidence. Med Care 2007;**45**(4):340-9.
23. van den Eertwegh V, van Dulmen S, van Dalen J, et al. Learning in context: identifying gaps in research on the transfer of medical communication skills to the clinical workplace. Patient Educ Couns 2013;**90**(2):184-92.
24. van den Eertwegh V, van Dalen J, van Dulmen S, et al. Residents' perceived barriers to communication skills learning: comparing two medical working contexts in postgraduate training. Patient Educ Couns 2014;**95**(1):91-7.
25. van den Eertwegh V, van der Vleuten C, Stalmeijer R, et al. Exploring residents' communication learning process in the workplace: a five-phase model. PLoS One 2015;**10**(5):e0125958.
26. van der Vleuten C, van den Eertwegh V, Giroldi E. Assessment of communication skills. Patient Educ Couns 2019;**102**(11):2110-13.
27. Ericsson KA. Deliberate practice and acquisition of expert performance: a general overview. Acad Emerg Med 2008;**15**(11):988-94.
28. Ericsson KA. Deliberate practice and the acquisition and maintenance of expert performance in medicine and related domains. Acad Med 2004;**79**(10 Suppl):S70-81.
29. van de Wiel MW, Van den Bossche P, Janssen S, et al. Exploring deliberate practice in medicine: how do physicians learn in the workplace? Adv Health Sci Educ Theory Pract 2011;**16**(1):81-95.
30. Wouda JC, van de Wiel HB. How to attain expertise in clinical communication? Paediatr Respir Rev 2013;**14**(4):213-8.
31. Greco M, Brownlea A, McGovern J. Impact of patient feedback on the interpersonal skills of general practice registrars: results of a longitudinal study. Med Educ 2001;**35**(8):748-56.
32. Hulsman RL, van der Vloodt J. Self-evaluation and peer-feedback of medical students' communication skills using a web-based video annotation system. Exploring content and specificity. Patient Educ Couns 2015;**98**(3):356-63.

33. Renting N, Dornan T, Gans RO, et al. What supervisors say in their feedback: construction of CanMEDS roles in workplace settings. Adv Health Sci Educ Theory Pract 2016;**21**(2):375-87.
34. Renting N, Gans RO, Borleffs JC, et al. A feedback system in residency to evaluate CanMEDS roles and provide high-quality feedback: Exploring its application. Med Teach 2016;**38**(7):738-45.
35. Watling C. The uneasy alliance of assessment and feedback. Perspect Med Educ 2016;**5**(5):262-4.
36. Watling C, Driessen E, van der Vleuten CP, et al. Learning from clinical work: the roles of learning cues and credibility judgements. Med Educ 2012;**46**(2):192-200.
37. Barth J, Lannen P. Efficacy of communication skills training courses in oncology: a systematic review and meta-analysis. Ann Oncol 2011;**22**(5):1030-40.
38. Stiefel F, de Vries M, Bourquin C. Core components of Communication Skills Training in oncology: A synthesis of the literature contrasted with consensual recommendations. Eur J Cancer Care (Engl) 2018;**27**(4):e12859.
39. Dures E, Hewlett S, Ambler N, et al. A qualitative study of patients' perspectives on collaboration to support self-management in routine rheumatology consultations. BMC Musculoskeletal Disorders 2016;**17**:129.
40. Dures E, Fraser I, Almeida C, et al. Patients' Perspectives on the Psychological Impact of Inflammatory Arthritis and Meeting the Associated Support Needs: Open-Ended Responses in a Multi-Centre Survey. Musculoskeletal Care 2016:n/a-n/a.
41. Helland Y, Dagfinrud H, Haugen M-I, et al. Patients' Perspectives on Information and Communication About Sexual and Relational Issues in Rheumatology Health Care. Musculoskeletal Care 2017;**15**(2):131-39.
42. Ferguson S, Trupin L, Yazdany J, et al. Who receives contraception counseling when starting new lupus medications? The potential roles of race, ethnicity, disease activity, and quality of communication. Lupus 2016;**25**(1):12-17.
43. Dures E, Hewlett S, Ambler N, et al. Rheumatology clinicians' experiences of brief training and implementation of skills to support patient self-management. BMC Musculoskeletal Disorders 2014;**15**:108-08.
44. Elwyn G, Frosch DL, Kobrin S. Implementing shared decision-making: consider all the consequences. Implement Sci 2016;**11**:114.
45. Nota I, Drossaert CH, Taal E, et al. Patient participation in decisions about disease modifying anti-rheumatic drugs: a cross-sectional survey. BMC Musculoskelet Disord 2014;**15**:333.
46. Brekke M, Hjortdahl P, Kvien TK. Involvement and satisfaction: a Norwegian study of health care among 1,024 patients with rheumatoid arthritis and 1,509 patients with chronic noninflammatory musculoskeletal pain. Arthritis Rheum 2001;**45**(1):8-15.
47. Kjeken I, Dagfinrud H, Mowinckel P, et al. Rheumatology care: Involvement in medical decisions, received information, satisfaction with care, and unmet health care needs in patients with rheumatoid arthritis and ankylosing spondylitis. Arthritis Rheum 2006;**55**(3):394-401.
48. Makoul G, Clayman ML. An integrative model of shared decision making in medical encounters. Patient Educ Couns 2006;**60**(3):301-12.

49. Stiggelbout AM, Van der Weijden T, De Wit MP, et al. Shared decision making: really putting patients at the centre of healthcare. Bmj 2012;**344**:e256.
50. Elwyn G, Frosch D, Thomson R, et al. Shared decision making: a model for clinical practice. J Gen Intern Med 2012;**27**(10):1361-7.
51. Farinha F, Freitas F, Agueda A, et al. Concerns of patients with systemic lupus erythematosus and adherence to therapy - a qualitative study. Patient Prefer Adherence 2017;**11**:1213-19.
52. Garrido-Cumbrera M, Hillmann O, Mahapatra R, et al. Improving the Management of Psoriatic Arthritis and Axial Spondyloarthritis: Roundtable Discussions with Healthcare Professionals and Patients. Rheumatol Ther 2017.
53. Smolen JS, Aletaha D, Bijlsma JWJ, et al. Treating rheumatoid arthritis to target: recommendations of an international task force. Annals of the Rheumatic Diseases 2010;**69**(4):631-37.
54. Smolen JS, Landewé R, Breedveld FC, et al. EULAR recommendations for the management of rheumatoid arthritis with synthetic and biological disease-modifying antirheumatic drugs: 2013 update. Annals of the Rheumatic Diseases 2014;**73**(3):492-509.
55. Hochberg MC, Altman RD, April KT, et al. American College of Rheumatology 2012 recommendations for the use of nonpharmacologic and pharmacologic therapies in osteoarthritis of the hand, hip, and knee. Arthritis Care & Research 2012;**64**(4):465-74.
56. Gossec L, Smolen JS, Ramiro S, et al. European League Against Rheumatism (EULAR) recommendations for the management of psoriatic arthritis with pharmacological therapies: 2015 update. Ann Rheum Dis 2016;**75**(3):499-510.
57. Mathijssen EGE, van den Bemt BJF, Wielsma S, et al. Exploring healthcare professionals' knowledge, attitudes and experiences of shared decision making in rheumatology. RMD Open 2020;**6**(1).
58. van Veenendaal H, van der Weijden T, Ubbink DT, et al. Accelerating implementation of shared decision-making in the Netherlands: An exploratory investigation. Patient Educ Couns 2018;**101**(12):2097-104.
59. Legare F, Ratte S, Gravel K, et al. Barriers and facilitators to implementing shared decision-making in clinical practice: update of a systematic review of health professionals' perceptions. Patient Educ Couns 2008;**73**(3):526-35.
60. Joseph-Williams N, Lloyd A, Edwards A, et al. Implementing shared decision making in the NHS: lessons from the MAGIC programme. Bmj 2017;**357**:j1744.
61. Cook DA, Levinson AJ, Garside S, et al. Internet-based learning in the health professions: a meta-analysis. Jama 2008;**300**(10):1181-96.
62. Sitzmann T, Kraiger K, Stewart D, et al. THE COMPARATIVE EFFECTIVENESS OF WEB-BASED AND CLASSROOM INSTRUCTION: A META-ANALYSIS. Personnel Psychology 2006;**59**(3):623-64.

CHAPTER 2

Postgraduate communication training in the field of rheumatology: what is done and how does this relate to evidence-based learning? A mixed method study involving an online questionnaire, semi-structured interviews and a scoping review

Laura JC Kranenburg | Annelieke Pasma | Mary EW Dankbaar | Natalja Basoski | Haske van Veenendaal | Walter W van den Broek | Johanna MW Hazes

Under review

Abstract

Background

Chronic diseases requires optimal doctor-patient communication. Communication skills are primarily taught during pre-graduate training, but should develop during postgraduate training to a higher level. Residents in the field of rheumatology underline the importance of good communication skills, but lack training. The most important enhancing factor in post-graduate communication training is encouragement to deliberately practice these skills in an environment in which the value of communication is recognized and support is institutionalized with appropriate feedback from role models. Purpose is to study 1) which methods are used to train doctor-patient communication skills among residents in the field of rheumatology in the Netherlands, 2) how these are perceived by residents and educators and 3) how these relate to evidence regarding postgraduate training of communication skills.

Methods

This mixed method study uses an online questionnaire to answer the first research question, semi-structured interviews to answer the second research question and a scoping literature search to answer the third research question.

Results

Cornerstones of methods used during residency training are the attendance of the practice of rheumatologists and trial-and-error. Experienced as most valuable were observation with direct feedback, video-on-the-job and training with actors, however these were rarely used during residency training. Main principles of evidence-based communication training mentioned in literature are 1) awareness of communication skills and reflection, 2) deliberate practice and 3) a safe and supportive environment.

Conclusions

Regular formative assessments followed by (multi-source) feedback should be implemented in order to increase awareness and the support of deliberate practice. Furthermore, staff should be trained to increase awareness on the effect of different communication techniques and to improve skills on feedback related to communication aspects.

Trial registration

None, no health care intervention on human participants involved

Key words

Post-graduate learning; rheumatology; communication training; evidence-based learning

Background

Long-term doctor-patient relationships require worthy communication skills. Chronic diseases, such as e.g. rheumatic diseases, influence daily life in many ways on functioning, work, social contacts and sexuality.[1-4] Variation in disease activity and life events ask for ongoing attention and adjustment of treatment.[1 2 5 6] Most patients want to be more actively involved in medical decision-making.[7-9] Adherence to therapy is poor due to patients' limited knowledge of their disease and treatment options.[10-13] Patient involvement is associated with better outcomes on health status, self-management, adherence, coping behavior and satisfaction with care.[7-9] Doctors report that they are aware of the importance of adequate doctor-patient communication, however they face difficulties when trying to apply those communication skills in their actual workplace.

The basis for doctor-patient communication skills is made during pre-graduate training. During post-graduate training these basic skills should transform to skilled communication, combining increasing medical knowledge with emergent communication skills.[14] Recent study showed that residents in the field of rheumatology in the Netherlands highly value good doctor-patient communication, but feel they lack skills how to apply this. There is unawareness of their own performance in techniques such as Shared Decision Making and misjudgment of patients' preferences.

Extensive research has been performed regarding training of communication skills. Doctor-patient communication skills can be taught in courses, are learnt, but are easily forgotten if not maintained by practice.[15] Effective training strategies include active, practice-oriented strategies.[16] The most important enhancing factor in post-graduate communication training is encouragement to deliberately practice in an environment in which the value of communication skills is recognized and support is institutionalized with appropriate feedback from role models.[14 17 18] Deliberate practice is known effective to improve performance. It involves the provision of immediate feedback, time for problem-solving and evaluation, and opportunities for repeated performance.[19] The transformation towards skilled communicator is also a process of deliberate practice that starts with confrontation with (un)desired behavior, raising awareness, searching for alternative approaches, personalization and finally internalization of renewed communication techniques.[14] Safety and cognitive & emotional space are labelled as overall conditions influencing this learning process.[14]

Little is known about the methods used by residents and educators in the field of rheumatology to train and improve communications skills. In order to improve future training and communication skills we have to start with an inventory of the methods used during residency training. Analysis of perceived effect of these methods related to findings in literature might give insight in current shortcomings in education and training. Therefore, we need to map the literature on a post-graduate communication training to identify key concepts of evidence-based post-graduate communication training.

Aim of this study is to create an overview of methods used to train doctor-patient communication skills amongst residents in the field of rheumatology in the Netherlands and how these are perceived by residents and educators. Furthermore to evaluate how these training methods correspond with evidence-based key elements regarding postgraduate training of communication skills.

Methods

This mixed method study combines quantitative and qualitative analyses involving an online questionnaire, semi-structured interviews and a scoping literature search.

Online questionnaire

In order to quantify which methods are used to improve communication skills an online questionnaire was sent out amongst residents. Question 17 from an existing online questionnaire was used, see appendix I. The questionnaire was developed for another study investigating opinion, experience and barriers related to Shared Decision Making among residents in the field of rheumatology (unpublished). Aspirant members of the Dutch Society of Rheumatology were invited to answer the online questionnaire by invitation of the Dutch Society of Rheumatology by email. A reminder was sent after two weeks. The addressed group was mixed: residents in their pre-training (year 1-3), specialty training for rheumatology (year 4-6) and rheumatologists who just finished their training. Anonymous participation was possible. It was possible to apply contact details in order to be able to contact participants for an additional semi- structured questionnaire. As membership to the Dutch Society of Rheumatology is obligatory, all Dutch residents were approached for participation. Written consent was given. Residents could select training methods from a predefined list with an open option for additional methods. This resulted in a list with training methods. Quantitative data was collected from the online survey as described above using an online survey program. Export was made to Microsoft Excel 2010.

Semi-structured interviews

Interviews were performed by LK, female Medical Doctor in training for rheumatology (5th year of training). Interviewer had prior experience in research on communication aspects in postgraduate training. Two out of eight participants were from the same educational region as the interviewer, there was no prior relationship with other participants.

Participants did not have any knowledge of the personal views of the interviewer related to the aim of the study. For transparency, the investigator stated personal assumptions and personal interests in the research topic prior to the interviews. The theoretical framework is phenomenology, to describe the meaning and significance of experiences of residents and educators. Participants were selected by purposive sampling. Residents who replied to the questionnaire and indicated that they were willing to participate in the interviews were approached by e-mail to participate. Educators were selected as registered by the Dutch Society of Rheumatology. When the principal educator was not available for the interview it was performed with the second educator. Educators were invited by e-mail, followed by an invitation by phone. The number of residents and educators approached depended on saturation of the answers. A representative sample of residents (year of training, region) and educators (region) participated. There were no participants who refused to participate. Residents and educators were interviewed independently. Interviews were performed by telephone between the interviewer and the respondents. Verbal consent was given during the interview. In the semi-structured interviews residents and educators were asked which training methods they perceived as valuable. Residents were asked about their experience with the used training methods investigating perceived factors for success and perceived barriers. Educators were asked about their approach in training of communication skills and perceived success. Overview of the questions used for the semi-structured interviews are provided in appendix II. No audio-recording was made, field notes were made during the interview, followed by a transcript directly after the interview, which was sent to the participants for comment. Data analysis was performed using a qualitative description as Sandelowski described it [20 21] in order to provide an overview of the current status quo of postgraduate communication training in rheumatology. This analysis was divided into several steps. First, the interview transcripts were read several times to allow the readers to become acquainted with the content by LK and AP. Transcripts were analyzed independently by a rheumatologist in training(LK) and psychologist (AP) stating main perceived effect of the training options, perceived factors for success and barriers in improving communication skills. Descriptions were then discussed and adjusted until agreement was reached (LK and AP). All the authors read the descriptions and participated in the stepwise analysis and discussions along the way. Agreement was reached about added value of different training options, factors for success and main barriers.

Scoping literature search

A scoping literature search[22 23] was conducted for evidence of effective elements in postgraduate training of communication skills. Our protocol was drafted using the Preferred Reporting Items for Systematic Reviews and Meta-analysis Protocols (PRISMA-ScR)[23], which was revised by the research team. Articles were considered eligible when these were published, English written and published after 2002, as from this moment there was increasing attention for communication skills after the introduction of competency based education. Initial search was conducted in Pubmed on 2021/02/01. Search was extended to ERIC, SCOPUS and Psycnet on 2021/11/10. Search was performed using the following terms: "Communication" AND "postgraduate" AND ("training" OR "education") Filters applied: from 2002/1/1 - 3000/12/12 AND English language. Articles were scanned by LK on title and abstract. Articles were included in the review when they discussed evidence related to postgraduate medical training of communication skills. Reference lists of key articles and articles identified in the search were checked for additional studies. Data from eligible studies were charted to identify key elements describing principles of evidence-based communication training. These retrieved key elements were clustered into themes. Finally themes were grouped into three main groups.

Results

Forty-two of the 111 (38%) aspirant members responded. All eight regions in the Netherlands were represented. Respondents were mainly in year 4-6 of their training (88%). The male/female ratio amongst participants was higher than amongst the registered aspirant members (10/90% versus 20/80%). This difference is not significant.

Sixteen of the 42 residents provided contact details and were willing to provide an additional interview. Eight of sixteen residents were contacted for an additional interview, which were purposively selected from different regions and with varying years of training. Educators from four of eight regions were available for participation to the interview.

Online questionnaire: Methods used to acquire communication skills
Residents in the field of rheumatology use different approaches in order to train doctor-patient communication skills as is presented in figure 1, illustrating the frequency that training options were selected from a pre-defined list in the questionnaire.

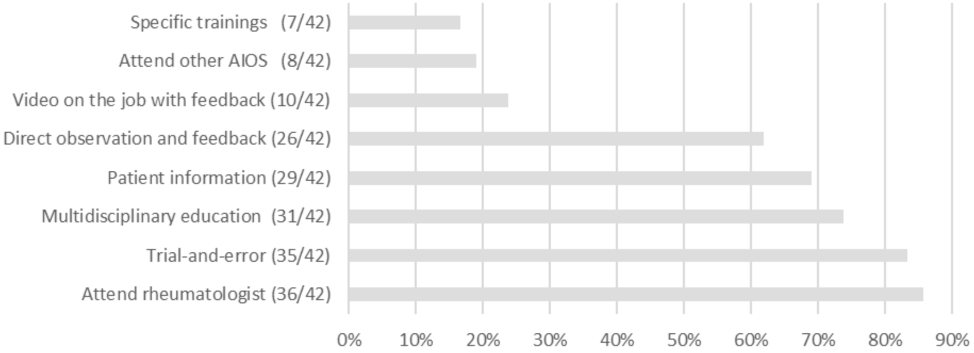

Figure 1. Methods used by residents to train communication skills; residents could choose multiple options from a predefined list of methods.

Semi-structured interview

Qualitative analysis of the interviews with residents shows that through the years residents become more confident about their role as a doctor, which goes hand in hand with more confidence about their doctor-patient communication skills. Many residents indicate that they just do what they think is right. They try new approaches and experience the effect of it. The basis is mostly formed in extensive communication training during pre-graduate medical education. During their postgraduate training there are certain experiences that have resulted in a sustainable changed behavior. Essential components resulting in changed behavior are awareness of undesired behavior, a personalized learning question, adequate feedback and a safe environment.

Qualitative analysis of the interviews with educators shows that the basic skills for doctor – patient communication are learnt before the specialization for rheumatology starts. They also notice that residents differ in the level of interest in doctor-patient communication and in the ability to learn/improve communication skills. Educators indicate that there are many possibilities for feedback, for example video-taping and feedback. Three of four indicate that this can be difficult to realize in practice, mainly due to lack of time. Other barriers are lack of scheduling and lack of initiative by the resident. All educators indicate that video-taping of doctor-patient contact is always possible, but depends on initiative of the resident. Although all educators support the principle of Shared Decision Making, the experience with the subject and how to train their residents differs per center.

Attending the doctor-patient contact of different rheumatologists was chosen by 85% of the residents in the questionnaire and was perceived as very valuable in the interviews (4 out of 8). Residents appreciate the possibility to learn both medical expertise as different communication styles. In this way a rich repertoire is built of both phrases, wording and metaphors as well as non-verbal communication that can be used. Combined doctor-patient consultations are highly valued, especially when performed on a regular basis for a longer period of time. In this way the role of the residents shifts from strict observation to own consultations with observation by the rheumatologist. Feedback, when given, is mostly strictly medical and rarely targeted on communication skills. Explanation given is that the main focus of the combined consultations is on medical content and feedback on communication skills is often forgotten. Drawback mentioned is that it is not always possible to find time to implement combined consultations into daily practice.

Trial-and-error was mentioned by 83% of the residents in the questionnaire. Two out of eight residents explicitly mentioned this as very helpful during the interviews. Trying different approaches and fine-tuning in wording and approach helps to build experience and to get more confidence. Feedback from patients was perceived as helpful. When residents start their specialization they merely focus on medical content. During years of training residents experiment with different communication strategies. Factors for success are deliberate experimenting in approach, followed by reflection on the experienced effect. When asked, residents in the in-depth interview indicate that these key-factors 'deliberate experimenting' and 'reflection on effect' often lack in daily practice.

Multidisciplinary education was mentioned by 74% of the respondents in the questionnaire, but was perceived as valuable by two out of eight residents in the interviews. Two residents were very negative on this subject, stating they have not learnt much of it. Factors that contribute to success are small groups, the quality of training actors, quality of feedback and quality of the teacher. It helps to experience the effect of different communication strategies. It also helps when there is a clear learning question in advance and when timing is right.

Information for patients is studied to improve communication skills by 69% of the respondents of the questionnaire. When asked six out of eight used this specifically at the start of their training to acquire simple wording to explain diagnosis and treatment options. This was regarded as valuable. Several residents state that they want to give their information in line with the other information provided to patients. Furthermore, illustrations in Information for patients are used in daily practice. It also gives insight into patients perceptions and perspectives, especially online patient fora.

Direct observation and feedback was mentioned by 62% of the residents in the questionnaire. During the interviews four out of eight respondents indicated this as an important strategy in improving communication skills. It gives confidence about the chosen approach and insight in small mistakes/misunderstandings that might be missed during supervision on paper. Factors for success mentioned are a clear learning question in advance, high quality feedback and a personalized approach adjusted to the learning style of the resident. Frequency during the first three years of internal medicine is 4-5 times a year. During the last three years this is mostly on demand. New patients are also seen by a rheumatologist, but these contact moments are rarely used for explicit observation and feedback regarding the communication skills of the resident. Availability of a supervisor who has time to attend is mentioned as a barrier both by residents as educators. Furthermore, the attendance of the supervisor might influence the doctor-patient contact.

Video on the job with feedback was only mentioned by 24% residents in the questionnaires. All residents in the interviews were enthusiastic about the potency to improve communication skills. Factors for success are a clear learning question prior to the video-taping and targeted feedback on the learning questions. Availability of technical possibilities and supervisors who can be approached for feedback are both essential. Educators indicate that video-recording depends on initiative by the resident. Many residents describe resistance and shame prior to video-taping and by analyzing the recordings. Installing of the cameras is also mentioned as a drawback. However, the lessons learnt and the acquired self-esteem are mentioned as rewarding and worth the trouble. Most experience with video-taping and feedback took place in the first three years of training.

Attending other residents was mentioned by 19% of the respondents of the questionnaire, but by none of the residents approached for the in-depth interview. Therefore no information can be given about the perceived effect and underlying mechanisms.

Specific trainings were mentioned by 17% of the residents in the questionnaire. In the in-depth interviews all five experiences were indicated as successful and leading to structural changes in communication skills. The trainings mentioned were regional education for rheumatology in small groups from two different regions, a training dedicated on medically unexplained physical symptoms (MUPS) and two trainings dedicated to train teaching skills. Reasons for success mentioned during the interview were the possibility to exchange experiences, approaches and phrasing in specific situation, with difficult real-life situations as a starting point. Other factors for success were observation of role play and gaining awareness of your own pitfalls by giving feedback to others.

Role play with training actors was not explicitly mentioned as an option in the questionnaire provided to the residents, but was referred to as highly valuable by three residents in the in-depth interviews. Factors that contribute to learning are small groups, the quality of training actors, quality of the feedback and the quality of the teacher. A safe learning environment is essential. It helps when there is a clear learning question in advance. It also helps to experience the effect of different communication strategies. It offers a save situation to try different strategies that would not be possible in daily practice. Several residents state that they have changed their approach in daily practice on a long-term basis. Drawback might be that residents do not feel confident to act in the way they would do facing a real patient.

Supervision on paper is mentioned by educators as most frequently used to train/improve communication skills. Educators (2 out of 4) actively question residents about their approach to discuss certain aspects, for example treatment options and tapering medication. One of them indicated that depending on the resident a clear progress in communication skills and the ability to address certain subjects with patients can be observed during a stage of six months.

Group discussions are frequently mentioned by educators. They stimulate discussion in small groups with rheumatologists and residents to discuss difficult cases and to exchange experiences. One region describes summer classes, where one educator discusses difficult situations with residents in order to share experiences and learn from each other.

Scoping literature search

Initial literature search in Pubmed resulted in 1317 articles. When reviewing the literature, a total of 18 articles was selected describing postgraduate residency training of communication skills. Additionally, 15 relevant articles were retrieved by reviewing reference lists. Additional search in other databases: ERIC, SCOPUS and Psycnet resulted in 30 additional articles. See figure 2 for a flowchart.

Evidence for effectiveness of communication training programs varied[16 24-33]. An effective training program should include twelve key elements, which can be categorized into three main groups: 1) Awareness and reflection, 2) Deliberate practice and 3) Environment. Table 1 represents these essential elements categorized into the three main groups.

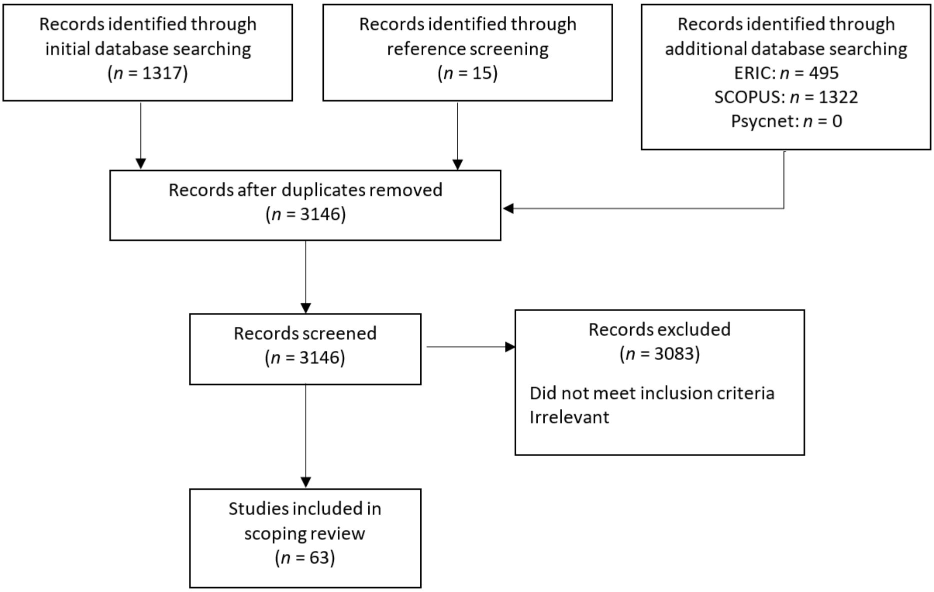

Figure 2. flow chart of study inclusion for scoping review

Table 1. Key elements for effective communication training categorized in three main groups

Key elements from selected articles	Themes	Main groups
Personalized well-defined learning goals [27 34 35]	Awareness	Awareness and reflection
Confrontation with and conscious of own behavior[14 36-39]		
Debriefing and reflection[37 38 40-45]	Reflection	
Training and video examples[45-57]		
Role play/simulation[16 28 36 38 39 45 47 49-51 55 58-62]	Trial and error	Deliberate practice
Sufficient time spent on repeated experiential training and practicing skills[15 16 27 28 32 35-37 43 45-47 58 62-68]		
Deliberate practice[17 27 40 43 60 69 70]		
Personalized, informative, structured, constructive feedback by supervisor, peers, self or patients[16 17 27 28 34 36-41 43 46 53 55 57-64 71-76]	Feedback	
Rich clinical context with opportunities for immediate feedback, reflection and corrections[15 32 36 39 62 70 75 77]	Environment	Environment
Working environment supports teaching and learning of communication for staff and residents[14 17 35 67 75 77]		
Safety[14 38 62]	Staff	
Trained and competent facilitator/teacher[17 27 34 35 39 41 46 55 62]		

Discussion

This study shows that workplace learning is the cornerstone for the post-graduate training of communications skills amongst rheumatologists in the Netherlands, comparable to postgraduate medical education in other fields[78]. Role modelling and trial-and-error are the main principles in the training. Most valuable were observation followed by feedback, training with actors and video-on-the-job followed by feedback. Although experienced as most valuable, these activities were extremely rare during specialist training.

Awareness of communication skills and reflection is one of the three main principles in evidence-based training of communication skills. The transformation towards skilled communicator starts with confrontation with (un)desired behavior. Nearly all residents in the interviews who experienced effect of a communication training intervention were aware of a communication problem and had a related learning question prior to the training intervention. The importance of awareness and reflection is mentioned in the five-phase learning process regarding communication training by van den Eertwegh[14]. Methods used that support raising awareness and reflection are video on the job, observation and feedback and training with actors, but these methods are not frequent. These kind of training methods should be implemented on a regular basis, mainly to increase awareness of own behavior and shortcomings. Furthermore, trial-and-error and discussion during supervision on paper might also contribute to increasing awareness. In order to support reflection, the resident must search for alternative approaches. Specific trainings, discussion in small groups, attending rheumatologists or combined consultations might be ideal to gather alternative approaches.

Deliberate practice is the second cornerstone of evidence-based communication training. It is important that sufficient time is spent on repeated experiential training and practicing skills. However, only a fraction of postgraduate training is actually spent on experimenting with different communication techniques. Being aware of a problem as a starting point, experimenting, the observation of the (effect of the) own behavior, 'good' feedback and reflection are described in this study as important aspects leading to sustainably changed communication skills. This describes the essence of postgraduate medical education as a process of deliberate practice. This is mostly experienced during observation followed by feedback, by training with actors and by video-on-the-job followed by feedback. Although experienced as most valuable, these activities were extremely rare during specialist training. This is also in line with other studies. Direct observation and feedback is not structurally implemented in postgraduate training[17 79]. Supervisors tend to focus their feedback on diagnostic and technical skills and provide relatively little feedback on generic competencies such as communication, professionalism and collaboration[80]. The quality of feedback on generic competencies is often limited and mostly focused on time-management and trying to make the patient feel understood rather than listening and really understand[81].

An environment in which the value of communication skills is recognized and support is institutionalized is essential for residency training of communication skills[14 17 18]. Safety and cognitive & emotional space are labelled as overall conditions influencing this learning process[14]. These were also mentioned in this study as relevant. Educators who underline the importance of good communication skills tend to discuss these aspects during supervision. Appropriate feedback from role models is described as very important[14]. Therefore, it is important that supervisors fully understand the importance of good doctor-patient communication and are trained to support residents to transform towards skilled communicators.

This study has several limitations: First, we recognize a potential inclusion bias, whereby residents interested in improving communication skills might be overrepresented in the questionnaire and the interviews. Residents who are unconsciously incompetent might therefore be underrepresented. However, this should not influence outcome on frequency of training methods and their perceived strengths and barriers. A response rate of 38% is underestimated. The Dutch Society for Rheumatology registers residents as aspirant members, but do not have exact information on the status of these members. Therefore, a substantial part of the registered aspirant members is already finished or is still in pre-training. The actual number of residents in training for rheumatology is estimated at 72. This means that the actual response rate is approximately 58% Furthermore, data is regarded representative due to the variation in region and years of training in our purposive sample. Last limitation is the small number of educators that was involved. However, the main focus of the study is the perceived effect by residents. Strengths are the combination of various methods related to literature and the involvement of educators (MD, WvdB), a psychologist (AP), rheumatologists (LK, MH, NB) and a communication specialist (HvV).

Finally, there are several recommendations for future postgraduate communication skills training. First, in order to increase awareness regular (formative) assessments should be implemented, followed by feedback. Video-taping has the advantage of the possibility of self-evaluation and the possibility to provide multi-source feedback. Also, residents should be encouraged to reflect on the verbal and non-verbal communication with their patients and ask patients how they have experienced the consultation and what was the essence of the consultation. Second, in order to improve deliberate practice, feedback on communication techniques based on real-life observations, video-taping or interaction with training actors should be implemented on a regular basis, for example every one-two months. Residents should be stimulated to formulate learning goals in advance. The availability of a supervisor for attendance of the consultation might be challenging. A video-recording might be a solution, although there may be technical and privacy challenges to take care of. There are increasing options for safe videotaping by means of own devices, where videos are stored directly in a protected database. From there on videos can be forwarded to registered supervisors for feedback. These options are very promising, certainly given the necessary precautions due to the COVID-19 pandemic. Finally, more awareness and training should also be given to supervisors, as an environment in which the value of communication skills is recognized and support is institutionalized is essential for residency training of communication skills.

Conclusions

Given the importance of good doctor-patient communication in chronic diseases this deserves full attention in post-graduate education programs. Evidence-based aspects are 1) raising awareness of communication skills and reflection, 2) deliberate practice and 3) a safe and supportive environment. Cornerstones of methods used during residency training are the attendance of the practice of rheumatologists and trial-and-error. Experienced as most valuable were observation with direct feedback, video-on-the-job and training with actors, however these were rarely used during residency training. Recommendations for further training are regular (formative) assessments followed by feedback to raise awareness and to deliberate practice communication skills. Residents should be stimulated to ask feedback from patients and to formulate learning goals. The use of video-taping can be very supportive. Staff should be trained to give adequate feedback on communication aspects. And last but not least, the environment should be safe and stimulating in order to continuously improve communication skills.

References

1. Dures E, Hewlett S, Ambler N, et al. A qualitative study of patients' perspectives on collaboration to support self-management in routine rheumatology consultations. BMC Musculoskeletal Disorders 2016;**17**:129.
2. Dures E, Fraser I, Almeida C, et al. Patients' Perspectives on the Psychological Impact of Inflammatory Arthritis and Meeting the Associated Support Needs: Open-Ended Responses in a Multi-Centre Survey. Musculoskeletal Care 2016:n/a-n/a.
3. Helland Y, Dagfinrud H, Haugen M-I, et al. Patients' Perspectives on Information and Communication About Sexual and Relational Issues in Rheumatology Health Care. Musculoskeletal Care 2017;**15**(2):131-39.
4. Ferguson S, Trupin L, Yazdany J, et al. Who receives contraception counseling when starting new lupus medications? The potential roles of race, ethnicity, disease activity, and quality of communication. Lupus 2016;**25**(1):12-17.
5. Dures E, Hewlett S, Ambler N, et al. Rheumatology clinicians' experiences of brief training and implementation of skills to support patient self-management. BMC Musculoskeletal Disorders 2014;**15**:108-08.
6. Elwyn G, Frosch DL, Kobrin S. Implementing shared decision-making: consider all the consequences. Implement Sci 2016;**11**:114.
7. Nota I, Drossaert CH, Taal E, et al. Patient participation in decisions about disease modifying anti-rheumatic drugs: a cross-sectional survey. BMC Musculoskelet Disord 2014;**15**:333.
8. Brekke M, Hjortdahl P, Kvien TK. Involvement and satisfaction: a Norwegian study of health care among 1,024 patients with rheumatoid arthritis and 1,509 patients with chronic noninflammatory musculoskeletal pain. Arthritis Rheum 2001;**45**(1):8-15.
9. Kjeken I, Dagfinrud H, Mowinckel P, et al. Rheumatology care: Involvement in medical decisions, received information, satisfaction with care, and unmet health care needs in patients with rheumatoid arthritis and ankylosing spondylitis. Arthritis Rheum 2006;**55**(3):394-401.
10. Barton JL, Schmajuk G, Trupin L, et al. Poor knowledge of methotrexate associated with older age and limited English-language proficiency in a diverse rheumatoid arthritis cohort. Arthritis Res Ther 2013;**15**(5):R157.
11. Blalock SJ, DeVellis BM, DeVellis RF, et al. Medication risk communication during rheumatology office visits. Arthritis Care Res (Hoboken) 2015;**67**(2):161-8.
12. Blalock SJ. Medication Risk Communication in Rheumatology: Where Are We and Where Do We Go from Here? Curr Rheumatol Rep 2017;**19**(2):7.
13. Sowden E, Hassan W, Gooden A, et al. Limited end-user knowledge of methotrexate despite patient education: an assessment of rheumatologic preventive practice and effectiveness. J Clin Rheumatol 2012;**18**(3):130-3.
14. van den Eertwegh V, van der Vleuten C, Stalmeijer R, et al. Exploring residents' communication learning process in the workplace: a five-phase model. PLoS One 2015;**10**(5):e0125958.

15. Aspegren K. BEME Guide No. 2: Teaching and learning communication skills in medicine-a review with quality grading of articles. Medical Teacher 1999;**21**(6):563-70.
16. Berkhof M, van Rijssen HJ, Schellart AJ, et al. Effective training strategies for teaching communication skills to physicians: an overview of systematic reviews. Patient Educ Couns 2011;**84**(2):152-62.
17. van den Eertwegh V, van Dalen J, van Dulmen S, et al. Residents' perceived barriers to communication skills learning: comparing two medical working contexts in postgraduate training. Patient Educ Couns 2014;**95**(1):91-7.
18. van der Vleuten C, van den Eertwegh V, Giroldi E. Assessment of communication skills. Patient Educ Couns 2019;**102**(11):2110-13.
19. Ericsson KA. Deliberate practice and acquisition of expert performance: a general overview. Acad Emerg Med 2008;**15**(11):988-94.
20. Sandelowski M. Whatever happened to qualitative description? Res Nurs Health 2000;**23**(4):334-40.
21. Sandelowski M. What's in a name? Qualitative description revisited. Res Nurs Health 2010;**33**(1):77-84.
22. Peters MD, Godfrey CM, Khalil H, et al. Guidance for conducting systematic scoping reviews. Int J Evid Based Healthc 2015;**13**(3):141-6.
23. Tricco AC, Lillie E, Zarin W, et al. PRISMA Extension for Scoping Reviews (PRISMA-ScR): Checklist and Explanation. Ann Intern Med 2018;**169**(7):467-73.
24. Ditton-Phare P, Loughland C, Duvivier R, et al. Communication skills in the training of psychiatrists: A systematic review of current approaches. Aust N Z J Psychiatry 2017;**51**(7):675-92.
25. Kieran K, Jensen NM, Rosenbaum M. See, Do, Teach? A Review of Contemporary Literature and Call to Action for Communication Skills Teaching in Urology. Urology 2018;**114**:33-40.
26. Kramer AW, Düsman H, Tan LH, et al. Acquisition of communication skills in postgraduate training for general practice. Med Educ 2004;**38**(2):158-67.
27. Maatouk-Bürmann B, Ringel N, Spang J, et al. Improving patient-centered communication: Results of a randomized controlled trial. Patient Educ Couns 2016;**99**(1):117-24.
28. Weiland A, Blankenstein AH, Willems MH, et al. Post-graduate education for medical specialists focused on patients with medically unexplained physical symptoms; development of a communication skills training programme. Patient Educ Couns 2013;**92**(3):355-60.
29. Aspegren K, Lønberg-Madsen P. Which basic communication skills in medicine are learnt spontaneously and which need to be taught and trained? Med Teach 2005;**27**(6):539-43.
30. Makoul G. The SEGUE Framework for teaching and assessing communication skills. Patient Educ Couns 2001;**45**(1):23-34.
31. Delvaux N, Merckaert I, Marchal S, et al. Physicians' communication with a cancer patient and a relative: a randomized study assessing the efficacy of consolidation workshops. Cancer 2005;**103**(11):2397-411.
32. van Dalen J, Kerkhofs E, van Knippenberg-Van Den Berg BW, et al. Longitudinal and concentrated communication skills programmes: two dutch medical schools compared. Adv Health Sci Educ Theory Pract 2002;**7**(1):29-40.

33. Rao JK, Anderson LA, Inui TS, et al. Communication interventions make a difference in conversations between physicians and patients: a systematic review of the evidence. Med Care 2007;**45**(4):340-9.
34. Stiefel F, de Vries M, Bourquin C. Core components of Communication Skills Training in oncology: A synthesis of the literature contrasted with consensual recommendations. Eur J Cancer Care (Engl) 2018;**27**(4):e12859.
35. Wouda JC, van de Wiel HB. How to attain expertise in clinical communication? Paediatr Respir Rev 2013;**14**(4):213-8.
36. Barker M, Fejzic J, Mak AS. Simulated Learning for Generic Communication Competency Development: A Case Study of Australian Post-Graduate Pharmacy Students.1109-23.
37. Bhagat PR, Prajapati KM, Bhatt RB, et al. Development and introduction of a communication skills module for postgraduate students of ophthalmology. Indian Journal of Ophthalmology 2019;**67**(11):1810-15.
38. Gough JK, Frydenberg AR, Donath SK, et al. Simulated parents: Developing paediatric trainees' skills in giving bad news. Journal of Paediatrics and Child Health 2009;**45**(3):133-38.
39. Kowalski C, Sathanandan S. The use of simulation to develop advanced communication skills relevant to psychiatry. BMJ Simulation and Technology Enhanced Learning 2015;**1**(1):29-32.
40. Beaudoin PL, Labbe M, Fanous A, et al. Teaching communication skills to OTL-HNS residents: multisource feedback and simulated scenarios. J Otolaryngol Head Neck Surg 2019;**48**(1):8.
41. Dohms MC, Collares CF, Tibério IC. Video-based feedback using real consultations for a formative assessment in communication skills. BMC Med Educ 2020;**20**(1):57.
42. Stigt JA, Koele JH, Brand PLP, et al. Workplace mentoring of residents in generic competencies by an independent coach. Perspectives on Medical Education 2018;**7**(5):337-41.
43. Ericsson KA. Deliberate practice and the acquisition and maintenance of expert performance in medicine and related domains. Acad Med 2004;**79**(10 Suppl):S70-81.
44. Balslev T, de Grave W, Muijtjens AMM, et al. The Development of Shared Cognition in Paediatric Residents Analysing a Patient Video versus a Paper Patient Case.557-65.
45. Borghi L, Meyer EC, Vegni E, et al. Twelve years of the italian program to enhance relational and communication skills (Percs). International Journal of Environmental Research and Public Health 2021;**18**(2):1-14.
46. MacDonald-Wicks L, Levett-Jones T. Effective teaching of communication to health professional undergraduate and postgraduate students: A Systematic Review. JBI Libr Syst Rev 2012;**10**(28 Suppl):1-12.
47. Arora S, Hull L, Fitzpatrick M, et al. Crisis Management on Surgical Wards: A Simulation-based Approach to Enhancing Technical, Teamwork, and Patient Interaction Skills. Annals of Surgery 2015;**261**(5):888-93.
48. Boissy A, Windover AK, Bokar D, et al. Communication Skills Training for Physicians Improves Patient Satisfaction. Journal of General Internal Medicine 2016;**31**(7):755-61.

49. Cannone D, Atlas M, Fornari A, et al. Delivering Challenging News: An Illness-Trajectory Communication Curriculum for Multispecialty Oncology Residents and Fellows. MedEdPORTAL : the journal of teaching and learning resources 2019;**15**:10819.
50. Downar J, Knickle K, Granton JT, et al. Using standardized family members to teach communication skills and ethical principles to critical care trainees. Critical Care Medicine 2012;**40**(6):1814-19.
51. Fiorentino M, Mosenthal AC, Bryczkowski S, et al. Teaching Residents Communication Skills around Death and Dying in the Trauma Bay. Journal of Palliative Medicine 2021;**24**(1):77-82.
52. Hoffmann TC, Bennett S, Tomsett C, et al. Brief training of student clinicians in shared decision making: A single-blind randomized controlled trial. Journal of General Internal Medicine 2014;**29**(6):844-49.
53. Lozano P, McPhillips HA, Hartzler B, et al. Randomized trial of teaching brief motivational interviewing to pediatric trainees to promote healthy behaviors in families. Archives of Pediatrics and Adolescent Medicine 2010;**164**(6):561-66.
54. Patki A, Puscas L. A Video-Based Module for Teaching Communication Skills to Otolaryngology Residents. Journal of Surgical Education 2015;**72**(6):1090-94.
55. Raper SE, Resnick AS, Morris JB. Simulated disclosure of a medical error by residents: Development of a course in specific communication skills. Journal of Surgical Education 2014;**71**(6):e116-e26.
56. Riess H, Kelley JM, Bailey RW, et al. Empathy training for resident physicians: A randomized controlled trial of a neuroscience-informed curriculum. Journal of General Internal Medicine 2012;**27**(10):1280-86.
57. Sekar DR, Siropaides CH, Smith LN, et al. Adapting Existing Resources for Serious Illness Communication Skills Training for Internal Medicine Residents. Southern medical journal 2021;**114**(5):283-87.
58. Dave S. Simulation in psychiatric teaching. Advances in Psychiatric Treatment 2012;**18**(4):292-98.
59. Gillis AE, Morris MC, Ridgway PF. Communication skills assessment in the final postgraduate years to established practice: A systematic review. Postgraduate Medical Journal 2015;**91**(1071):13-21.
60. Nakagawa S, Fischkoff K, Berlin A, et al. Communication Skills Training for General Surgery Residents. Journal of Surgical Education 2019;**76**(5):1223-30.
61. Nikendei C, Bosse HM, Hoffmann K, et al. Outcome of parent-physician communication skills training for pediatric residents. Patient Education and Counseling 2011;**82**(1):94-99.
62. Subramanian P, Sathanandan K. Improving communication skills using simulation training. British Journal of Medical Practitioners 2016;**9**(2).
63. Dacre J, Richardson J, Noble L, et al. Communication skills training in postgraduate medicine: the development of a new course. Postgrad Med J 2004;**80**(950):711-5.
64. Stigt JA, Koele JH, Brand PLP, et al. Workplace mentoring of residents in generic competencies by an independent coach. Perspect Med Educ 2018;**7**(5):337-41.

65. Barth J, Lannen P. Efficacy of communication skills training courses in oncology: a systematic review and meta-analysis. Ann Oncol 2011;**22**(5):1030-40.
66. Razavi D, Merckaert I, Marchal S, et al. How to optimize physicians' communication skills in cancer care: results of a randomized study assessing the usefulness of posttraining consolidation workshops. J Clin Oncol 2003;**21**(16):3141-9.
67. Bylund CL, Banerjee SC, Bialer PA, et al. A rigorous evaluation of an institutionally-based communication skills program for post-graduate oncology trainees. Patient Education and Counseling 2018;**101**(11):1924-33.
68. Keen A. Communication skills and postgraduate medical training programs. Journal of Postgraduate Medicine 2005;**51**(2):90-91.
69. Brand PL, Stiggelbout AM. Effective follow-up consultations: the importance of patient-centered communication and shared decision making. Paediatr Respir Rev 2013;**14**(4):224-8.
70. Wouda JC, van de Wiel HB. The communication competency of medical students, residents and consultants. Patient Educ Couns 2012;**86**(1):57-62.
71. Junod Perron N, Nendaz M, Louis-Simonet M, et al. Impact of postgraduate training on communication skills teaching: a controlled study. BMC Med Educ 2014;**14**:80.
72. Hulsman RL, van der Vloodt J. Self-evaluation and peer-feedback of medical students' communication skills using a web-based video annotation system. Exploring content and specificity. Patient Educ Couns 2015;**98**(3):356-63.
73. Greco M, Brownlea A, McGovern J. Impact of patient feedback on the interpersonal skills of general practice registrars: results of a longitudinal study. Med Educ 2001;**35**(8):748-56.
74. Brinkman WB, Geraghty SR, Lanphear BP, et al. Effect of multisource feedback on resident communication skills and professionalism: a randomized controlled trial. Arch Pediatr Adolesc Med 2007;**161**(1):44-9.
75. Newcomb AB, Trickey AW, Porrey M, et al. Talk the Talk: Implementing a Communication Curriculum for Surgical Residents. Journal of Surgical Education 2017;**74**(2):319-28.
76. O'Shaughnessy SM. Peer teaching as a means of enhancing communication skills in anaesthesia training: trainee perspectives. Irish Journal of Medical Science 2018;**187**(1):207-13.
77. Essers G, van Dulmen S, van Es J, et al. Context factors in consultations of general practitioner trainees and their impact on communication assessment in the authentic setting. Patient Educ Couns 2013;**93**(3):567-72.
78. Dornan T. Workplace learning. Perspect Med Educ 2012;**1**(1):15-23.
79. Mendick N, Young B, Holcombe C, et al. How do surgeons think they learn about communication? A qualitative study. Med Educ 2015;**49**(4):408-16.
80. Renting N, Gans RO, Borleffs JC, et al. A feedback system in residency to evaluate CanMEDS roles and provide high-quality feedback: Exploring its application. Med Teach 2016;**38**(7):738-45.
81. Renting N, Dornan T, Gans RO, et al. What supervisors say in their feedback: construction of CanMEDS roles in workplace settings. Adv Health Sci Educ Theory Pract 2016;**21**(2):375-87.

Appendix I

1. Contact information and center of education

Name
Year of education
Centre of education
Former education
E-mail address

17. What have you done so far to improve your communication skills? (multiple options possible)

Interventions to train communication skills (Please select, multiple possible)
Multidisciplinary education on communication
Video on the job; feedback by medical specialist or psychologist
Direct observation of patient contact and feedback by medical specialist
Attending doctor-patient interaction of other residents rheumatology
Attending doctor-patient interaction of various rheumatologists
Trial-and-error
Reading of patient information
Specific trainings, please specify

General remark: only the relevant questions as used for this manuscript is shown here. The complete survey can be found in Appendix I chapter 3.

Appendix 2 – script semi-structured interviews

Interview residents

- What kind of situation, experience or intervention in your current training contributed the most to your skills in doctor-patient communication? Did this experience change the way you work and communicate as a doctor? Is you experienced a change, can you explain why?
- Residents indicated in the questionnaire which interventions they have tried to improve communication skills. Residents are asked for each of these interventions whether they worked and to evaluate how or why they worked or not.
- What would you like to change regarding the current curriculum for rheumatologists in training to improve communication skills?

Interview educators

- Which techniques are used to train communication skills for residents? Educators are asked whether these techniques work and why or how they work.
- Which techniques are used to train skills for Shared Decision Making amongst residents? Educators are asked whether these techniques work and why or how they work.
- Do you use ICT tools / feedback tools to enhance learning of communication skills?
- Educators are asked whether these tools work and why or how they work.
- Do you use video-taping of doctor-patient encounters for feedback? Educators are asked whether this works and why or how they work.
- What would you like to change regarding the current curriculum for rheumatologists in training to improve communication skills?

CHAPTER 3

3

Shared Decision Making amongst residents rheumatology: perceived competences, needs and barriers

Laura JC Kranenburg | Mary EW Dankbaar | Natalja Basoski | Haske van Veenendaal | Walter W van den Broek | Johanna MW Hazes

Under review

Abstract

Objectives

Specifically the field of rheumatology endorses Shared Decision Making (SDM). Despite good intentions, realization in daily practice is disappointing. The basis for shared care is made during residency training. How do residents rheumatology use SDM and which barriers they perceive?

Methods

Mixed method study, combining literature search for barriers with quantitative and qualitative data collected from an online questionnaire amongst residents rheumatology who were registered as such in January 2018 by the Dutch Society of Rheumatology.

Results

The opinion on SDM amongst residents rheumatology in the Netherlands is positive but residents' experience with SDM is limited. Identified barriers in the implementation of SDM in daily practice are 1) Unclearness of concept, 2) Lack of ability and skills, 3) Misjudgment of patient preferences, 4) No recognition that SDM is not applied and 5) Uncertainty in treatment decision and outcome.

Conclusion

Residents rheumatology underline the importance of SDM. However, experience and knowledge of SDM is limited and there is limited consultation of patient preferences.

Practice implications

We believe more training and support in SDM is needed. More attention should be paid to SDM during resident training. Ideally there are regular moments of observation and structured feedback.

KEY WORDS

Shared Decision Making, postgraduate communication training, Rheumatology, RA, PSA, SDM

Introduction

The role of a medical specialists changes to that of a medical counselor, who guides patients by providing the best medical care adjusted to patients' values and preferences. That is the essence of Patient Centered Care, where Shared Decision Making (SDM) plays the main role. SDM allows patients and healthcare professionals to jointly make a decision based on the best available evidence for treatment options while respecting patient's values and preferences.[1-3]

Specifically the field of Rheumatology endorses SDM. Rheumatic diseases influence daily life in many ways on functioning, work, social contacts and sexuality.[4-7] Furthermore, patient involvement is associated with better health outcomes, such as health status, self-management, adherence, coping behavior and satisfaction with care.[8-10] The urge for a central role of SDM is supported by patients[11-13] and international guidelines.[14-17] Barriers in the implementation of SDM in the field of Rheumatology are time constraints, the incompatibility with clinical practice guidelines and beliefs that patients are not able to take an active role and do not want SDM.[18]

As the basis for proficiency in shared care and patient participation is made during residency training, an interesting question is how residents rheumatology are trained for SDM. The aim of this study is 1) To explore opinion on SDM amongst residents rheumatology in the Netherlands, 2) To explore residents' experience with SDM, 3) To get an idea of how they self-assess their skills on communication and SDM, 4) Finally to identify barriers in the implementation of SDM.

Methods

This mixed method study combines quantitative and qualitative analyses involving an online questionnaire and literature study. For this purpose an online questionnaire was developed, see appendix I. In the development of the questionnaire a SDM specialist (HvV) and a rheumatologist with experience in implementing SDM (LK, NB) were consulted for content and face validity.

In order to explore the opinion on SDM the CanMEDS framework was used to gain insight how residents value doctor-patient communication in general. [19] Residents were asked to organize these roles in order of relative importance (see question 2 appendix I). Qualitative data on residents' view on SDM was gathered by an open question, see question 12 appendix I.

In order to explore the experience with SDM in daily practice quantitative information was gathered by asking residents which aspects they communicate when discussing a new diagnosis or treatment, see question 8 and 10 appendix I. Options in the list were selected from various studies on doctor-patient communication[20-24] and from national initiatives on patient information and decision aids.[252627] Residents were asked which methods they use to optimize their doctor-patient communication (see question 11 appendix I). Qualitative information about residents' experience with SDM was collected by the use of an open question (see question 12 appendix I).

In order to evaluate self-assessment of communication skills and SDM skills quantitative data on general communication skills was collected by asking residents to self- assess their communication skills on a five-point scale, see questions 3 to 6, appendix I. Quantitative data on SDM skills was collected by asking residents to assess their ability to deal with the four steps of SDM[28] on a visual analog scale from 0 to 100. See questions 13-16, appendix I. Quantitative data on skills to discuss various rheumatologic diseases and treatment options with patients were collected by asking residents to rate their competence on a five-point scale. See questions 7 and 9, appendix I.

In order to explore barriers in the implementation of SDM in daily practice literature study was performed in order to identify known barriers for healthcare providers to implement SDM. Publications were screened for barriers in the implementation of SDM on the level of the health care provider. Potential barriers are represented in table 1, more detailed information based on the literature are presented in appendix II.

Table 1. Barriers identified in literature.

Barriers identified in literature
Potential negative attitude towards SDM
Unclearness of concept and lack of knowledge on how to apply SDM
Lack of ability and skills related to SDM, poor physician communication
Dealing with different patient characteristics, misjudgment of patient preferences
No recognition that Shared Decision Making is not applied ('we are already doing it')
Uncertainty in the treatment decision and concern regarding adverse effects
Time constraints

Aspirant members of the Dutch Society of Rheumatology were invited to answer the online questionnaire. Anonymous participation was possible. The addressed group is mixed: residents in their pre-training (year 1-3), specialty training for rheumatology (year 4-6) and rheumatologists who just finished their training. Qualitative data was analyzed by thematic content analysis[29 30]. Themes were identified by the relative frequency of aspects mentioned in the answers to the open questions. All provided information was reported.

Data was related to identified barriers in the use of SDM amongst medical specialists as found in literature.

Results

Forty-two of the 111 (38%) aspirant members responded. All eight regions in the Netherlands were represented. The male/female ratio amongst participants was higher than amongst the registered aspirant members (10/90% versus 20/80%). This difference is not significant. See table 2.

Table 2. Characteristics of respondents (n=42)

Parameter	respondents n=42 (%)
Complete responders	37 (88)
Year 1-3 (pre-training)	4 (10)
Year 4	15 (36)
Year 5	13 (31)
Year 6	9 (21)
Years of training unknown	1 (2)
Male responders	4 (10)
Female responders	35 (83)
Sex not reported	3 (7)

Residents rate the CanMED role of Communicator as second important after the role of Medical Expert, see figure 1. Residents report that doctor-patient communication in general and more specific SDM is important. The possibility to improve adherence to therapy and the doctor-patient relationship was mentioned as an advantage. They state that SDM is indispensable within the field of rheumatology and should be obligatory. SDM is referred to as an ethical imperative. It is stated that it is necessary to investigate patients' values and preferences related to therapy. Two residents indicate that they try to influence the decision to what in their opinion is medically right. One participant mentions that patients are often satisfied anyway, with or without SDM.

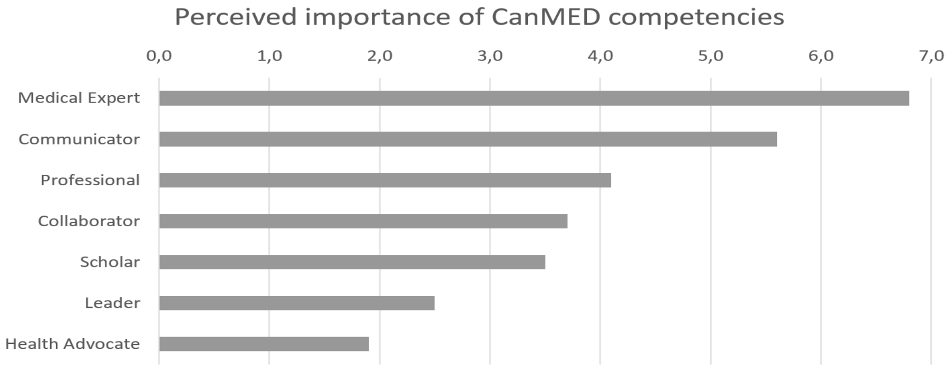

Figure 1. Perceived importance of CanMEDS competencies as rated by residents. Higher scores imply higher perceived importance.

Experience with SDM in daily practice is rare and there is a plea for more training. Discussing patient's preferences is reported only by 33% of the residents on a regular basis. Most discussed items are explanation of diagnosis, pharmaceutical treatment, potential treatment effect end possible side effects. Other aspects, e.g. non-pharmaceutical treatment options are clearly less frequently discussed (appendix III , figure 1).

To support SDM and communication in general, residents use various tools and methods (see table 3). Decision aids are rarely used. Qualitative analysis shows that less than a third of the residents indicate that they use SDM on a daily basis (10 out of 37). A need is clearly expressed for more education, regardless of the level of experience (20 out of 37). Residents are mainly interested in feedback about their own performance on SDM and concrete and practical advice on how to apply SDM in daily practice. Residents report more difficulties with SDM when patients are newly diagnosed. They also indicate that SDM is more difficult with patients with rare diseases.

Self-assessment of skills for communication and SDM is overall average to good. Median rating for "Effective relationship" and "Listening and information gathering" is 4 on a 5 point scale. Median rating for "Provision of information to patients and family" and for "adequate oral report" is slightly lower (Median scores 4 and 3). The perceived ability is not higher for residents with more training (table 4).

Median self-ratings for performance on the four steps of SDM vary between 66 to 73 on a scale from 0-100, see figure 2. Self-reported skills for communication different diagnosis and treatment options increases for the reported years of training (appendix III, figures 2 and 3). Residents experience more difficulty in doctor-patient communication for rare diseases (appendix III, figure 2) and for treatment options that are less frequently used (appendix III, figure 3).

Table 3. Tools used to support doctor – patient communication and SDM. Resident could select multiple options from a pre-defined list. Last option provided the possibiliy to enter other suggestions.

Possible tools to support doctor – patient communication	respondents n=42 (%)
Printed information / folders	38 (90%)
Information and counseling by specialized nurse	35 (83%)
Structuring and prioritizing of information	25 (60%)
Drawing /sketches	22 (52%)
Referral to patient groups and patient societies	15 (36%)
Providing percentages and changes and examples	13 (31%)
Information dedicated to the patient through website : 'Voorlichting op maat'	6 (14%)
Audiotaping of the consult	3 (7%)
Online decision aid: http://www.reumamedicatiekeuzehulp.nl	2 (5%)
Power point presentations / video	0
Method 'Samen Beslissen: drie goede vragen' (SDM intervention)	0
Other, input from open field: - use of pictures on the internet - referral to good websites (not specified) - website of the Dutch Arthritis Foundation - Stimulating patient to write down questions at home and request for a new appointment when needed to discuss these questions.	

Table 4. Median scores (Q1:Q3) on self-assessment on four aspects of competence communication on a five-point scale (1 = not competent; 5 = very competent) - categorized by years of education. In brackets interquartile range is represented by first and third quartile scores.

	All years (n=41)	Year 1-3 (n=4)	Year 4 (n=15)	Year 5 (n=12)	Year 6 (n=9)	Unknown (n=1)
Effective relationship	4 (4:4)	4 (4:4)	4 (3,5:4)	4 (4:4)	4 (4:4)	3 (-)
Listening and information gathering	4 (4:4)	4 (4:4,25)	4 (3:4)	4 (3,75:4)	4 (4:4)	3 (-)
Provision of information to patients and family	4 (3:4)	3,5 (3:4,25)	4 (3:4)	3,5 (3:4)	4 (3:4)	3 (-)
Adequate oral report	3 (3:4)	3 (3:3,25)	3 (3:4)	3 (3:4)	4 (4:4)	3 (-)

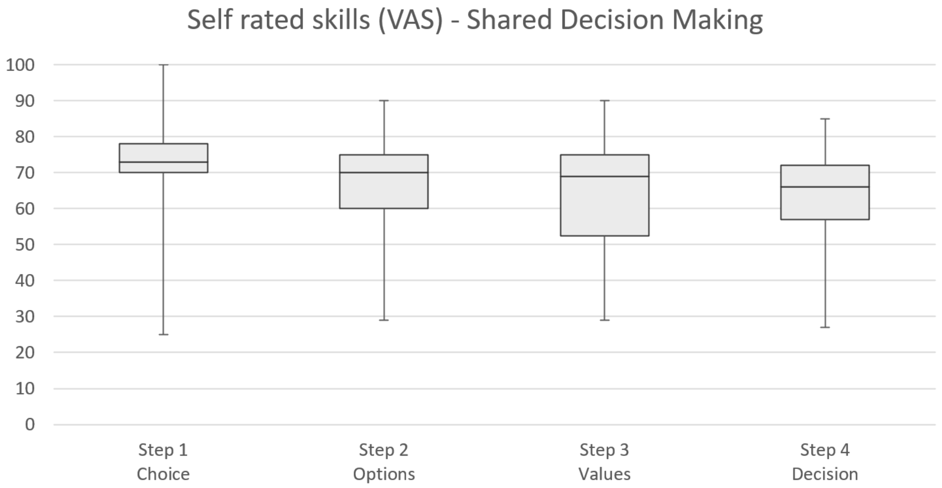

Figure 2. Self rated skills on a Visual Analog Scale for the four steps for Shared Decision Making

There are several barriers described for in the implementation of SDM, see table 1. First, a "potential negative attitude towards SDM" was not mentioned by respondents. Secondly, "Unclearness of concept" was recognized; residents indicate that the concept of SDM is rather vague and they are insecure about their own performance in practice. Thirdly, "Lack of ability and skills" was reported; residents find it difficult to determine whether they apply SDM correctly. They experience difficulty in explaining all options. They state that patients seem to have problems in understanding options and find it difficult to make a decision. Supportive tools such as Patient decision aids (PtDAs) are rarely used to support SDM. Fourthly, there is "Misjudgment of patient preferences". Residents describe difficulties recognizing which patients want to make a Shared Decision. Many mention that some patients might not be fit for SDM. Hereby respondents do not report any difficulty inquiring about patients' preferences in decision making and treatment options. Fifth, there is the barrier "We are already doing it", illustrated by the self-assessment on the four steps of SDM. A sixth barrier is "Uncertainty in treatment decision". Residents report difficulty in the use of SDM in rare diseases and self-reported skills are lower than for other diseases. Finally, the barrier "Time constraints" was only mentioned once.

Discussion

This study shows that residents rheumatology in the Netherlands value the concept of Shared Decision Making, but that their experience is limited. Self-assessment of general communication skills and SDM are average to good. Identified barriers in the implementation of SDM in daily practice are 1) Unclearness of concept, 2) Lack of ability and skills, 3) Misjudgment of patient preferences, 4) No recognition that SDM is not applied and 5) Uncertainty in treatment decision and outcome.

Residents argue that SDM is highly relevant in the field of Rheumatology by providing good information about treatment options and to ask about patient's needs. This is in line with the results found by Mathijssen[18] regarding healthcare professionals' perspectives of SDM in rheumatology. Residents indicate that the concept of SDM is rather vague. They believe not all patients are fit for SDM. The question arises, whether patients are not up to SDM or residents lack the necessary skills.[18] More vulnerable patient populations report less interest in SDM, while in particular these vulnerable groups might benefit from a decision which matches their personal situation and preferences.[31 32] Residents also experience difficulties with rare diseases and treatment options, where evidence lacks due to small patient numbers. [33 34] However, especially in these situations with limited evidence, patients' preferences should play a key role in the decision. This pleas for training how to address SDM for different patient groups.

Residents indicate that they try to implement SDM into daily practice, but plea for feedback and training. The reported self-assessment for SDM skills are overall moderate to good. Overrating of performance on SDM is a known problem.[18] These results may indicate over-confidence in these skills. Feedback on actual performance may lead to awareness on shortfalls, which is the starting point for learning and improvement. The most important enhancing factors in post-graduate communication training are encouragement to deliberately practice in an environment in which the value of communication skills is recognized and support is institutionalized with appropriate feedback from role models.[35-37] Supportive tools such as Patients Decision Aids (PtDAs) are rarely used and should get more attention, as PtDAs improve decision quality [38]. PtDAs help patients to understand their involvement in choosing and to communicate their values to their practitioners.[39]

It is striking that participants claim it is difficult to explain options, but do not mention difficulty in identifying patients' preferences. It is possible that there are no difficulties in assessing patients' preferences, but it is more likely that these are misjudged. Patients' preferences are not discussed on a regular basis. Misjudgment of patient preferences is a barrier in the implementation of SDM.[31 32 40-42]

Conclusion

Important conclusions are that residents underline the importance of SDM. However, experience and knowledge of SDM is limited and there is limited consultation of patient preferences. Important barrier in the implementation of SDM is unclearness of concept and lack of training. Residents are often not aware that they fail to identify patient preferences and lack tools to implement SDM for certain groups. Specific attention should be paid for Shared Decision Making with patients who want to leave the decision up to the doctor and in Shared Decision Making regarding rare diseases.

Practice implications

We believe more training and support in SDM and the use of decision aids is needed during resident training. Ideally there are regular moments of observation and structured feedback. Specific attention should be paid to patient groups that need support in decision making and rare diseases.

References

1. Makoul G, Clayman ML. An integrative model of shared decision making in medical encounters. Patient Educ Couns 2006;**60**(3):301-12.
2. Stiggelbout AM, Van der Weijden T, De Wit MP, et al. Shared decision making: really putting patients at the centre of healthcare. Bmj 2012;**344**:e256.
3. Elwyn G, Frosch D, Thomson R, et al. Shared decision making: a model for clinical practice. J Gen Intern Med 2012;**27**(10):1361-7.
4. Dures E, Hewlett S, Ambler N, et al. A qualitative study of patients' perspectives on collaboration to support self-management in routine rheumatology consultations. BMC Musculoskeletal Disorders 2016;**17**:129.
5. Dures E, Fraser I, Almeida C, et al. Patients' Perspectives on the Psychological Impact of Inflammatory Arthritis and Meeting the Associated Support Needs: Open-Ended Responses in a Multi-Centre Survey. Musculoskeletal Care 2016:n/a-n/a.
6. Helland Y, Dagfinrud H, Haugen M-I, et al. Patients' Perspectives on Information and Communication About Sexual and Relational Issues in Rheumatology Health Care. Musculoskeletal Care 2017;**15**(2):131-39.
7. Ferguson S, Trupin L, Yazdany J, et al. Who receives contraception counseling when starting new lupus medications? The potential roles of race, ethnicity, disease activity, and quality of communication. Lupus 2016;**25**(1):12-17.
8. Kjeken I, Dagfinrud H, Mowinckel P, et al. Rheumatology care: Involvement in medical decisions, received information, satisfaction with care, and unmet health care needs in patients with rheumatoid arthritis and ankylosing spondylitis. Arthritis Rheum 2006;**55**(3):394-401.
9. Nota I, Drossaert CH, Taal E, et al. Patient participation in decisions about disease modifying anti-rheumatic drugs: a cross-sectional survey. BMC Musculoskelet Disord 2014;**15**:333.
10. Brekke M, Hjortdahl P, Kvien TK. Involvement and satisfaction: a Norwegian study of health care among 1,024 patients with rheumatoid arthritis and 1,509 patients with chronic noninflammatory musculoskeletal pain. Arthritis Rheum 2001;**45**(1):8-15.
11. Dures E, Hewlett S, Ambler N, et al. Rheumatology clinicians' experiences of brief training and implementation of skills to support patient self-management. BMC Musculoskeletal Disorders 2014;**15**:108-08.
12. Farinha F, Freitas F, Agueda A, et al. Concerns of patients with systemic lupus erythematosus and adherence to therapy - a qualitative study. Patient Prefer Adherence 2017;**11**:1213-19.
13. Garrido-Cumbrera M, Hillmann O, Mahapatra R, et al. Improving the Management of Psoriatic Arthritis and Axial Spondyloarthritis: Roundtable Discussions with Healthcare Professionals and Patients. Rheumatol Ther 2017.
14. Smolen JS, Aletaha D, Bijlsma JWJ, et al. Treating rheumatoid arthritis to target: recommendations of an international task force. Annals of the Rheumatic Diseases 2010;**69**(4):631-37.

15. Smolen JS, Landewé R, Breedveld FC, et al. EULAR recommendations for the management of rheumatoid arthritis with synthetic and biological disease-modifying antirheumatic drugs: 2013 update. Annals of the Rheumatic Diseases 2014;**73**(3):492-509.
16. Hochberg MC, Altman RD, April KT, et al. American College of Rheumatology 2012 recommendations for the use of nonpharmacologic and pharmacologic therapies in osteoarthritis of the hand, hip, and knee. Arthritis Care & Research 2012;**64**(4):465-74.
17. Gossec L, Smolen JS, Ramiro S, et al. European League Against Rheumatism (EULAR) recommendations for the management of psoriatic arthritis with pharmacological therapies: 2015 update. Ann Rheum Dis 2016;**75**(3):499-510.
18. Mathijssen EGE, van den Bemt BJF, Wielsma S, et al. Exploring healthcare professionals' knowledge, attitudes and experiences of shared decision making in rheumatology. RMD Open 2020;**6**(1).
19. Frank JR, Danoff D. The CanMEDS initiative: implementing an outcomes-based framework of physician competencies. Med Teach 2007;**29**(7):642-7.
20. Blalock SJ, DeVellis BM, DeVellis RF, et al. Medication risk communication during rheumatology office visits. Arthritis Care Res (Hoboken) 2015;**67**(2):161-8.
21. Blalock SJ, Slota C, Devellis BM, et al. Patient-rheumatologist communication concerning prescription medications: getting to the gist. Arthritis Care Res (Hoboken) 2014;**66**(4):542-50.
22. Blalock SJ, DeVellis RF, Chewning B, et al. Gist and verbatim communication concerning medication risks/benefits. Patient Educ Couns 2016;**99**(6):988-94.
23. Schwartz LM, Woloshin S. The Drug Facts Box: Improving the communication of prescription drug information. Proc Natl Acad Sci U S A 2013;**110 Suppl 3**:14069-74.
24. Schwartz LM, Woloshin S, Welch HG. The drug facts box: providing consumers with simple tabular data on drug benefit and harm. Med Decis Making 2007;**27**(5):655-62.
25. http://www.reumamedicatiekeuzehulp.nl/home
26. https://voorlichtingopmaat.erasmusmc.nl/login
27. https://3goedevragen.nl/
28. Stiggelbout AM, Pieterse AH, De Haes JC. Shared decision making: Concepts, evidence, and practice. Patient Educ Couns 2015;**98**(10):1172-9.
29. Sandelowski M. Whatever happened to qualitative description? Res Nurs Health 2000;**23**(4):334-40.
30. Sandelowski M. What's in a name? Qualitative description revisited. Res Nurs Health 2010;**33**(1):77-84.
31. Legare F, Ratte S, Gravel K, et al. Barriers and facilitators to implementing shared decision-making in clinical practice: update of a systematic review of health professionals' perceptions. Patient Educ Couns 2008;**73**(3):526-35.
32. Légaré F, Witteman HO. Shared decision making: examining key elements and barriers to adoption into routine clinical practice. Health Aff (Millwood) 2013;**32**(2):276-84.

33. de Snoo-Trimp JC, Brom L, Pasman HR, et al. [Perspectives of medical specialists on sharing decisions in cancer care: a qualitative study concerning chemotherapy decisions with patients with recurrent glioblastoma]

Wel of geen kankerbehandeling in de resterende tijd? Specialisten over gedeelde besluitvorming bij recidiverend glioblastoom. Ned Tijdschr Geneeskd 2015;**159**:A9790.

34. Covvey JR, Kamal KM, Gorse EE, et al. Barriers and facilitators to shared decision-making in oncology: a systematic review of the literature. Support Care Cancer 2019;**27**(5):1613-37.
35. van den Eertwegh V, van Dalen J, van Dulmen S, et al. Residents' perceived barriers to communication skills learning: comparing two medical working contexts in postgraduate training. Patient Educ Couns 2014;**95**(1):91-7.
36. van den Eertwegh V, van der Vleuten C, Stalmeijer R, et al. Exploring residents' communication learning process in the workplace: a five-phase model. PLoS One 2015;**10**(5):e0125958.
37. van der Vleuten C, van den Eertwegh V, Giroldi E. Assessment of communication skills. Patient Educ Couns 2019;**102**(11):2110-13.
38. O'Connor AM, Llewellyn-Thomas HA, Flood AB. Modifying unwarranted variations in health care: shared decision making using patient decision aids. Health Aff (Millwood) 2004;**Suppl Variation**:VAR63-72.
39. Elwyn G, O'Connor A, Stacey D, et al. Developing a quality criteria framework for patient decision aids: online international Delphi consensus process. Bmj 2006;**333**(7565):417.
40. van Veenendaal H, van der Weijden T, Ubbink DT, et al. Accelerating implementation of shared decision-making in the Netherlands: An exploratory investigation. Patient Educ Couns 2018;**101**(12):2097-104.
41. Cassidy BP, Getchell LE, Harwood L, et al. Barriers to Education and Shared Decision Making in the Chronic Kidney Disease Population: A Narrative Review. Can J Kidney Health Dis 2018;**5**:2054358118803322.
42. Driever EM, Stiggelbout AM, Brand PLP. Shared decision making: Physicians' preferred role, usual role and their perception of its key components. Patient Educ Couns 2020;**103**(1):77-82.

Appendix I

1. Contact information and center of education

Name
Year of education
Centre of education
Former education
E-mail address

2. Which competence do you rate as most important as a rheumatologist. Rate the competences from most important to least important.

CanMED competence	Rating
Medical Expert	Choose importance from 1 - 7
Communicator	Choose importance from 1 - 7
Collaborator	Choose importance from 1 - 7
Scholar	Choose importance from 1 - 7
Health Advocate	Choose importance from 1 - 7
Leader	Choose importance from 1 - 7
Professional	Choose importance from 1 - 7

Intermezzo
What is your self-evaluation of your competence of communication?
How would you rate your own skills to explain certain diseases and treatment options?
What would you like to learn?

3. To what extent are you capable to build an effective doctor-patient relationship?
→ Self-evaluation on a five-point scale (1 = not competent; 5 = very competent)

4. To what extent are you capable to listen and gather information?
→ Self-evaluation on a five-point scale (1 = not competent; 5 = very competent)

5. To what extent are you capable to provide information to patients and family?
→ Self-evaluation on a five-point scale (1 = not competent; 5 = very competent)

6. To what extent are you capable to give an adequate oral report?
→ Self-evaluation on a five-point scale (1 = not competent; 5 = very competent)

7. Self-evaluation of communication skills to explain the following diagnoses to patient

Diagnosis	Self-evaluation
Undifferentiated arthritis	All diagnoses are rated on a five-point scale rating the level of expertise of communication skills 1= knowledge of 2= strict supervision 3= limited supervision 4 = unsupervised 5 = level rheumatologist
Reumatoid arthritis	
Spondyloarthritis (axial/perifere)	
Psoriatic arthritis	
Cristal arthritis	
PMR/Arteriitis temporalis	
SLE	
Systemic Sclerosis	
M. Sjögren	
Arthrosis	
Fibromyalgia/ Pain syndrom	
Osteoporosis	
Clinical suspect arthralgia	

8. When a diagnosis is made, how do you communicate these aspects?

	Always	On request	Via nurse	Not at all
Explanation diagnosis				
Pathofysiology				
Prognosis				
Medicamental treatment				
Non-medicamental treatment				
Influence on Pregnancy				
Influence on work				
Influence on sexuality				
Influence on social aspects				
Cardiovascular risc				
Risc for osteoporosis				

9. Self-evaluation of communication skills to explain treatment options to the patient

Diagnosis	Self-evaluation
Corticosteroïds	All treatment options are rated on a five-point scale rating the level of expertise of communication skills 1= knowledge of 2= strict supervision 3= limited supervision 4 = unsupervised 5 = level rheumatologist
NSAIDs	
Conventional DMARDs	
TNF-a blockage	
Anti-IL6, anti-B cel, T-cel blockage	
Anti-IL 17 / 23	
Cyclophosphamid	
Uric acid lowering therapy and profylaxis	
Bisphosphonates, calcium and vitamin D	
Revalidation in fibromyalgia	

10. When you start a treatment, how do you communicate these aspects?

	Always	On request	Via nurse	Not at all
Course of disease without treatment				
Potential treatment effect				
Possible side-effects				
Frequency and severeness of side-effects				
Control of side effects				
Consequences for pregnancy				
Consequences for alcohol				
Alternatives and patient preferences				

11. To what extent do you use the following tools (multiple options possible)

Possible tools to support doctor – patient communication (Please select, multiple possible)
Drawing /sketches
Providing percentages and changes and examples
Structuring and prioritizing of information
Audiotaping of the consult
Power point presentations / video
Information and counseling by specialized nurse
Printed information / folders
Online decision aid: Reuma medicatie keuzehulp http://www.reumamedicatiekeuzehulp.nl
Information dedicated to he patient through website : 'Voorlichting op maat'
Referral to patient groups and patient societies
Method 'Samen Beslissen: drie goede vragen' (Shared Decision Making intervention)
Other (please explain)

12. What is your opinion about Shared Decision Making?
Do you have any experience? Would you like to learn more?
→ Open field question

Intermezzo

To what extent do you feel competent to apply the steps from Shared Decision Making?

Imagine a case where Shared Decision Making is possible and answer the following questions.

Examples of possible cases:
- **A young 19 years old woman with erosive Juvenile Idiopathic Arthritis and uveïtis, acutal medication methotrexate and anti-TNF, no awareness of disease and possible complications and active desire and attempts for pregnancy.**
- **A 80 years old lady with erosive and active Rheumatoïd Arthritis, always treated with local steroïd injections out of fear for DMARD therapy.**
- **Or: a situation in your own daily practice where a choice was made**

13. Step 1. Inform patient that a shared decision has to b e made
Make sure that the patient understands that a decision should be made, that there are multiple options which are possible. Make sure that the setting is right.
→ To what extent on a visual analog scale from 0 to 100 (0= not capable at all 100= vary capable) do you think you are capable to discuss this first step.

14. Step 2. Explain pros and cons of each option
Explain all options (including no action) in a neutral way, explain pros and cons of each option in a concrete manner.
→ To what extent on a visual analog scale from 0 to 100 (0= not capable at all 100= vary capable) do you think you are capable to discuss this second step.

15. Step 3. Discuss patients values
Make an inventory what matters to the patient. What are his/her goals, expectations, preferences, worries? What does this mean for the possible options?
→ To what extent on a visual analog scale from 0 to 100 (0= not capable at all 100= vary capable) do you think you are capable to discuss this third step.

16. Step 4. Discuss shared decision and following steps
Can we *make* a decision? Is more time or information needed?
→ To what extent on a visual analog scale from 0 to 100 (0= not capable at all 100= vary capable) do you think you are capable to discuss this fourth step.

17. What have you done so far to improve your communication skills? (multiple options possible)

Interventions to train communication skills (Please select, multiple possible)
Multidisciplinary education on communication
Video on the job; feedback by medical specialist or psychologist
Direct observation of patient contact and feedback by medical specialist
Attending doctor-patient interaction of other residents rheumatology
Attending doctor-patient interaction of various rheumatologists
Trial-and-error
Reading of patient information
Specific trainings, please specify

18. What would you like to add to the training for rheumatologist? (multiple options possible)

Preferences to enrich training of communication skills in future (Please select, multiple possible)
Video-examples of explanation of certain diseases by colleagues
Video-examples of explanation of pros and cons of medication and possible risks
Video-examples of how to deal with different types of patients
Training in Shared decision making
Training in how to deal with Medically unexplained physical symptoms (MUPS)
Training in how to deal with different types of patients
Training with simulated patients (Actors)
Video-taping of patient doctor contact + feedback
Audio-taping of patient doctor contact + feedback
Feedback by other residents rheumatology
Feedback by various rheumatologists
Feedback by psychologist
Feedback by patients
Other (please explain)

Appendix II – reported barriers into the implementation of SDM in literature

Barriers identified in literature	**Summary from literature**	**Analysis for residents rheumatology**
Potential negative attitude towards SDM	Literature shows that a potential negative attitude towards SDM is a barrier for the implementation of SDM by health care professionals.[1] In the field of chronic kidney disease lack of engagement of the health care professional was a key barrier in the implementation of SDM related to decisions regarding end stage renal disease.[2]	Does not apply; there is a strong wish for SDM
Unclearness of concept and lack of knowledge on how to apply SDM	Literature shows that unclearness of the concept of SDM and lack of knowledge on how to apply SDM can be a barrier in implementation[1]. Legaré [3 4] states that the *clinical situation* can be a major constraint in the implementation of SDM. In certain situations, e.g. vaccination for flu, the preferred decision for healthcare might not reflect patient's preferences, which can influence decision making.	Unclearness of concept is reported; there is a plea for more feedback and training on SDM
Lack of ability and skills related to SDM, poor physician communication	In literature lack of ability and skills how to perform SDM is repeatedly mentioned as a barrier[1 5], for example in decision making in the field of oncology[6] or end stage renal disease[2]. A lack in utilizing effective methods of transferring knowledge to patients was reported. A cross-sectional survey amongst of 785 physicians showed that physicians tend to limit SDM to discussing treatment options and fewer made clear that a decision had to be made or explored the patient's wish how to be involved in decision making[5].	Knowledge on techniques for SDM and decision aids is limited
Dealing with different patient characteristics, misjudgment of patient preferences	Literature identifies misjudgment of patient preferences as an important barrier in the implementation of SDM[1-5]. The severity of the patients' conditions, increasing age, low education and male sex are predictors of a preference for a passive role in the doctor-patient relationship, but these characteristics explain only 20% or less of the variability in preferences[7]. Also, culture and ethnicity have often been cited as barriers in establishing an effective and satisfying doctor–patient relationship[8]. Above this, variables in personality are strongly predictive for preferences in doctor-patient interaction, despite the patients demographic profile[9]. The only way a physician can gain insight into an individual patient's desire to participate in decision making is through direct enquiry [7]. Physicians often revert to a paternalistic approach, despite a positive attitude towards SDM, especially when they judged the patient to be incapable of participating in decision making[5]. *Patient characteristics* are perceived as a barrier for the implementation of SDM, while more vulnerable patient populations report less interest in SDM[3 10-12]. This pleas for other ways to address SDM, while in particular these vulnerable groups might benefit from a decision which matches their personal situation and preferences[3 4].	Difficulty is experienced with certain patient characteristics, assumption that not all patients are fit for SDM

Barriers identified in literature	Summary from literature	Analysis for residents rheumatology
No recognition that Shared Decision Making is not applied ('we are already doing it')	Literature shows that the lack of recognition that SDM is not applied is an important barrier[1,3,13]. A frequently mentioned objection to SDM by physicians is: "But we already do that". However there is a wealth of somewhat depressing evidence that physicians and patients do not communicate well[14]. A qualitative study in oncology showed that although medical specialists considered SDM to be important, they did not adhere to its elements. Stopping treatment was not considered equal to continuing treatment. Exploration of the patients' wishes was done implicitly, and shared responsibility for the decision was not highly recognized. Decisions for starting or continuing treatment were made on assumptions of physicians about knowing what patients want[15]. Despite a positive attitude to SDM, assessment of the level of SDM in routine rheumatology consultations showed rather disappointing results with a mean OPTION score of 28.3 (SD=15.1), higher OPTION scores indicating higher levels of SDM (scale 0-100)[16].	There is a wish for feedback on performance on SDM. Possible lack between self-ratings and actual performance given the high self-ratings.
Uncertainty in the treatment decision and concern regarding adverse effects	A systematic literature review within oncology identified *uncertainty in the treatment decision and the concern regarding adverse effects* as specific barriers in the use of SDM[6]. Treatment in oncology shows similarity with the treatment of rare diseases in rheumatology, as evidence is limited and adverse outcomes might be severe. Specifically in these situation it is important to involve patients' preferences into decision making.	Difficulty is experienced in SDM in rare diseases; self-reported skills for discussing rare diseases and treatment options is less compared to more common variants.
Time constraints	*Literature reports time* as a frequent perceived barrier[3,4,17]. Prior study performed in the field of rheumatology identifying barriers and facilitators showed that time constraints were one of the main concerns[17]. However several studies have shown that doctors trained in some of these communications skills do not take significantly longer to conduct patient interviews.[18-20] Combined results from more than a hundred randomized trials provide no robust evidence that more time is required to engage in SDM in clinical practice than to offer usual care[3,4].	Reported by one respondent

1. van Veenendaal H, van der Weijden T, Ubbink DT, et al. Accelerating implementation of shared decision-making in the Netherlands: An exploratory investigation. Patient Educ Couns 2018;**101**(12):2097-104.
2. Cassidy BP, Getchell LE, Harwood L, et al. Barriers to Education and Shared Decision Making in the Chronic Kidney Disease Population: A Narrative Review. Can J Kidney Health Dis 2018;**5**:2054358118803322.
3. Legare F, Ratte S, Gravel K, et al. Barriers and facilitators to implementing shared decision-making in clinical practice: update of a systematic review of health professionals' perceptions. Patient Educ Couns 2008;**73**(3):526-35.
4. Légaré F, Witteman HO. Shared decision making: examining key elements and barriers to adoption into routine clinical practice. Health Aff (Millwood) 2013;**32**(2):276-84.
5. Driever EM, Stiggelbout AM, Brand PLP. Shared decision making: Physicians' preferred role, usual role and their perception of its key components. Patient Educ Couns 2020;**103**(1):77-82.
6. Covvey JR, Kamal KM, Gorse EE, et al. Barriers and facilitators to shared decision-making in oncology: a systematic review of the literature. Support Care Cancer 2019;**27**(5):1613-37.
7. Benbassat J, Pilpel D, Tidhar M. Patients' preferences for participation in clinical decision making: a review of published surveys. Behav Med 1998;**24**(2):81-8.
8. Schouten BC, Meeuwesen L. Cultural differences in medical communication: A review of the literature. Patient Education and Counseling 2006;**64**(1):21-34.
9. Braman AC, Gomez RG. Patient personality predicts preference for relationships with doctors. Personality and Individual Differences 2004;**37**(4):815-26.
10. Stiggelbout AM, Van der Weijden T, De Wit MP, et al. Shared decision making: really putting patients at the centre of healthcare. Bmj 2012;**344**:e256.
11. Legare F, Stacey D, Turcotte S, et al. Interventions for improving the adoption of shared decision making by healthcare professionals. Cochrane Database Syst Rev 2014(9):Cd006732.
12. Légaré F, Politi MC, Drolet R, et al. Training health professionals in shared decision-making: An international environmental scan. Patient Education and Counseling 2012;**88**(2):159-69.
13. Joseph-Williams N, Lloyd A, Edwards A, et al. Implementing shared decision making in the NHS: lessons from the MAGIC programme. Bmj 2017;**357**:j1744.
14. Towle A, Godolphin W. Framework for teaching and learning informed shared decision making. BMJ : British Medical Journal 1999;**319**(7212):766-71.
15. de Snoo-Trimp JC, Brom L, Pasman HR, et al. [Perspectives of medical specialists on sharing decisions in cancer care: a qualitative study concerning chemotherapy decisions with patients with recurrent glioblastoma] Wel of geen kankerbehandeling in de resterende tijd? Specialisten over gedeelde besluitvorming bij recidiverend glioblastoom. Ned Tijdschr Geneeskd 2015;**159**:A9790.
16. Mathijssen EGE, Vriezekolk JE, Popa CD, et al. Shared decision making in routine clinical care of patients with rheumatoid arthritis: an assessment of audio-recorded consultations. Annals of the Rheumatic Diseases 2020;**79**(2):170-75.

17. Mathijssen EGE, van den Bemt BJF, Wielsma S, et al. Exploring healthcare professionals' knowledge, attitudes and experiences of shared decision making in rheumatology. RMD Open 2020;**6**(1).
18. Stewart M, Brown JB, Weston WW. Patient-Centred Interviewing Part III: Five Provocative Questions. Can Fam Physician 1989;**35**:159-61.
19. Roter DL, Hall JA, Kern DE, et al. Improving physicians' interviewing skills and reducing patients' emotional distress. A randomized clinical trial. Arch Intern Med 1995;**155**(17):1877-84.
20. Clark NM, Gong M, Schork MA, et al. Impact of education for physicians on patient outcomes. Pediatrics 1998;**101**(5):831-6.

Appendix III – additional figures

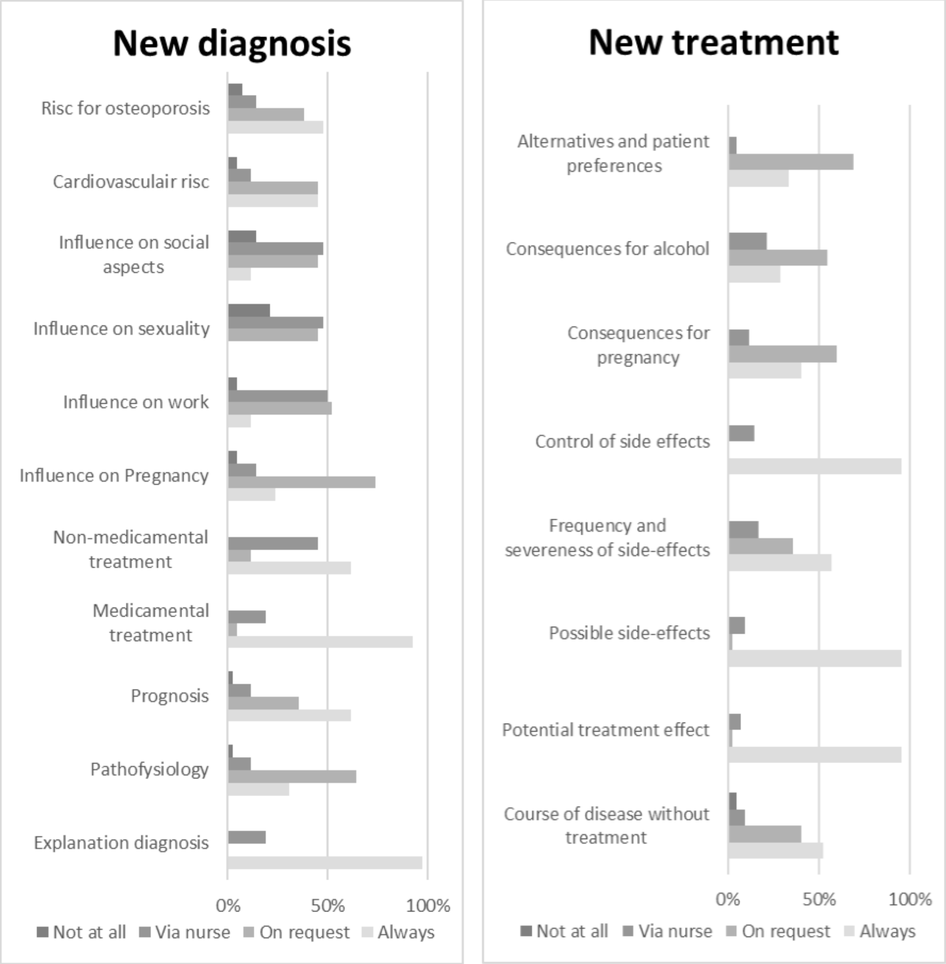

Figure 1. Aspects to discuss when explaining a new diagnosis on the left; aspects to discuss when starting a new treatment on the right.

Residents could choose multiple options (always, on request, via specialized nurse or not at all). Horizontal bars represent percentages.

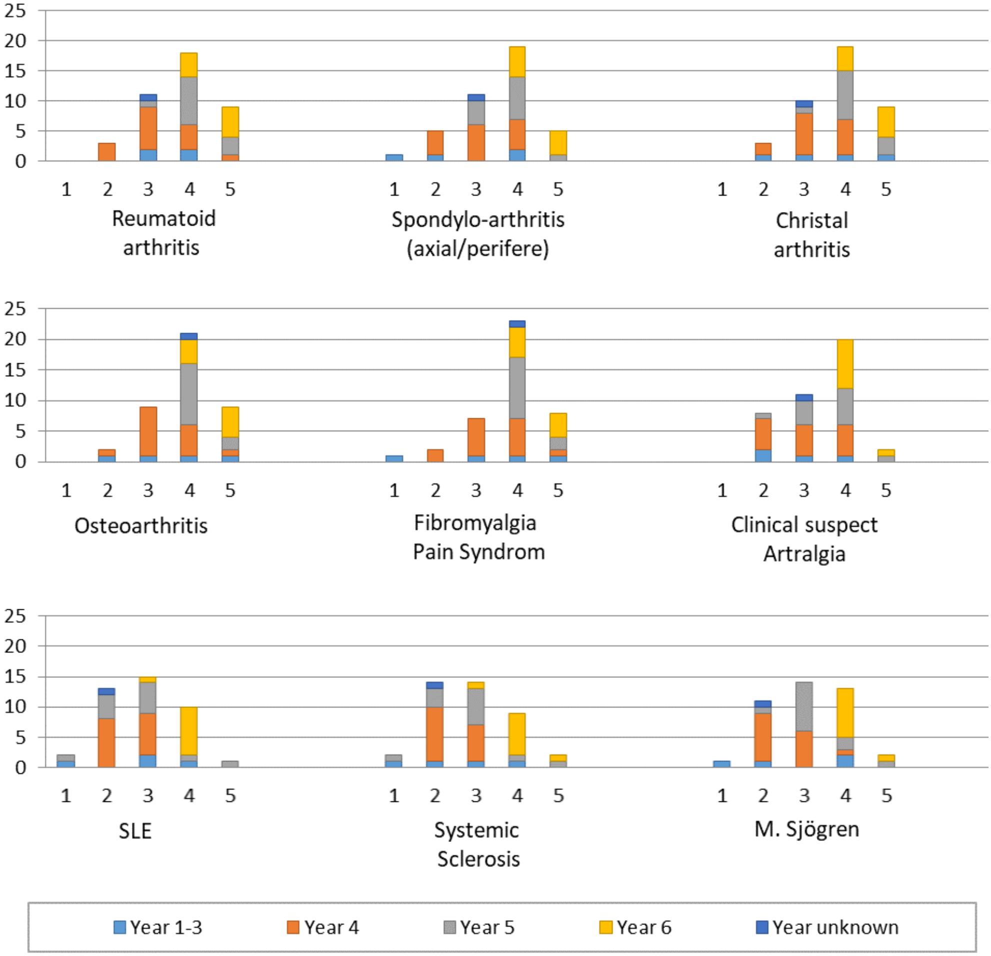

Figure 2. Self-rated skills for discussing different rheumatologic diagnosis.
Vertical axis number respresents the number of respondents. Horizontal axis shows the self-rated level of expertise: 1 knowledge of; 2: strict supervision; 3 limited supervision; 4 unsupervised; 5 level rheumatologist.

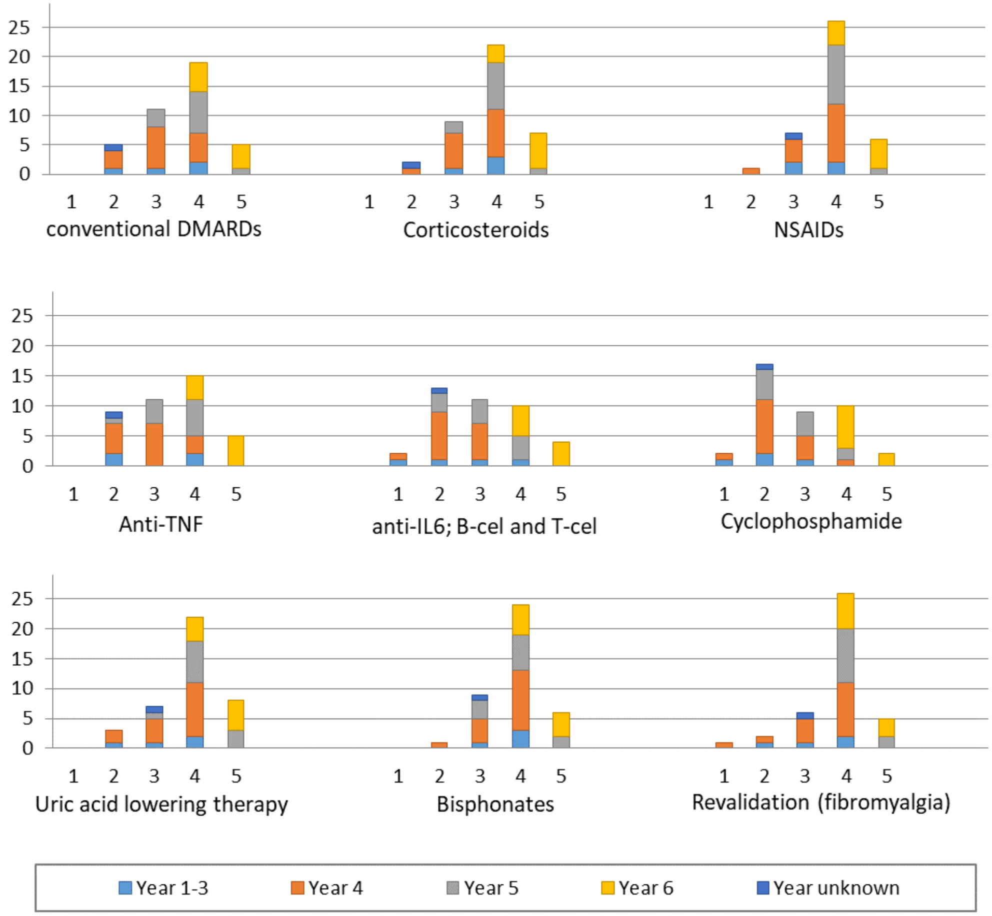

Figure 3. Self-rated skills for discussing different treatment options.
Vertical axis number respresents the number of respondents. Horizontal axis shows the self-rated level of expertise: 1 knowledge of; 2: strict supervision; 3 limited supervision; 4 unsupervised; 5 level rheumatologist.

CHAPTER 4

4

Global application of DSD-related electronic (e) resources: e-learning, e-consultation and e-information sharing

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Abstract

The past 20 years have seen proliferation of electronic (e) resources that promote improved understanding of disorders of sex development (DSD): e-learning for physicians and trainees, e-consultation between clinicians, and e-information for families and affected individuals. Recent e-learning advances have emerged from the European Society for Paediatric Endocrinology's online learning portal for current physicians and trainees. Developed with attention to developing clinical competencies incorporating learning theory, and presenting material that represents international best practice, this e-learning portal offers advances in training, making information more accessible for clinicians and trainees. Multiple levels of instruction, authentic case examples, collaborative forums for physicians and trainees, individualized feedback and user-friendly tools represent advances in trainee and physician learning that can take place in any location. e-consultation is an emerging tool that aims to connect physicians with specialists experienced in DSD care. Although it faces logistical challenges, e-consultation carries the potential to improve DSD care, especially in remote areas with limited access to DSD specialists. e-information for families and patients of all ages is widely accessible online, often with focus on DSD biology, medical care, and psychological and social support. e-information tools aid self-management and support of those affected by DSD. Efforts to improve these resources should aim to map information to individual users, incorporate optimally clear nomenclature, and continue as a 'shared enterprise' of clinicians, affected individuals, families and researchers. Improving the quality of DSD-related e-learning and e-information and developing e- consultation carries the potential to transform DSD care and support for patients, families and physicians worldwide.

Introduction

Since the original description in 1989 that defined distance or distributed learning as "the application of telecommunications and electronic devices which enable students and learners to receive instruction from some distant location" [1] , electronic-learning (e-learning) has evolved greatly due to the advent of the internet. In some areas of medical e-learning, resources and websites have matured and are producing high-quality information for learners and observers. Within the context of disorders of sex development (DSD), e-learning has similarly grown and developed in medical training.

Additionally, electronic consultation (e-consultation) has emerged as a valuable tool to aid communication between primary care and specialist clinicians [2,3] . Lastly, information offered electronically (e-information) is created specifically with patients, parents and families in mind[4,5]. In recent studies, e-information is regarded as a valuable tool to help patients with chronic diseases self-manage a condition [6]. Today, it is impossible to imagine life without electronic learning, consulting, and sharing information.

In this chapter, we first focus on DSD e-learning, incorporating current concepts of competency development and assessment in medical education. Then we discuss recent developments of e-consultation and make proposals specifically applicable for DSD care at a global level. Thirdly, we review current e-information available on DSD for physicians, families and patients and we discuss important topics in e-information development. At the end we propose strategies for further developing of these online resources.

E-learning for DSD

In an effort to combine instruction and formative assessment in learning as well as Competency-Based Medical Education (CBME), the European Society for Paediatric Endocrinology launched the initiative to develop an interactive e-learning portal for Paediatric Endocrinology [7]. The ESPE e-learning portal (http://www.espe-elearning.org) provides a rich source of information for experts, fellows, residents and students. The portal is freely accessible for medical students and professionals through a simple login procedure. Available chapters on DSD concisely describe physiology and pathophysiology, along with practical approaches to management and treatment. In addition, real-life cases are integrated so students may consider solutions for the diagnostic and management concerns in a step-wise and interactive manner. Here we briefly discuss some pertinent issues for e-learning in relationship to DSD.

How can e-learning be effective for clinicians?

During clerkships and residency, clinicians learn via an apprenticeship model. Today, this model faces several challenges [7], not only because of educational issues, which will be addressed in a next paragraph, but also because the development of medical expertise is happening very quickly, making collaboration with many specialists for the treatment of DSD necessary. Moreover, the relationships between doctors and patients are shifting; doctors are challenged to learn from patients' experiences. The ESPE portal offers a possibility to learn via a distance-learning model in which up-to-date knowledge is presented: anytime, anywhere, and independently. Combining the best elements from these two models—online learning and face-to-face instruction— [8], the portal emanates from the idea of blended learning. Blended learning brings together the clinicians' daily practice of high-quality interactions between colleagues with clinical theory, cases, extra exercises, and immediate feedback on multiple choice questions. Therefore, the ESPE e-learning portal is not meant to be a stand-alone learning environment; it incorporates the blended learning model and should thus be imbedded in the daily practice.

Maximizing the use of the ESPE e-learning portal, users can communicate their thoughts directly to a facilitator; this allows self-directed learning to fuse with social networking capabilities of the portal. The portal's introduction of online, high-quality interaction between experts and clinicians is highly valued by the users [9].

Can e-learning be helpful to attain clinical reasoning?

Clinical reasoning is a cornerstone of the care clinicians provide. Even with ample publications on clinical reasoning, there is no clear consensus regarding what clinical reasoning entails or how it might be best taught and assessed. Durning et al [10, 11] propose a model that acknowledges the complexity of the situation. The expertise of clinical reasoning is a performance within a set of boundaries, created by dynamic interaction between physician, patient and practice. For teachers as well as assessors, it is important to explore the situation in its entirety by asking: 'What is actually occurring?' and 'Are we recognizing all of clinical reasoning's component parts?' Consequently, we cannot define clinical reasoning as the result of a single chain of consecutive events. Rather, there are multiple, equally acceptable paths that help develop clinical reasoning. Implications for learning and assessment focus on activities undertaken before and during a task and reflection during and after a task. The results of these activities depend on both the environment and the participants. These items include physician factors such as knowledge and experience, patient factors such as presentation and acuity of illness, and practice factors such as appointment length and staffing [10, 11].

Within the ESPE learning portal, chapters on DSD are authored by clinical care providers, as well as by individuals who have insight on specialized DSD clinical care. Incorporating both research and the experience of the authors, the chapters aim to convey elements of clinical reasoning. The open questions after each section focus on the specific items learned. But, around the world, specific religious and cultural aspects might influence clinical reasoning and decision-making. So, the outcome depends on the boundaries of the case in the specific environment and on the particular interaction between the case components. To facilitate reflection on their activities, residents and fellows are linked with an expert to enable comprehensive discussions on components of medical care in cases of DSD.

What learning theoretical requirements need to be set for the environment?

For those involved in designing instructional materials, an adequate understanding of how human learning works is important. The cognitive learning theory explains how mental processes transform information received by the eyes and the ears into knowledge and skills in human memory [12] . In short, the learner selects the most important information, works this through his limited working memory, integrates the auditory and visual information, and relates the information with existing knowledge from long-term memory. With repeated rehearsal during practice, the information will be stored in the long-term memory. Retrieval depends on the quality of storage. From this information processing theory, we learn that information must be well-organized by a) stressing learning objectives and main points, b) managing complexity by braking lessons into parts, c) delivering examples and well-structured tasks, d) giving (immediate) feedback, e) being easily accessible by glossary, overview and hyperlinks, f) presenting information in words and in visualisations, animations, simulations, graphics.

The constructivist learning theory stresses that learning is an active, constructive process. Learning is the active linking of existing knowledge with information, offered by a new experience. We may conclude that everyone has a different arsenal of knowledge and that each person builds a unique representation of the world. The new information is always linked with a certain experience and its specific context. This so-called ‘situated cognition’ works like an index of knowledge and skills. From the constructive learning theory, we learn that we can make learning more effective by a) presenting information in a meaningful, relevant context, b) stimulating active engagement, c) offering different situations to apply the information, d) ending with a review and encouraging reflection on what has been learnt.

Also with respect to andragogy, the studies of adult learning, the above-mentioned educational requirements form the base of the ESPE portal. Some features need further explanation:

I. Offering different levels: to build effectively on former constructs of participants (e.g. medical students, residents, and fellows), the information should connect with former knowledge. In the portal, two levels of difficulty are offered, and the participant may choose where to start.

II *Using authentic cases*: meaningful experiences are valuable tools for learners. Adults seek relevant information on issues that are immediately applicable. Personal experiences and previous conclusions are the building blocks while learning on the job. Since medical cases are meaningful learning experiences, a number of well-chosen cases are presented in the portal.

III. *Working together*: In the 'Forum,' users create or work out a case together and help each other with a diagnosis. Users are asked to critique each other's reasoning, compare their reasoning with others', and evaluate while collaborating. In so doing, they may discover new possibilities. By having interactions of teacher—teacher, student—student, and teacher—student, the learners are personally engaged and control their own learning.

IV. *Getting feedback*: In the portal, immediate feedback is given on any multiple choice question. In certain exercises, senior endocrinologists give personal feedback on open questions, assignments, and writings of the learner. This case-specific feedback facilitates instructive, hands-on learning without patient risk. Additionally, learners can contact an expert or the Forum with their questions.

V. *Being user friendly*: The portal is learner-oriented; the learner can make choices as to cases, level, theory, and way of learning and support. To navigate easily, there is a library called Mediclopedia, consisting of an alphabetically-ordered list of subjects with links and chapters. The users can incorporate their personal strategy, starting with theoretical information or with a specific case. After the development of the first cases and theory chapters, users offered ideas for improvements, which were incorporated in the learning portal.

Assessment in E-Learning

Competency-based learning

Competencies are defined as tasks (simple or complex) a successful candidate must be able to handle, and during which s/he uses the correct and relevant knowledge, skills, attitudes and meta-cognitions at the appropriate time to manage a task successfully [13]. The Canadian Medical Education Directions for Specialists (CanMEDS) competencies contain the domains: Medical Expert, Communicator, Collaborator, Manager, Health Advocate, Scholar and Professional. [14,15] .

The ESPE e-learning portal provides real-life cases with specific questions in order to train to achieve the seven competences relevant for clinicians. For example, the competence ' Communicator' is trained by offering open questions on how to communicate with parents regarding genital surgeries. Feedback is provided both by standard answer models with essential guidelines and personalized feedback provided by experts. In several small-scale pilot studies, both the applicability of the portal for competency based medical education as well as the use of personalized feedback were evaluated. These pilot studies suggested that personalized feedback can appropriately train CanMEDS roles of communicator and health advocate. Independent learning and questions with fixed answers seem appropriate for training roles of medical expert and scholar [9] .

Assessment for learning

In recent years, thoughts on the role of assessment in education have changed. This is best characterized as a shift from assessment of learning to assessment for learning. Traditionally, examinations take place at the end of the instruction and are separated from the educational process. This is referred to as *summative assessment*. The purpose of such an assessment is to determine whether the student has acquired sufficient knowledge, skills, etc. Assessment for learning, however, is an approach that embeds the assessment process within the educational process, is maximally information-rich, and serves to steer and foster each student's learning to the maximum of his/her ability. This is referred to as *formative assessment*. [13, 16].

Assessments of competence need to be well structured. First, a minimum number of supervised procedures to be carried out by the trainee should be specified. As competence is case and context-specific, one observation by a single assessor is neither valid nor reliable; several observations are necessary to achieve a valid judgment of the competence of a trainee. Second, knowledge should be part of the assessment of competence since clinical performance is positively correlated with a well-organized knowledge database [17] .

Observation and assessment of trainee performance in 'real-life' professional settings has been a cornerstone of health professions education for centuries. Current assessment practices are characterized by emphasis on workplace-based assessment including implementation of competency-based curricula, and improved supervision and assessment of medical trainees [18].

Assessing individual differences regarding competencies, knowledge, skills and attitudes requires assessment instruments capable of capturing these differences and empirically translating them in the domain of interest into meaningful numbers. [19] Extensive recommendations regarding competency assessment have been formulated recently [13]. Formative and summative assessments do not represent contrasting but rather complementary approaches [20].

Data on the use and effectiveness of e-learning are limited in the assessment of competencies of residents and clinical fellows in their course of clinical specialty or subspecialty training. In previous reports, educational considerations for using interactive web portal in addition to practical experiences were provided. [7, 9]. The development and evaluation of both *formative* and *summative* assessments are two of the main objectives in the ongoing improvement of the ESPE e-learning portal. Special attention is paid to the development of *formative assessments* for compentency-based learning.

A unique feature of medical education (and assessment) research is that it needs to be both locally and internationally relevant [13]. An international working group has formulated a long list of recommendations regarding assessment research. [13]. For both students and faculty, assessment is an issue of high importance. Different countries may have different procedures regarding ethical consent for educational research. In some countries, ethics committees rule educational research automatically as exempt, whereas in others, a full ethics review regarding the educational research is needed [13]. This presents challenges for assessment within the e-learning portal.

E-consultation

Electronic consultation (e-consultation) is rapidly emerging as a valuable tool to aid communication between clinicians—both primary care providers and specialist— in the care of a particular patient. To conduct e-consultation, clinicians may use either a web-based program or a shared electronic medical record. Examples of development of electronic consultation (e-consultation) systems between primary care providers and specialists on a regional basis have been recently reported [21, 22,23]. It is particularly relevant at a global level for the management of rare diseases such as DSD.

Diagnostic consultation platforms for rare diseases and networks of centers of expertise are being developed, such as I-DSD [24] . Developing a robust and secure e-consultation service within the international DSD clinical community was recently proposed as a vital component of the services provided from emerging networks; e-consultation services would help extend the current database and e-learning facilities with an objective to provide expert opinion on a worldwide basis [3]. In order to ensure that patient data are secure and to be clear about who is accountable for decisions and treatments offered through such a network, standards of care need to be carefully set, and there needs to be greater consensus about both information and clinical governance. Additional issues to consider include: traditional physician -patient relationship, malpractice liability, and the costs of the consultation.

The management of the system would require clear definition of the tasks and responsibilities of the various stakeholders. A steering committee with a fixed-term membership should oversee the scope, monitor governance, deal with complaints, record consultations, and have face-to-face e-meetings. Further issues to be considered include: safe translation between languages, understanding and respecting cultural differences while maintaining legal or ethical standards. However, the clear clinical advantages of such a network providing access to expert opinion on a worldwide basis far outweigh the bureaucratic hurdles that would be necessary for governance of such a system.

E-information

Electronic information (e-information) is a term that encompasses online resources of websites, tools, and videos available for individuals with DSD, their families, and others. Many of these tools developed in parallel with the Internet. Some online tools include DSD condition-specific resources such as Prof. Garry Warne's (1997) *Complete Androgen Insensitivity Syndrome* [25]. This booklet is accessible online in PDF format. Other resources take the form of websites that change with ongoing updates (see: dsdfamilies.org, AccordAlliance.org).

Recent scholars discuss the emergence of specific websites and communities for individuals with DSD. The Internet has aided the development of geographically diverse communities for DSD advocacy and support; out of those communities one finds many examples of DSD e-information for affected individuals and families [26, 27]. These e-information resources aim to help individuals to understand DSD diagnoses comprehensively, and to receive support outside of the clinic. E-information resources are found on a number of websites, including sites of support and advocacy organizations, hospitals, and medical organizations, among others.

Existing DSD e-information can be divided into categories: biology, medical care, and psychological & social support. Some of the available resources span multiple categories. *Table 1* illustrates examples of each category of DSD e-information, including sites with informational crossover. Selections have been narrowed to English-language websites and tools that represent different components of E-information on DSD. Easy-to-understand illustrations with clear, age-appropriate descriptions can make e-information resources more accessible to families, young people, and adults alike

Disorders of sex development are understood today with increasing complexity, from recent updates in DSD nomenclature and classification [28], to increased identification of DSD genetic etiology [29]. E-information aids patient and family understandings of biological, medical, genetic, psychosocial, and personal components of DSD. However, the impact of new etiologic and genetic information on the quality of life for patients and families is not fully understood [29]. In turn, it often proves difficult to develop E-information that speaks to the patient and family's quality of life. Questions remain: What, for instance, is most important to address when providing DSD e-information? How should those topics be presented to physicians, families, and patients? To arrive at answers, additional information from patients, families, and physicians is needed. Research should further a focus on those needs, and on the most effective ways to communicate with families and patients of all ages.

Table 1. Examples Of Online information on DSD for Patients, Parents & Professionals

DSD Biology: Specific information on DSD

Genetics

- http://www.dsdgenetics.org

General information

- UK Intersex Association – www.ukia.co.uk
- Intersex Society of North America – www.isna.org *

Condition-Specific Information

DSD and Typical Sex Development

- AboutKidsHealth.ca from the Hospital for Sick Children, Toronto http://www.aboutkidshealth.ca/En/HowTheBodyWorks/SexDevelopmentAnOverview/Pages/default.aspx

Congenital Adrenal Hyperplasia

- Congenital Adrenal Hyperplasia Education and Support Network – www.congenitaladrenalhyperplasia.org *
- Your Child with Congenital Adrenal Hyperplasia – www.rch.org.au/cah_book/index.cfm?doc_id=1375
- Adrenal Hyperplasia Network – www.ahn.org.uk

Androgen Insensitivity Syndrome

- eMedicine – http://emedicine.medscape.com/article/924996-overview
- Complete Androgen Insensitivity Syndrome – www.rch.org.au/publications/CAIS.html

XY/XO Gonadal Dysgenesis

- xyTurners – www.xyxo.org

Hypospadias

- Hypospadias Support Group – www.hypospadias.co.uk

DSD Medical Care

DSD Consensus Statement documents and information

- AccordAlliance.org *

Hospital-based websites

- Seattle Children's Hospital Website— http://www.seattlechildrens.org/clinics-programs/disorders-sex-development/
- University of Michigan Hospital Website— http://www.med.umich.edu/yourchild/topics/dsd.htm

Clinical and research networks

- The Scottish Genital Anomaly Network – www.sgan.nhsscotland.com
- EuroDSD – www.eurodsd.eu/index.php
- The International DSD Network and Registry (I-DSD); www.i-dsd.org
- The International CAH Network and Registry (I-CAH); www.i-cah.org
- DSD-Translational Research Network (DSD-TRN); https://dsdtrn.genetics.ucla.edu/
- DSD Network Registry; https://endovl.org.au/dsdnetwork/
- DSD Life; www.dsd-life.eu/

DSD in psychosocial context

Online resource-based organizations

- Dsdfamilies.org *

Support and advocacy organization specific websites (see other chapters for additional information on support and advocacy organizations)

- Advocates for Informed Choice; www.aiclegal.org
- Androgen Insensitivity Syndrome Support Group (UK); www.aissg.org1
- Androgen Insensitivity Syndrome and Disorder of Sex Development Support Group (USA);
- www.ais-dsd.org
- Androgen Insensitivity Syndrome Support Group (Australia); www.aissga.org.au
- Beautiful You MRKH Foundation; http://www.beautifulyoumrkh.org
- Climb Congenital Adrenal Hyperplasia UK Support Group; www.livingwithcah.com
- Hypospadias & Epispadias Association (USA); http://www.heainfo.org

In additional languages

- Androgen Insensitivity Syndrome Support Group, España (Spain); http://www.grapsia.org
- Asexye – supporting families living with bladder extrophy, cloacal bladder exstrophy and epispadias (Spain); http://www.asexve.es
- Androgen Insensitivity Syndrome-Disorders of Sex Development Support Group, Japan; http://aissgjp.org
- DSDNederland (the Netherlands); http://www.dsdnederland.nl
- AISIA: Associazione Italiana Sindrome Insensibilità Adrogeni (Italy); http://www.sindromedimorris.org/

* These resources fit into multiple categories within the table

The "shared enterprise" of DSD e-information

Some e-information is produced by medical professionals or support and advocacy groups separately; others are collaborative efforts that join together individuals involved in DSD support and care. Other websites and organizations, such as dsdfamilies and Accord Alliance, incorporate affected individuals, stakeholder organizations, and members of medical teams within their organizations and their available resources. Developing e-information with interdisciplinary perspectives around DSD—incorporating medical, biological, psychological, and social information—represents an important shift. Although many hospital websites contain information regarding DSD, websites have increasingly featured collaboration between medical professionals and families and patient stakeholders, with AccordAlliance.org and dsdfamilies.org again serving as examples.

The history of collaboration is longstanding. It includes the work undertaken by Prof. G. Warne and the AISSG-UK in the 1990s to produce "Complete Androgen Insensitivity Syndrome" [25, 30] (Raising Awareness, http://www.aissg.org/12_history.htm#Warne). As discussed by Ellie Magritte of dsdfamilies.org in her invited plenary address at the 4th International-DSD Symposium in Glasgow, Scotland, the improvement of DSD clinical care, research, education and training, and patient and family support is a "shared enterprise" of clinicians and patient/family stakeholders. Thus, collaboration is necessary for medical teams, stakeholder groups, affected youth, adults, and families alike to create and promote e-information of the highest quality and relevance.

The importance of self-management to DSD e-information

Like e-information for chronic conditions, e-information is one tool that enables individual patient and family self-management of a DSD condition [6](Johnston 2012). As a tool for self-management, e-information should further address patient-centered factors outside of diagnosis and treatment to help individuals understand DSD and enhance individual confidence.

Care for individuals with DSD is characterized by varying short and long-term challenges that may emerge throughout one's lifespan [31] —from urogenital variation, to concerns about fertility, gonadal malignancy, adrenal complications, hormone replacement, bone health, and psychosocial support, among others. These complex challenges extend to patients and families who seek to understand DSD in biological, medical, and psychosocial contexts. Clinicians widely acknowledge that family and patient support is needed as challenges are met throughout the lifespan [31].

E-information is one way to provide ongoing support to patients and families. Clinicians recommend comprehensive information sharing in areas of sexual health and fertility, particularly in young patients with DSD [31]. E-information can help patients and families manage those issues positively and proactively as they are encountered. Moving forward, e-information should increasingly include age-appropriate material on sexual health and activity, vaginal dilation, fertility challenges, and on discussing DSD with friends, partners, and loved ones.

Issues in developing e-information: nomenclature

Pursuant to changes in DSD classification and nomenclature [28], the way we discuss DSD has changed greatly in the past 10 years; e-information needs to adapt medical information effectively, that is in accessible forms, for families and affected individuals. Because interdisciplinary DSD teams share a variety of perspectives on DSD with patients and families, e-information should include those perspectives which help to provide clear and accurate understandings of DSD in its many contexts. With regard to one's quality of life, recent research notes the uncertain impact of sharing DSD genetic diagnoses with families [29]. Further research needs to evaluate how discussing DSD with families affects family and patient quality of life, as well as the treatment pathways chosen.

Efforts to make DSD e-information more concise and accessible will refine available e-information. Additionally, e-information will need to help families navigate nuanced DSD terminology by clarifying any confusion around diagnosis, terminologies, and family histories. To consider approachable ways to improve clarity and accessibility in developing e-information, a brief compilation of preferred terms and rationales is attached as Table 2.

Mapping e-information to user

Research widely recognizes the different information needs of patients and families, recommending that children should be told progressively about DSD in ways that are age-appropriate and fitted to each individual (DSD [26, 31] Different e-information users (parents of infants, young children, children; pre-teens; teenagers; young adults; adults; and the elderly) have different needs when accessing information on DSD. These needs should be reflected in the available e-information resources. Attention to each specific e-information audience is critical. Those developing e-information should pay particular attention to the differing needs of clinicians, affected individuals, and families.

Such organizations as dsdfamilies.org and Accord Alliance provide substantive e-information for parents—information needed for a parent to make sense of DSD conditions outside of the clinical environment. Additionally, child and young adult-friendly DSD e-information requires examples of age-appropriate information for children and young adults. To this end, images and visuals—like those on AboutKidsHealth.ca—may promote age-appropriate understandings among youth with DSD. As children with DSD enter puberty, topics including changes in appearance, sexual health, fertility, care

Table 2. Abridged table of preferred terminology on DSD developed by authors and administrators of dsdfamilies.org.

Words to avoid in e-information	Preferred terminology (Objective, accurate, sensitive)	Brief explanation
'Normal/abnormal'	Typical/not so typical/ atypical; Usual/unusual; Common/less common/ varying/different pathways	While statistically DSD may be abnormal in the sense of 'rare', in a social context the word 'abnormal' has a very negative connotation and should be avoided when communicating with parents, patients, and families.
Potential value-laden terms such as: 'Error' 'Problem' 'Insufficient' 'poor' (e.g. "poor virilization")	Mutation, difference, high/low presence, alternative sequence, high/low androgen response	These terms help patients and families understand and accept diagnoses by using descriptive terms to explain the different developmental pathway of the child's body, avoiding words that might give negative connotations.
'Correction of genitalia'	Surgery; genital surgery	'Correction of genitalia' implies that that something is wrong and correction is necessary. This is not helpful when trying to support parents coming to terms with the different anatomical development of the genitals of their child.
'Blind Pouch'	'A vagina without the presence of a uterus and without a cervix'	A vagina is a vagina— even without a uterus or a cervix. "Blind pouch" might seem as though there is a sort of deficiency with a vagina. In a medical context, it means something descriptive, however it is easily misunderstood by patients, families, and parents to mean that the vagina doesn't work properly or is somehow deficient.
'Male/female fetus', 'genetically male/female' (used during assignment process)	Genetic/chromosomal XY/XX/ etc. fetus; baby with XX/XY/XXY/etc sex-linked chromosomes;	By insisting on saying 'the male fetus' or "The baby is genetically male' and has 'male hormones', doctors make sex development of a child confusing and difficult for parents to understand. When there may be doubt regarding gender of rearing, avoid implying that a baby's body is problematically at odds with the chromosomes. Instead, emphasize the complexity of sex development and the specific role of the chromosomal make-up, hormones, and gonads in the development of the child's genitals.

for the gonads, and preparing for paediatric to adult medical care transitions come to the fore. High quality e-information should also address emerging youth concerns outside of the clinic. To accomplish this, e-information developers should seek out and incorporate user feedback—especially from children, teens, and young adults with DSD conditions.

Repurposing related e-information

Patients with DSD conditions often have concerns similar to those of other patient groups: hormone replacement, bone health concerns, and fertility challenges, among others. DSD e-information should consider repurposing this already available information for individuals affected by DSD. Earmarking existing, but not explicitly "DSD," resources may be a cost and time-effective way of rapidly expanding the supply of high-quality e-information.

Developing high quality e-information

It is important that affected individuals and families have access to e-information of good quality. But what constitutes a quality resource? Little is known at present about how e-information is developed and piloted by those responsible for the pertinent websites. Who creates and approves the available information? How is the information disseminated and revised? How does a resource discuss differing treatments and pathways available, or present the advantages, complications, and challenges of the same? As little material exists regarding how to develop high quality DSD e-information, we hope to identify steps and considerations for those developing and refining those resources.

A step-by-step approach to e-resource development

In DSD e-information, the development process and the resulting resource are interdependent—how a resource is developed helps determine its quality and how effectively it can be used. To recommend development moving forward, the authors suggest re-purposing previously proposed plans of developing e-consultation [22] in order to further develop DSD e-information, detailed in fig. 1. The figure is generally adapted to e-resources at large; the legend details the process to further develop e-information, e-learning, and e-consultation.

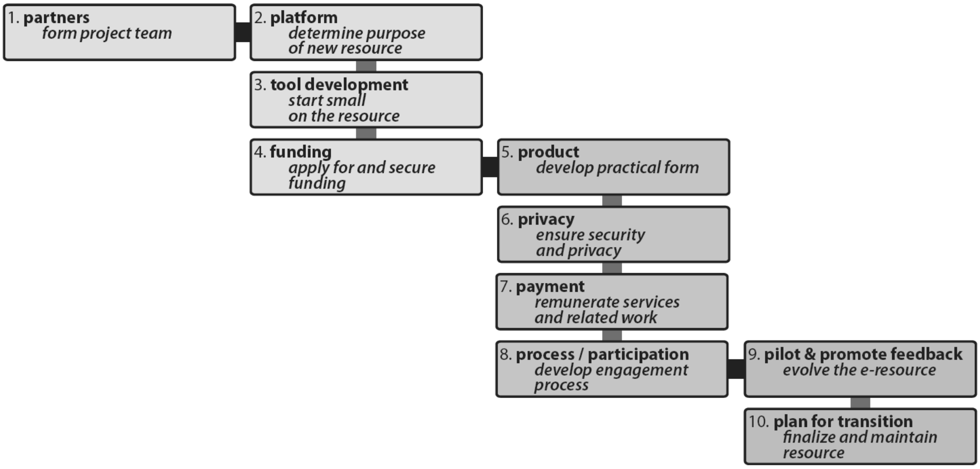

Figure 1. 10 steps to consider when designing electronic (e) resource development—e-learning, e-consultation and e-information sharing (after Liddy et al 2013,[22])

Step 1 Partners: form project team with clinicians, stakeholder organizations, and affected individuals/ families; Step 2 Platform: determine resources available, determine purpose of new resource, and consider how it will be used.; Step 3 Tool Development: develop the timeline and start small on the resource; Step 4 Funding: apply for and secure funding to support e-resource ; Step 5 Product: develop practical form (most times, a website) and map a resource design; Step 6 Privacy: ensure security and privacy concerns are met for those accessing the e-resource; Step 7 Payment: remunerate services and related work on e-resource; Step 8 Process/Participation: develop engagement process for students/ fellows etc, affected individuals, parents, and specialists respectively to improve the e-resource; Step 9 Pilot and Promote Feedback: evolve the e-resource based on user and clinician feedback and current evidence; Step 10 Plan for Transition: finalize resource, develop plan to promote and sustain e-resource

Conclusions and Recommendations

As shown in fig 1, technical, administrative and strategic steps have been formulated recently that are equally relevant when establishing e-learning, e-consultation, and e-information [22] (Liddy 2013). Moving forward collaboration in the development of e-learning, e-consultation and e-information has the potential to present DSD more creatively, clearly, and positively, and to fill-in existing gaps. Long distance e-consultation across geographical and national boundaries offers the opportunity to broaden access to healthcare for all DSD patients, and to widen discussion across the DSD community.

Developing quality e-learning, e-consultation, and e-information presents a number of challenges for the individuals, organizations, and teams developing them. For example, presenting e-information that is accurate, updated to best practice, and sensitive to the needs of affected individuals and families may present challenges, as understandings and considerations of DSD may differ internationally. Additionally, the costs of developing and maintaining quality websites are high, and even then, funding to compensate contributors and work on the available information is considerable. Finally, just as it is critical to engage patients and patient groups to hone e-resources, so too is it critical to manage engagement and tool development when forming, piloting, and disseminating an e-information resource.

Given the different target groups of electronic learning, consultation, and information sharing, we would like to make a plea for active interaction. As shown and outlined in fig 2, there is considerable overlap between these categories and high potential for mutually profitable interaction of clinician, family, affected individual, and support/advocacy groups.

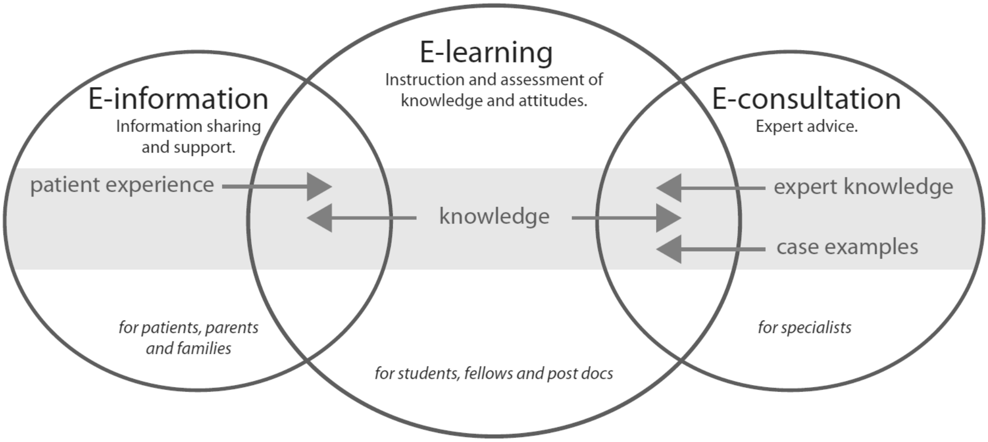

Figure 2. A plea for integration

The aim of ***e-learning*** is to provide a certain standard of state of the art knowledge and attitudes towards patient care and management for target groups of students, residents fellows/postdocs. In addition, the aim is to reach out to "educationally underprivileged" areas in the world, bypassing traditional geographic boundaries by using the internet.

The objective of ***e-consultation*** is to provide forums for structured expert opinions for clinicians and specialists. Before seeking expert advice, it is prudent to access up to date knowledge and current practice. Experts may provide important practical knowledge based on case examples.

The target group of ***e-information*** is patients, parents and families. It is critically important that relevant knowledge is shared with each target group in styles and tones that are fully appropriate. In addition, how patients, parents, and families perceive DSD, how they cope, and how they self-manage DSD are relevant for medical students/fellows/ specialists to understand ramifications of clinical approaches in DSD care.

References

1 Bruder I. Distance learning: what's holding back this boundless delivery system? Electron. Learn 1989; 8: 30-35

2 Horner K, Wagner E, Tufano J. Electronic consultations between primary and specialty care clinicians: early insights. Issue Brief (Commonw Fund). 2011;23:1-14.

3 Drop SL, Mure PY, Wood D, El-Ghoneimi A, Faisal Ahmed S. E-consultation for DSD: a global platform for access to expert advice. J Pediatr Urol. 2012;8:629-32.

4 Wagner E, Austin B, Davies C, Hindmarsh M, Scaefer J, Bonomi A: Improving chronic illness care: translating evidence into action. Health Aff (Millwood) 2001; 20:64–78.

5 Epping-Jordan J, Pruitt S, Bengoa R, Wagner E: Improving the quality of health care for chronic conditions. Qual Saf Health Care 2004; 13:299– 305.

6 Johnston S, Irving H, Mill K, Rowan MS, Liddy C. The patient's voice: an exploratory study of the impact of a group self-management support program. BMC Fam Pract. 2012;13:65.

7 Grijpink-van den Biggelaar K, Drop SL, Schuwirth L. Development of an e-learning portal for paediatric endocrinology: educational considerations. Horm Res Paediatr. 2010;73:223-30.

8 Sleator RD. The evolution of eLearning background, blends and blackboard.... Sci Prog. 2010;93: 3:319-34.

9 Kranenburg-van Koppen L.J.C. , Kalinka Grijpink-van den Biggelaar K, Drop SL. An interactive E-Learning Portal in Paediatric Endocrinology: Practical Experience. Pharmacy 2013; 1: 160-171

10 Durning SJ, Artino A, Boulet J, La Rochelle J, Van der Vleuten C, Arze B,Schuwirth L. The feasibility, reliability, and validity of a post-encounter form for evaluating clinical reasoning. Med Teach. 2012;34:30-7.

11 Durning SJ, Artino AR Jr, Schuwirth L, van der Vleuten C. Clarifying assumptions to enhance our understanding and assessment of clinical reasoning. Acad Med. 2013;88:442-8.

12 Clark RC, Mayer RE. e-Learning and the Science of Instruction: Proven Guidelines for Consumers and Designers of Multimedia Learning, ed. 3, revised. San Francisco, Pfeiffer, 2011.

13 Schuwirth LW, Van der Vleuten CP. Programmatic assessment: From assessment of learning to assessment for learning. Med Teach. 2011;33:478-85.

14 Frank, J.R.; Danoff, D. The CanMEDS initiative: Implementing an outcomes-based framework of physician competencies. Med. Teach. 2007;29; 642–647.

15 Frank, J.R.; Snell, L.S.; Cate O.T.; Holmboe, E.S.; Carraccio, C.; Swing, S.R.; Harris, P.;Glasgow, N.J.; Campbell, C.; Dath, D.; et al. Competency-based medical education: Theory to practice. Med. Teach. 2010;32: 638–64

16 Van der Vleuten CP, Schuwirth LW, Scheele F, Driessen EW, Hodges B. The assessment of professional competence: building blocks for theory development. Best Pract Res Clin Obstet Gynaecol. 2010 ;24:703-19.

17 Dijksterhuis MG, Voorhuis M, Teunissen PW, Schuwirth LW, ten Cate OT, Braat DD, Scheele F. Assessment of competence and progressive independence in postgraduate clinical training. Med Educ. 2009 ;43:1156-65.

18 Govaerts MJ, Van de Wiel MW, Schuwirth LW, Van der Vleuten CP, Muijtjens AM. Workplace-based assessment: raters' performance theories and constructs. Adv Health Sci Educ Theory Pract. 2013 ;18:375-96.

19 Dijksterhuis MG, Jozwiak I, Braat DD, Scheele F. An exploratory study into the effect of time-restricted internet access on face-validity, construct validity and reliability of postgraduate knowledge progress testing. BMC Med Educ. 2013 ;13:147

20 van Mook WN, Van Luijk SJ, Fey-Schoenmakers MJ, Tans G, Rethans JJ, Schuwirth LW, van der Vleuten CP. Combined formative and summative professional behaviour assessment approach in the bachelor phase of medical school: a Dutch perspective. Med Teach. 2010;32:e517-31.

21 Straus SG, Chen AH, Yee H Jr, Kushel MB, Bell DS. Implementation of an electronic referral system for outpatient specialty care. AMIA Annu Symp Proc. 2011:1337-46.

22 Liddy C, Maranger J, Afkham A, Keely E. Ten Steps to Establishing an e-Consultation Service to Improve Access to Specialist Care. Telemed J E Health. 2013 Sep 27.

23 Keely E, Liddy C, Afkham A. Utilization, benefits, and impact of an e-consultation service across diverse specialties and primary care providers. Telemed J E Health. 2013;19:733-8.

24 Ahmed SF, Rodie M, Jiang J, Sinnott RO. The European disorder of sex development registry: a virtual research environment. Sex Dev. 2010;4:192-8.

25 Warne, GL. Complete Androgen Insensitivity Syndrome. The Department of Endocrinology and Diabetes Royal Children's Hospital. Parkville, Victoria, 1997.

26 Preves SE. *Intersex and identity: the contested self*. Rutgers University Press. Piscataway, New Jersey, 2003.

27 Still B. *Online Intersex Communities*… .Cambria Press. Amherst, New York, 2008. .

28 Hughes IA, Houk C, Ahmed SF, Lee PA 2006 Consensus statement on management of intersex disorders. *Arch Dis Child*. 2006; 91:554–563

29 Sarafoglou K, Ahmed SF. Disorders of Sex Development: Challenges for the Future. J Clin. Endocrinol Metab. 2012; 97:2292-2294.

30 Raising Awareness. (2011). Retrieved November 5, 2011, from http://www.aissg.org/12_HISTORY.HTM

31 Lee PA and Houk C. "Chapter 14 Disorders of Sex Development: Clinical Approaches." In Fenway Guide to Lesbian, Gay, Bisexual, and Transgender Health. American College of Physicians , Philadelphia 2008. pg 393-416.

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CHAPTER 5

An interactive E-Learning Portal in Paediatric Endocrinology: Practical Experience

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Abstract

Based on educational considerations, the European Society for Paediatric Endocrinology (ESPE) e-learning portal has been developed, providing an interactive learning environment for up-to-date information in paediatric endocrinology. From March 2011 to January 2012, five small-scale pilot studies were completed to assess the usefulness of the structure and content by senior experts, fellows, residents and medical students. Altogether, 8 cases and 4 chapters were studied by a total of 71 individuals: 18 senior experts, 21 fellows, 10 medical students, 9 regional paediatric ians and 13 residents, resulting in a total of 127 evaluations. Participants considered the portal content interesting and appreciated the way of learning compared to traditional learning from literature and textbooks. Special attention was paid to assess the personalized feedback given by experts to fellows and residents who completed the portal. Feedback from experts included both medical understanding and communication skills demonstrated by fellows and residents. Users highly appreciated the feedback of the medical experts, who brought perspectives from another clinic. This portal also offers educational opportunities for medical students and regional paediatric ians and can be used to develop various CanMEDS competencies, e.g., medical expert, health advocate, and scholar.

Keywords

E-learning; interactive learning; feedback; CanMEDS; paediatric endocrinology

Introduction

E-learning refers to the use of various kinds of electronic media and information and communication technologies (ICT) in education. The term "e-learning" includes all forms of educational technology that electronically or technologically supports learning and teaching. Various examples of e-learning in medical education programs have recently been described in the literature. Daetwyler [1] offered an example of using e-learning to enhance physician patient communication and in teaching how to present bad news to a patient. Dyrbye [2] concluded that e-learning has become an important tool in physician education, enabling advanced curriculum development, instruction, assessment, evaluation, educational leadership, and education scholarship.

Feedback plays a central role in learning. It offers students insight into their learning progress and supports students' skill development. Ideally, feedback is provided face-to-face and is concrete; it identifies strengths and weaknesses and offers improvement strategies [3]. Assessment methods provide two forms of feedback: summative and formative feedback. Feedback from the summative assessment plays a central role in certification. Feedback from the formative assessment provides primarily educational feedback [4–6]. Both summative and formative feedback play an important role in learning. Therefore, e-learning should provide both summative and formative assessments [7].

Feedback can be given in an e-learning portal, which is illustrated by Anderson's interactions in an online learning environment (see Figure 1). This figure illustrates the two major human actors, learners and teachers, and how they interact with each other and with the content. In the figure on the left there is interaction between learners and teachers and the content, using a variety of synchronous or asynchronous activities such as video, audio, computer conferencing or interactions. Here, feedback can be given personally. The right side illustrates the independent use of structured learning resources. Common tools used in this mode include computer-assisted tutorials, tests, drills and simulations [8]. Here feedback is given automatically, by predefined answers.

Not only knowledge, but also competences count in medical education. Since the nineties, Competency-Based Medical Education (CBME) has become a priority in many countries. A widely-used framework for CBME is the CanMEDS model from the Royal College of physicians and surgeons of Canada. CanMEDS is a derivative from Canadian Medical Education Directives for Specialists. It identifies and describes seven roles that lead to optimal health and health care outcomes: medical expert (central role), communicator, collaborator, manager, health advocate, scholar and professional [9–14].

Combining the instruction and formative assessment in learning and CBME, an interactive e-learning portal for Paediatric Endocrinology [15] was developed [16]. According to Deming's Plan-Do-Check-Act cycle [17], several small-scale pilots were performed to check on the user experience, quality of the teaching content, navigation and interaction. Furthermore, learning by personalized feedback was evaluated. One of the outcomes of the pilot studies was the experts' opinion that CanMEDS roles can be trained within the e-learning portal.

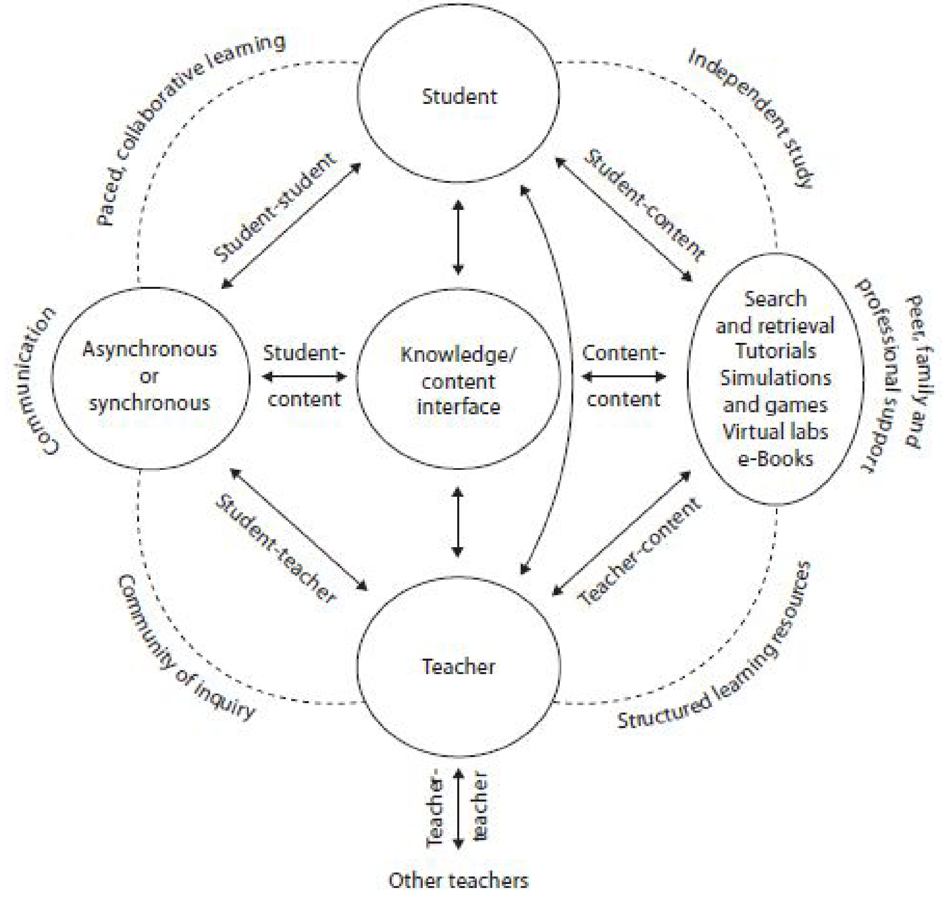


Figure 1. To create online educational experiences and contexts it is important to further all types of interaction.
Well known is the human interaction between student and teacher about learning content (vertical line). The student-teacher interaction can also take place using a variety of synchronous or asynchronous activities such as video-, audio-, computer conferencing or virtual interactions. In addition, there are structural learning tools associated with independent learning such as (computer assisted) tutorials, drills, and simulations (Anderson, T. 2004 [8]).

Experimental Section

The European Society for Paediatric Endocrinology (ESPE) sets standards for medical training and accreditation in EU member states in the field of Paediatric Endocrinology. In addition to conventional learning methods like daily clinical practice and textbooks, an e-learning portal "Paediatric Endocrinology" [15] was developed in 2010. The aim of developing the web portal was to provide an interactive learning environment for up-to-date paediatric endocrinology. The development of the portal is an on-going process, following Deming's Plan-Do-Check-Act cycle.

Plan

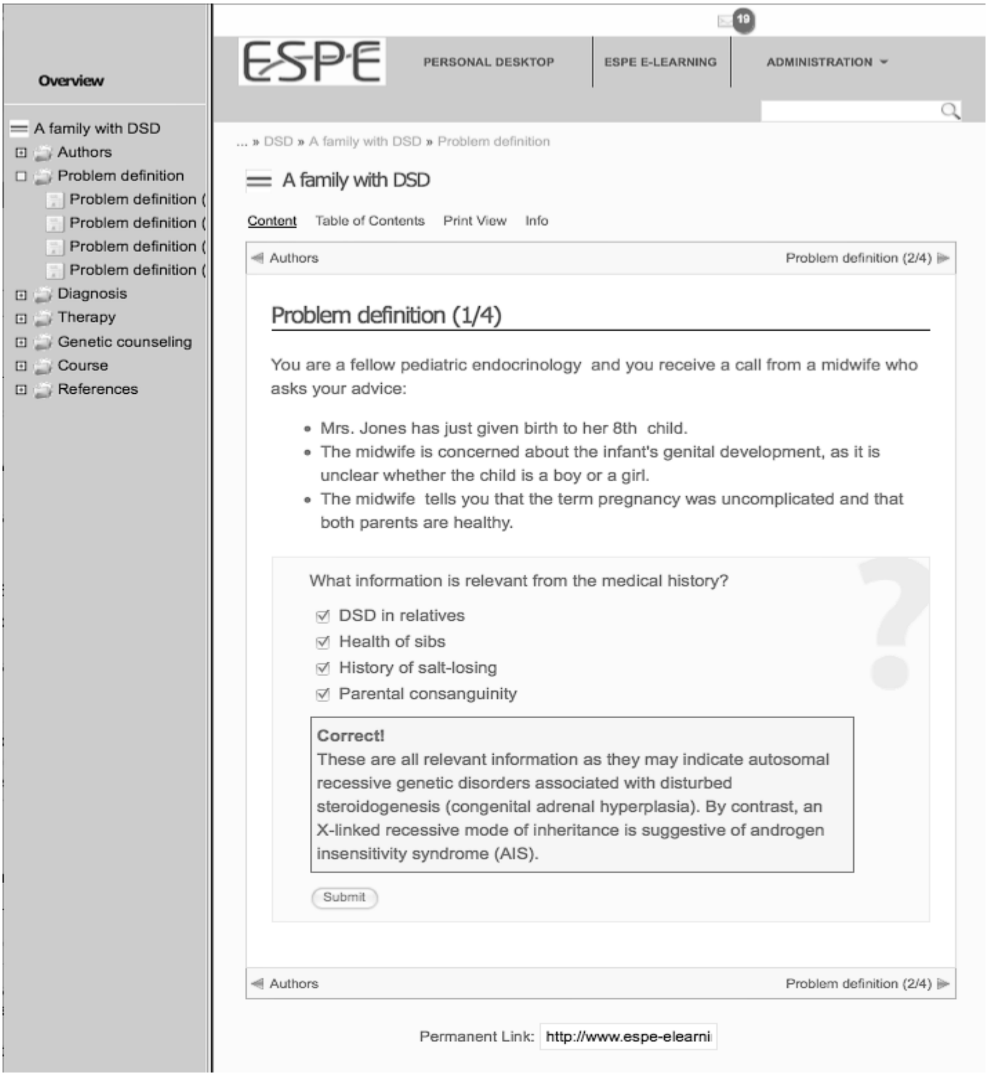
The pedagogical design was based on psychological learning theories and recent research described in the study of Grijpink-van den Biggelaar [16]. The most important feature of this design was the integration of learning, instruction, and assessment. The formative assessment offered immediate feedback to the independent learner [8]. The portal also aimed to support the interaction between aspirant and qualified paediatric endocrinologists to help them acquire and maintain their professional competencies. These assessments were used to train various competencies as described by the CanMEDS model [9–12].

Do

As part of the ESPE e-learning portal and as part of an EU project EURO-DSD (European Community's Seventh Framework Program (FP7/2007-2013) grant agreement No. 2014444) a special module on Disorders of Sex Development (DSD) was developed. Many specialists on DSD contributed to this module resulting in various chapters concisely describing physiology, pathophysiology, and practical approaches to management and treatment of DSD, such as the use of therapeutic agents. In addition, real-life cases were provided, inviting the student to solve diagnostic and management problems in a step-wise and interactive manner. The first version supported independent learning by providing formative assessments. Both multiple-choice and open questions with predefined answers were provided, written by experts on DSD. Examples of questions are provided in Table 1 and a visual illustration is provided in Figure 2. Based on the outcome of the first pilot, a second version of the portal was developed. This portal contained two cases, with each three extra feedback questions that enabled personalized feedback. Fellows were requested to submit their answer on these questions to an expert, who subsequently provided electronically personalized feedback. An example of these feedback questions is also provided in Table 1.

Table 1. Abbreviated example of an interactive case assessing various competencies.

<table>
<tr><td colspan="3"><i>You are a fellow paediatric endocrinologist and you receive a call from a paediatric ian who asks your advice: Mrs. Johnson has just given birth to her second child. The paediatric ian is concerned about the infant's genital development, as it is unclear whether the child is a boy or a girl.</i></td></tr>
<tr><th>Competence</th><th>Question</th><th>Assessment</th></tr>
<tr><td>Medical expert</td><td>What information do you need from the paediatric ian?</td><td>Multiple-choice question</td></tr>
<tr><td>Medical expert</td><td>What information is relevant to provide to the paediatric ian?</td><td>Open question, provide correct items</td></tr>
<tr><td>Collaborator</td><td>What is your advice to the paediatric ian?</td><td>Open question, feedback provided by expert</td></tr>
<tr><td>Communicator</td><td>What is your advice to the paediatric ian to tell the parents? What to tell family and friends?</td><td>Open question, feedback provided by expert</td></tr>
<tr><td colspan="3"><i>The genitalia of the otherwise healthy infant indeed look very ambiguous.</i></td></tr>
<tr><td>Medical expert</td><td>What information regarding the physical examination are you specifically interested in? What further information needs to be collected and what tests need to be performed after the initial physical examination?</td><td>Multiple-choice question</td></tr>
<tr><td colspan="3"><i>The karyotype is 46,XX; based on hormonal and ultrasound investigations you diagnose congenital adrenal hyperplasia.</i></td></tr>
<tr><td>Medical Expert</td><td>Deficiency of which enzyme is most likely responsible?</td><td>Multiple-choice question</td></tr>
<tr><td colspan="3"><i>Both parents are very relieved that they have a girl and the mother says: "From the beginning, I had the feeling that I had a daughter!"</i></td></tr>
<tr><td>Communicator</td><td>You discuss this condition with the infant's parents. What do you say?</td><td>Open question, feedback provided by expert</td></tr>
</table>



Problem definition (1/4)

You are a fellow pediatric endocrinology and you receive a call from a midwife who asks your advice:

- Mrs. Jones has just given birth to her 8th child.
- The midwife is concerned about the infant's genital development, as it is unclear whether the child is a boy or a girl.
- The midwife tells you that the term pregnancy was uncomplicated and that both parents are healthy.

What information is relevant from the medical history?

☑ DSD in relatives
☑ Health of sibs
☑ History of salt-losing
☑ Parental consanguinity

Correct!
These are all relevant information as they may indicate autosomal recessive genetic disorders associated with disturbed steroidogenesis (congenital adrenal hyperplasia). By contrast, an X-linked recessive mode of inheritance is suggestive of androgen insensitivity syndrome (AIS).

Permanent Link: http://www.espe-elearni

Figure 2. A screenshot from a multiple-choice question in the studied e-learning portal.

Check

From March 2011 to January 2012, five small-scale pilot tests were conducted to evaluate the developed portals by senior experts, general paediatric ians, fellows, residents and medical students. Experts are defined as paediatric ians specialized in endocrinology, fellows are paediatric ians in specialty training in paediatric endocrinology, residents follow a specialty training in paediatric s, and medical students are mostly in their 4–6th year. An overview of the pilot studies and the main characteristics is provided in

Table 2. Pilots 1–5 were designed to evaluate independent learning as provided in the first version of the pilot. The second pilot was specifically conducted to evaluate personalized feedback in the second version. In all pilot studies, participants were asked to fill in a standard questionnaire investigating opinions about content, user experience, time spent, and the questions used in the e-learning portal. In the second and fifth questionnaire, participants were also asked about interactivity and the ability of the portal to train competences described by the CanMEDS model. Pilots 3–5 focused on the use of the portal for specific user groups.

Table 2. Main characteristics of five pilot studies.

	Time period	Subject	Items studied	Participants
Pilot 1	March 2011	User experience and quality	3 cases, 3 chapters	9 experts, 1 fellows, 9 residents, 6 medical students, total 35
Pilot 2	August 2011	Interaction	2 cases	3 experts, 8 fellows, 3 residents, total 14
Pilot 3	October 2011	Use for medical students	2 cases	4 medical students
Pilot 4	November 2011	Use for regional paediatric ians	2 cases	9 paediatric ians, 1 fellow
Pilot 5	January 2012	Use for masterclass	2 cases	9 experts, 1 fellow

Pilot 1

This pilot was conducted to evaluate user experience, the didactic quality of the content, and navigation in the e-learning environment. Three cases and three chapters were selected for evaluation. A total of 54 people were invited to participate, including senior experts, fellows, residents and medical students worldwide. Chapters and cases contained a various amount of open and multiple-choice questions with standard answer models. All participants were asked to study one chapter and one case and subsequently answer a survey, e.g., does the content connect well to the level of knowledge of the different target groups? Are there sufficient questions in the cases and are these questions adjusted to the needs of the different users? How do experts think about the content of the chapters and the cases?

Pilot 2

In order to test and evaluate the feedback offered in the second version of the portal, 4 experts and 24 fellows or senior residents were asked to participate. In the period of 30 August to 22 September 2011, the fellows and residents, referred to as “students”, were asked to study 2 cases, each containing 3 feedback questions. The answers on these questions were sent to an expert appointed to the student, referred to as “teacher”. The teacher gave personalized feedback to the given response. An answer model with some suggestions was provided for the experts. The student received a notification and had the opportunity to engage in further discussion with the teacher. The experiences were evaluated with a questionnaire, consisting of questions regarding time spent, quality of feedback, evaluation of interaction and evaluation of usability of the e-learning portal to train CanMEDS competences.

Pilot 3

The third pilot evaluated the use of the portal in the master phase of medical student education. The study was conducted at the Erasmus University in Rotterdam, the Netherlands. 24 Master students, completing a minor in paediatric s, were asked to study a chapter and a case describing a real-life situation. Afterwards they were asked to fill in a questionnaire to evaluate time spent as well as the quality and level of the content and the navigation.

Pilot 4

The fourth pilot study evaluated the contribution of the e-learning portal to the on-going education of regional paediatric ians participating in post-graduate training for regional paediatric ians on DSD in the Netherlands in the Rotterdam area. A group of 36 paediatric ians and fellows were asked to study two cases in the portal and subsequently fill in the questionnaire.

Pilot 5

The fifth pilot study was conducted to evaluate the usability of the portal for on-going education for experts and fellows in DSD. The 29 participants in a masterclass were asked to study two cases and subsequently fill in the questionnaire.

Results and Discussion

Pilot 1 included 34 participants: 9 experts, 11 fellows, 9 residents and 5 medical students. For the second pilot 4 experts and 24 fellows or senior residents were asked to participate. Due to the short pilot period and summer period, 3 experts, 8 fellows and 3 residents were able to participate. Only 4 of the 24 medical students replied who were asked to participate in the third pilot. In the fourth pilot, 10 invitees responded. From the 29 participants in the masterclass who were invited for this pilot, 10 persons participated. See Table 2 for an overview of participants of the pilot studies. Altogether, 8 cases and 4 chapters were studied by a total of 71 people: 18 senior experts, 21 fellows, 10 medical students, 9 regional paediatric ians and 13 residents, with a participation rate of 42%. Some participants studied multiple cases or chapters, resulting in a total of 127 evaluations.

Evaluation of Usability and Time Spent

A standard survey was used in all pilot studies to evaluate the studied chapters and cases on content, illustrations, effectiveness of learning compared to other learning methods such as textbooks, and time spent to study a chapter or case. An overview of the results is given in Table 3 and is later discussed in more detail.

Table 3. Results of all pilot studies (n = number of respondents, sd = standard deviation)

Evaluation by subgroup	**N**	**Content (scale 1–10)**		**Illustrations (scale 1–10)**		**Effectiveness of learning**		**Time spent (min)**	
		mean	**sd**	**mean**	**sd**	**mean**	**sd**	**mean**	**sd**
Expert	30	8.3	1.7	7.1	2.2	8.2	1.0	41	37
Fellow	35	8.6	1.5	7.0	2.4	8.3	0.9	56	40
Medical student	20	8.0	0.9	7.2	1.5	7.8	1.2	37	18
Regional paediatric ian	20	8.4	0.9	7.4	1.7	8.2	0.8	19	8
Resident	22	8.3	1.4	7.2	1.6	7.9	1.3	53	45
Total	127	8.4	1.4	7.1	1.9	8.1	1.1	43	36

Regarding the level of difficulty of the studied content, the opinions varied. In general, experts and fellows judged the content as appropriate, but that depended on whether a case or chapter was considered easy or difficult. Medical students however judged the content as appropriate to difficult.

The time spent depended on the length of the case or chapter studied and ranged from a mean of 17 min for the shortest case to a mean of 69 min for the longest. Furthermore, the time spent varied per subgroup. On average residents and fellows spent most time on studying the content. The time spent was mostly judged as appropriate. Fellows indicated that they would like to have clear information about the time needed and the objectives before they start with the case. Experts indicated that "*lack of time was a great threat*", by which they were not able to spend the appropriate time and attention.

Overall, the respondents were enthusiastic about the content of the studied sections. There were some suggestions to break the text into shorter paragraphs in order to make the information easier to digest. More illustrations, photos, animations and diagrams were desired. Also, a short take-home message at the end of a chapter was requested as potentially helpful for students.

On average, respondents were satisfied by the number and the quality of open and multiple-choice questions. Several students remarked that they found questions helpful in the learning process. There were several requests to add more questions to check the knowledge retained. For example: "*I think a combination of multiple-choice and open questions is appropriate. Open questions stimulate to think more and take time. Multiple choice questions cost less time*".

In the first pilot, specific attention was paid to students' wishes for personalized feedback on certain questions. This potential option received an average rating of 8 on a scale of 1–10. There was a suggestion for each page to have a "Feedback" section where users could leave feedback or ask questions on it. It was suggested that personalized feedback may be difficult to implement, as it will require resources. However, as one respondent commented, *"I do not think that every open question should be given personal feedback. Some are easy to explain in general, some need help"*.

Personalized Feedback

The second pilot was especially conducted to evaluate personalized feedback given by experts to fellows and residents. Each studied case contained three feedback questions, where the answer of the fellow/resident was sent to an expert for personalized feedback. On average, fellows spent 30–40 min answering and discussing the three questions in one case while residents spent 40–45 min. Experts needed 15 min in average per case to give feedback. The way of interaction was qualified on a scale of 1 (worst)–10 (best); it showed an average score of 8.3 for experts, 6.3 for fellows and 7.3 for residents. Several fellows preferred receiving feedback by direct e-mail, although the involved experts did not want to use e-mail. The mode of interaction was evaluated as acceptable. The quality of the interaction received a mean score of 8 on a scale of 1–10 (worst-best). Fellows and residents appreciated the opinion of another expert, who brought in another perspective. It was also appreciated not only to receive feedback on pure medical aspects, but also to receive feedback on communication skills. In the pilot the answers of both students and teachers were anonymous, which was highly valued by several students and one teacher. “*Anonymity guarantees that the student feels free to answer*”.

Different Target Groups and Didactical Possibilities

One aspect the pilot studies tested was how applicable the e-learning portal was for different target groups. The number of medical students (4 out of 24) that participated in the third pilot showed little enthusiasm. Their comments mostly involved portal navigation. The content was rated interesting, however difficult. The fourth pilot was set up with regional paediatric ians. The content was evaluated as interesting (see Table 3), the level of material adequate, and the time spent varied from appropriate to short. One respondent stated that it was not fit for a general paediatric ian, but would be very useful for the fellow in the paediatric endocrinology or for endocrinologists for on-going learning.

Furthermore, the second and fifth pilot paid attention to the portal’s applicability of the CanMEDS competences for the education and training in an e-learning environment. In the opinion of the respondents, the e-learning portal can be used to train all competences, in particular the role of Medical Expert (see Figure 3).

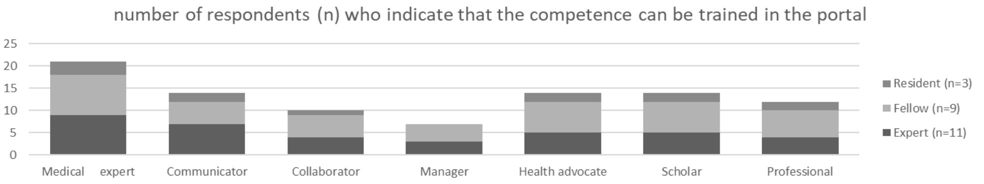


Figure 3. This figures illustrates the opinion of the named subgroups about the applicability to train CanMEDS competences in the studied e-learning portal.
The vertical axis counts the number respondents indicating that the portal is suitable to train the specific competence.

Discussion

The ESPE e-learning portal [15] provides a rich source of information for medical students, residents, fellows, specialists, consultants and teachers around the world. The portal is based on educational learning theories and is continuously improved in order to fit the daily practice of the users. In order to evaluate the content and user experience of the current portal, pilot studies were conducted. The outcomes of the pilot studies are just a preliminary “check” in the plan-do-check-act cycle, evaluating the first versions of the e-learning portal based on educational principles. The results of these “checks” are used for the further development of the e-learning portal.

We noticed that the response rate of these pilot studies was 42%, varying individually from 17%–63%. In literature, comparable response rates are found, for example a survey from a university in Cameroon [18] had a response rate of 51%. We do not know the reasons for not participating as we did not address specific attention to this respect. However, this might be due to the voluntary participation and the time needed to study the content. In future studies, we intend to study what factors affect individual participation and usage in more detail.

Participants of the pilot studies stated that the content was interesting and that the level of the content was appropriate. Valuable suggestions for improvement were made to break the text in shorter paragraphs and provide short take home messages at the end of a chapter or case. These suggestions will be incorporated in future versions of the portal.

We were particularly interested to learn about the wish of the students for personalized feedback and the portals feasibility to provide this. Also, we were interested in the response of both students and teachers to comply with the exchange of information. The quality of feedback exchange was rated highly by both students and teachers. Several fellows preferred receiving feedback by direct e-mail, although the involved experts did not want to use e-mail. Not unexpectedly, time spent was a serious issue. Thus, in future studies, more details on the time needed to complete a section will be provided.

An additional aim of the pilot studies was to obtain initial insight into the use of the CanMEDS competencies, both offering specific items related to CanMEDS roles in problem solving cases and evaluating these competencies. We think that the possibility of personalized feedback is appropriate to train roles such as communicator and health advocate. Independent learning and questions with fixed answers seem appropriate for roles such as medical expert roles and scholar. Our preliminary data suggest that these competencies can be addressed, and we intend to further develop formative assessment modules.

Based on the insights obtained in these pilot studies, we proceeded to further develop the ESPE e-learning portal using an open source learning management system ILIAS [19] with major emphasis on formative assessment and option for summative assessment.

Conclusions

The ESPE e-learning portal [15] provides a rich source of information for medical students, residents, fellows and specialists around the world. The portal was based on educational principles and is continuously improved in order to fit in to the daily practice of the users.

In order to evaluate the content and user experience of the current portal, a number of pilot studies were conducted.

The portal offers possibilities for medical education, both for knowledge and for competence purposes. The portal offers possibilities for independent learning, which seems fit to train competencies as medical expert and scholar. This was accomplished by providing chapters and real-life cases with formative assessment in the form of open and multiple-choice questions with standard answer models. Moreover, the portal offers possibilities to train other competencies such as communicator and health advocate by providing personalized feedback.

It has to be mentioned that the pilot study outcomes were preliminary "checks" in the Plan-Do-Check-Act cycle, evaluating the first versions of the e-learning portal based on educational principles. The results of these "checks" were used to further develop the e-learning portal. Based on the insights from the pilot studies, we further developed the ESPE e-learning portal using an open source learning management system ILIAS [7] with major emphasis on formative assessment and with options for summative assessment.

Conflicts of Interest

The authors declare no conflict of interest.

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References

1. Daetwyler, C.J.; Cohen, D.G.; Gracely, E.; Novack, D.H. Elearning to enhance physician patient communication: A pilot test of "doc.com" and "WebEncounter" in teaching bad news delivery. *Med. Teach.* **2010**, *32*, e381–e390.
2. Dyrbye, L.; Cumyn, A.; Day, H.; Heflin, M. A qualitative study of physicians' experiences with online learning in a masters degree program: Benefits, challenges, and proposed solutions. *Med. Teach.* **2009**, *31*, e40–e46.
3. Orrel, J. Feedback on learning achievement: Rhetoric and reality. *Teach. High. Educ.* **2006**, *11*, 441–456.
4. Gibbs, G.; Simpson, C. Conditions under which assessment supports students' learning. *Learn. Teach. High. Educ.* **2004**, *5*, 3–31.
5. Van den Vleuten, C.P.M.; Schuwirth, L.W.T. Assessing professional competency: From methods to programmes. *Med. Educ.* **2005**, *39*, 309–317.
6. Galbraith, M.; Hawkins, R.E.; Holmboe, E.S. Making self-assessment more effective. *J. Contin. Educ. Health.* **2008**, *28*, 20–24.
7. Schuwirth, L.W.T.; van den Vleuten, C.P.M. A plea for new psychometric models in educational assessment. *Med. Educ.* **2006**, *40*. 296–300.
8. Toward a theory of online learning. Available online: http://epe.lac-bac.gc.ca/100/200/300/athabasca_univ/theory_and_practice/ch2.html (accessed on 6 August 2013).
9. Frank, J.R.; Danoff, D. The CanMEDS initiative: Implementing an outcomes-based framework of physician competencies. *Med. Teach.* **2007**, *29*, 642–647
10. Frank, J.R.; Snell, L.S.; Cate O.T.; Holmboe, E.S.; Carraccio, C.; Swing, S.R.; Harris, P.; Glasgow, N.J.; Campbell, C.; Dath, D.; *et al*. Competency-based medical education: Theory to practice. *Med. Teach.* **2010**, *32*, 638–645.
11. Wong, R. Defining content for a competency-based (CanMEDS) postgraduate curriculum in ambulatory care: A delphi study. *Can. Med. Educ. J.* **2012**, *3*, Article 1.
12. CanMEDS: Better standards, better physicians, better care. Available online: http://www.royalcollege.ca/portal/page/portal/rc/resources/aboutcanmeds (accessed on 6 August 2013).
13. Stutsky, B.J.; Singer, M.; Renaud, R. Determining the weighting and relative importance of CanMEDS roles and competencies. *BMC Res. Notes* **2012**, *5*, doi:10.1186/1756-0500-5-354.
14. Michels, N.R.M.; Denekens, J.; Driessen, E.W.; van Gaal, L.F.; Bossaert, L.L.; de Winter, B.Y. A Delphi study to construct a CanMEDS competence based inventory applicable for workplace assessment. *BMC Med. Educ.* **2012**, *12*, doi:10.1186/1472-6920-12-86.
15. Espe e-learning portal. Available online: http://www.espe-elearning.org/ (accessed on 6 August 2013).
16. Grijpink-van den Biggelaar, K.; Drop, S.L.S.; Schuwirth, L. Development of an e-learning portal for paediatric endocrinology: Educational considerations. *Horm. Res. Paediatr.* **2010**, *73*, 223–230.
17. Deming, W.E. Out of the Crisis; MIT Center for Advanced Engineering Study: Cambridge, MA, USA, 1986.

18. Bediang, G.; Stoll, B.; Geissbuhler, A.; Klohn, A.M.; Stuckelberger, A.; Nko'o, S.; Chastonay, P. Computer literacy and e-learning perception in cameroon: The case of yaounde faculty of medicine and biomedical sciences. *BMC Med. Educ.* **2013**, *13*, doi:10.1186/1472-6920-13-57.
19. Open source e-learning. Available online: http://www.ilias.de/docu/goto.php?target=root_1 (accessed on 6 August 2013).

CHAPTER 6

6

Global application of assessment of competencies of paediatric endocrinology fellows in the management of differences of sex development (DSD) using the ESPE e-learning.org portal

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Summary

Background

To combine instruction and formative assessment (assessment with feedback during the learning process to improve learning) the European Society for Paediatric Endocrinology (ESPE) developed an interactive e-learning portal for pre- and postdoctoral education and training: (http://www.espe-elearning.org). The aim of the study was to evaluate the role of e-learning in the formative assessment of competencies (medical expert, communicator) of paediatric endocrinology fellows in informing patients and parents with a difference of sex development (DSD) about diagnosis and management

Materials and method

Sixty-eight fellows and 32 experts -from various countries- participated in the study. Fellows answered on- line multiple-choice and open questions related to two clinical interactive problem solving cases (case 1: a newborn with congenital adrenal hyperplasia; case 2: a 17-year-old girl with gonadal dysgenesis). Experts provided anonymous feedback on-line, using prepared response elements.

Results

Fifty percent of fellows achieved the correct answer on the first attempt for 16 of 20 questions (case 1 and 2 combined). Only 50% of the key elements of questions 1 and 2 (case 1) and question 2 (case 2) were mentioned. Personalized formative feedback to the open questions from 2-3 experts each was highly appreciated.

The most frequent cultural, religious and regional items mentioned by 12/32 experts were fertility and role of the family. Fellows in their first reply to the open questions did not mention them but acknowledged in their second reply their importance.

Conclusion

The ESPE e-learning portal, offering direct interaction between trainee and trainer, is applicable in on-line formative assessment of trainees.

Background

E-learning, defined as “the application of telecommunications and electronic devices which enable students and learners to receive instruction from some distant location” has evolved greatly due to the advent of the internet [1] . Today, it is impossible to imagine life without electronic learning, consulting, and sharing information. Major advantages of electronic learning are global availability, relatively low cost, options to (unlimited) extensions and regular updates, option to refer to and to link to current textbooks. Case-based learning in an e-learning environment has been proven to be an effective method to practice clinical reasoning [2] . Moreover, current technology allows construction of portals providing interaction between tutor and student/fellow at regional but also (inter) national/global level.

In an effort to combine instruction and formative assessment (i.e. assessment with feedback during the learning process to improve learning) as well as Competency-Based Medical Education (CBME), the ESPE launched the initiative to develop an interactive e-learning portal for Paediatric Endocrinology [3][4][5]. The ESPE e-learning portal (http://www.espe-elearning.org) can be viewed on a computer and also on mobile devices, such as smartphones. It provides a rich source of information on various aspects of paediatric endocrinology such as differences of sex development (DSD). Differences of sex development (DSD) comprise a wide array of congenital conditions in which the development of one or more of the gonads, reproductive structures and genitalia is atypical[6]. Many individuals are recognized in the newborn period when variation in genital development is noted. Later presentations may occur in children or adolescents because of delayed or incomplete pubertal development. Chapters within the e-learning portal concisely describe physiology, pathophysiology and practical approaches to investigation and management. In addition, real-life cases are presented, and the medical student, resident or fellow is invited to solve the diagnostic and management problems in a step-wise and interactive manner [3]. Access to the portal is provided through an automated login procedure via password protection and is global and unlimited.

The most important feature of the portal is the integration of learning, instruction and assessment in the management of patients with paediatric endocrine disorders [3]. According to the CanMEDS framework, “competence” is defined as “the minimally expected standard for an individual ready to start independent practice”[7][8][9]. Several competencies are highly relevant for optimal patient-centered care. In addition to developing skills as ‘Medical Expert’, the competencies of ‘Communicator’ and ‘Collaborator’ are crucial, because informing parents and patients with DSD in a sensitive and appropriate manner is extremely important [10] [11].

The further development of learning and competency assessment tools is necessary to extend the impact of the e-learning portal. Assessment of learners fulfils two distinct goals: formative assessment is defined as assessment during the learning process with direct feedback (assessment *for* learning), whereas summative assessment is a test or exam with a score at the end of a learning process (assessment *of* learning) [12].

Aims

This study was intended to evaluate the role of e-learning in the formative assessment of the 'Medical Expert' competency. We have chosen to focus on a group of fellows and experts representing one specialty, paediatric endocrinology.

An additional objective was to evaluate the development of communication skills in informing patients and parents about diagnosis and management. Special attention was paid to evaluate the value of personalized feedback and the feedback by different experts. A qualitative analysis of the communication skills such as wording and empathy will be discussed in more detail in a separate paper.

The third aim was to assess religious, regional and cultural aspects relevant to management of DSD by performing the study on a global level.

Study outline

Senior paediatric endocrinologists (experts) were approached and asked to provide names and email addresses of paediatric ians undergoing subspecialty training in paediatric endocrinology (referred to hereafter as fellows) in their respective departments who agreed to being approached to participate in the assessment study. The fellows were invited to login into a special e-learning course within the ESPE e-learning portal, where they had access to the general content of the system, including chapters on differences of sex development (DSD).

Access was provided to two interactive cases on the subject of DSD prepared as e-learning exercises in English. The two clinical cases are within the expertise of paediatric endocrinologists and it is likely these cases would be referred to these specialists. The study also addresses the role of the paediatric endocrinologist in an interdisciplinary DSD team.

Case 1, entitled 'A newborn with "ambiguous genitalia' focused on the stepwise approach to investigation and management of a newborn referred by a midwife because of atypical genital development due to congenital adrenal hyperplasia. Case 2, entitled 'Ranya: an adolescent girl with delayed menarche' addressed the approach to a 17-year-old girl with delayed menarche as a result of 46,XY gonadal dysgenesis. The format of these cases includes a step-wise clinical approach to the assessment of a patient with DSD, followed by multiple-choice (MC) questions testing medical knowledge ('Medical Expert' competency). The correct answer to each MC question is provided to the learner directly after a correct answer and if incorrect after the second attempt to answer it. At crucial stages of the problem-solving process, three open-ended questions (i.e. free text response) were included for each case, to which the fellow was invited to describe how (s)he would handle the situation, with particular focus on how the fellow would inform the parents and/or patient of the key information regarding diagnosis, implications and management. The open questions from both cases are presented in table 1.

The replies to the MC questions were stored centrally for further analysis, and the replies to the open-ended questions were submitted to 2-3 experts from various parts of the world (excluding the expert at the institution where the fellow was employed); at least one of the experts was chosen to be from the same geographic region as the fellow. The experts were invited to reply with a brief formative commentary to the fellows' responses. They were free to use an example answer prepared by the study team, see supplementary table 1. The fellows were asked to acknowledge and reply to the comments of the experts. All these procedures were handled anonymously through the e-learning portal.

To evaluate the responses of the fellows to the open questions as objectively as possible several key items were identified *a priori* by the study team as critical elements of an adequate response. The set of response elements (Case 1, question 1 N = 7; Case 1, question 2 N = 8; Case 2, question 1 N = 6) prepared by the study team was submitted to the group of experts with the request to indicate at least 5 key items that, in their opinion, should be mentioned in the answer by the fellow. From their responses a set of key items was derived, which was used to evaluate the answers of the fellows by scoring the number of key items mentioned in their response.

Finally, the fellows were invited to complete a brief web-based questionnaire regarding feasibility/receptivity and value of personalized feedback related to competence development.

Table 1. The three open questions presented to the fellows for each case

Case	#	Question text
A newborn with ambiguous genitalia	1	You are the paediatric endocrinology fellow in charge; you have obtained the medical history information and you have examined the baby. What do you tell the parents at this stage?
	2	You have just made the diagnosis of congenital adrenal hyperplasia (CAH), most likely as a result of 21α-hydroxylase deficiency. Give a detailed account of how you will inform the parents of the diagnosis.
	3	Many years later, when Eve is 17 years of age, you prepare a letter at the time of transition of her medical care to an internist-endocrinologist and gynaecologist. You provide her with a written summary of her medical history. Specifically you explain once more in understandable terms: a. what kind of condition she has b. what happened before she was born c. why she had surgery d. why she is being referred to an internist-endocrinologist / gynaecologist for ongoing care e. whether she is required to take medication all her life f. whether she will be able to bear children in the typical way and if so, the risk is that one of her children will also be affected Prepare this letter.
Ranya, an adolescent girl with delayed menarche	1	You have made the diagnosis 46,XY gonadal dysgenesis. Give a detailed account of how you will arrange the sharing of information to Ranya and her parents. Consider the following elements: the setting; allotted time; key elements to be discussed regarding the diagnosis; relevant therapeutic issues; long-term implications; follow up; other considerations
	2	Give a detailed account of how you will inform Ranya and her parents of the increased risk for development of a gonadal germ cell tumor and the therapeutic options. Consider the following elements: relative risk; time frame; therapeutic procedures and timing; follow up; other considerations.
	3	Ranya is now 19 years of age. Hormone replacement with estrogen and progesterone is resulting in regular withdrawal bleedings. A laparoscopic bilateral gonadectomy was performed at age 18, revealing the presence of a streak gonad on the left side and a streak with gonadoblastoma on the right side. The excision of the gonadoblastoma was complete and no further treatment was indicated. Some months after surgery, Ranya sends you an email message stating that she has a boyfriend and she asks you to summarize in understandable terms what kind of condition she has. Specifically, she asks you to explain: a. what kind of condition do I have? b. what is the cause? c. why did I have to undergo surgery?. d. Do I have to take medication all my life? Are there limitations to be expected regarding my general health? f. Other considerations

Statistical methods

The time fellows were logged in to the portal was registered automatically. Time spent was analysed by taking the median, as the distribution was skewed. For the calculation of the median outliers with a z-value greater than 2.68 were identified and eliminated. To compare the time spent in different parts of the world the time spent was represented in a box-and whisker plot for both cases. Whiskers represent minimum and maximum values; boxes represent first and third quartiles and the median; outliers with an absolute z-value greater than 2.68 were excluded (1 for case 1 and 2 for case 2). Apart from the registration of the time logged in to the portal fellows were asked in the web-based questionnaire to indicate the time range that the spent on both cases and how much time they spent on the answering of the questions and discussion with the expert for each separate case. The median time range was selected as being most illustrative.

For the analysis of the percentage of correct answers for multiple choice questions the percentage of correct answers on the first try was calculated. For the analysis of the open questions the number of key items that was discussed correctly in the reply was scored. A percentage was calculated of number of key items mentioned versus the total number of key items.

Results

Fifty-four experts (paediatric endocrinologists) from various parts of the world (Africa, Asia, Australia, Europe, North America, South America) were invited to participate; 41 agreed and 32 ultimately contributed. Of the 77 fellows proposed by the experts, 68 fellows participated from the following countries: Africa (5), Asia (12), Australia (4), Europe (27), North America (10), South America (10). The global distribution of participating experts and fellows is provided in Table 2.

Table 2. Distribution of the experts and fellows by continent.

	Africa	Asia	Australia	Europe	North America	South America
Experts (N=32)	1	5	2	16	5	3
Fellows (N=68)	5	12	4	27	10	10

Sixty-seven of participating fellows answered at least one of the questions for case 1; 62 fellows answered all MC questions for case 1. The median time they were logged into the portal for case 1 was 35 minutes, standard deviation 26 minutes. Sixty of the participating fellows answered at least one question for case 2; 58 completed all MC questions for case 2. The median time they were logged into the portal was 21 minutes, standard deviation 14 minutes. Fifty fellows answered all the open questions and MC questions for both cases. Time spent logged into the e-learning portal evaluating the cases and answering questions online is presented in Figure 1. The time registered by the portal system may not be a full representation of the time spent by the fellows as they may have worked additionally off-line on the cases.

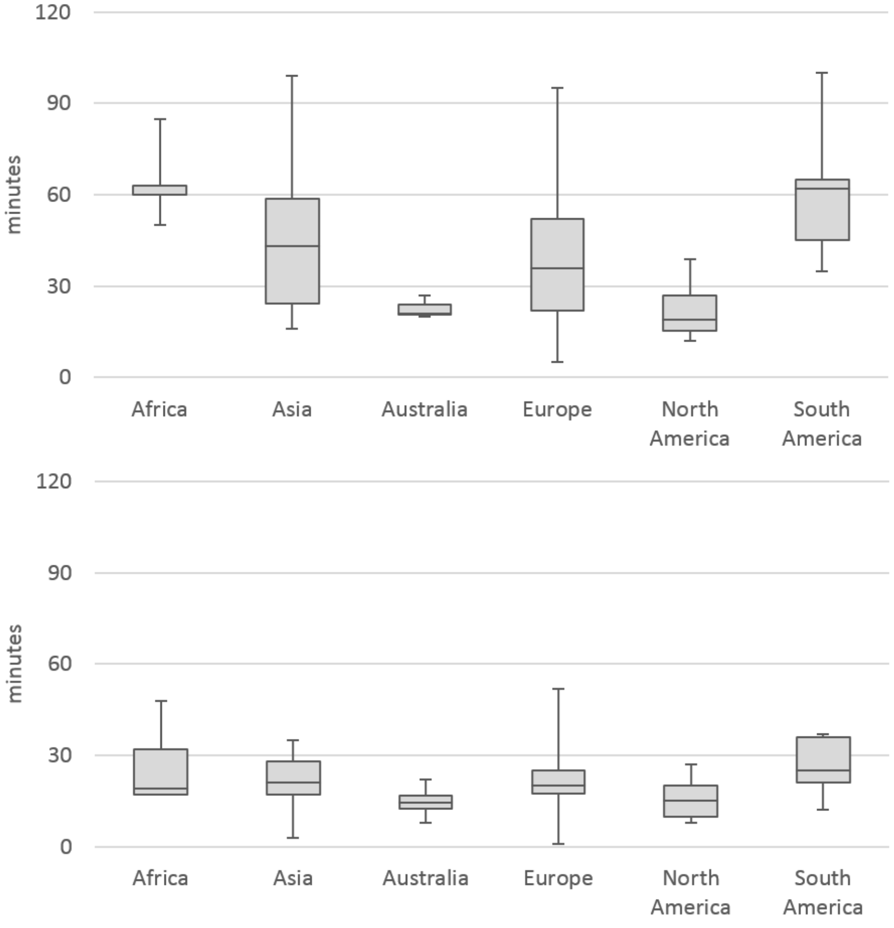

Figure 1 a and b. The time (in minutes) spent at the e-learning portal by fellows studying and responding to questions for case 1 (fig. 1a) and for case 2 (fig. 1b) is shown in box-and-whisker plots according to the continent in which the fellows worked.
Whiskers represent minimum and maximum values; boxes represent first and third quartiles and the median; outliers with an absolute z-value greater than 2.68 were excluded (1 for case 1 and 2 for case 2).

MC questions

The percentage of correct answers on the first attempt is given for each MC question for case 1 and case 2 (Figure 2). Notably, fewer than 50% of fellows achieved the correct answer on the first attempt for 16 of the 20 questions (case 1 and 2 combined); particularly low scores were observed for the questions relating to medical history (case 1: questions 1, 3, 6), differential diagnosis (case 2: questions 3, 5) and management (case 1: questions 11,13,14).

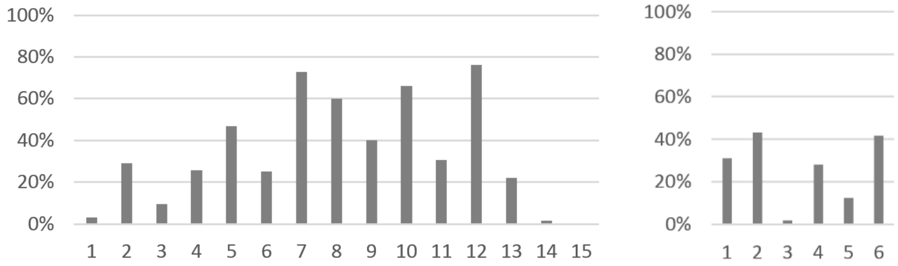

Figure 2 and b. Percentage fellows who gave the correct answer to the multiple-choice question on the first attempt.
Each question is listed by its number on the horizontal axis, fig. 2a for case 1 and fig. 2b for case 2. Note: Case 1, question15, had no correct response due to a technical error.

Open questions and key items

Sixteen experts responded to the request to identify at least 5 key elements needed for an adequate response to the open questions. Table 3 provides an overview of the key items and the number of experts naming these response elements pre-identified by the study team for each open question. While the great majority of experts noted the importance of having a diagnostic plan for both cases, only about half identified sex assignment for the newborn with congenital adrenal hyperplasia or discussion of normal and variant sex development as key items, and fewer than 40% indicated that follow-up was an important facet of the response for the young woman with gonadal dysgenesis.

Table 3. Key items mentioned by 16 experts and 50 fellows for questions 1 and 2 of case 1, and question 1 of case 2. The total number and percentage of experts and fellows naming the item is given.

	Key-items Identified a priori by Study Team	Number of Experts (% of 16 experts)	Number of Fellows (% of total 50 [case 1] and 45 [case 2])
Case 1 Question 1	1. Diagnostic plan	13 (81)	40 (80)
	2. Setting	11 (69)	36 (72)
	3. Reassurance	11 (69)	26 (52)
	4. Informing family and friends	11 (69)	13 (26)
	5. Sex assignment	9 (56)	6 (12)
	6. Explanation of normal and variant sexual development	8 (50)	11 (22)
	7. DSD team and follow up	6 (38)	10 (20)
Case 1 Question 2	1. Diagnostic plan	13 (81)	45 (90)
	2. Therapeutic options	11 (69)	34 (68)
	3. Follow up	9 (56)	9 (18)
	4. Long term implications	9 (56)	8 (16)
	5. Option of surgery	9 (56)	23 (46)
Case 2 Question 1	1. Explanation of diagnosis	15 (94)	35 (77)
	2. Long term implications	12 (75)	15 (33)
	3. Therapeutic options	13 (81)	28 (62)
	4. Setting	10 (63)	36 (80)
	5. Gonadal tumor risk	11 (69)	20 (44)
	6. Follow up	6 (38)	14 (31)

Fifty fellows responded to the open questions for case 1 and 45 fellows responded to the open questions for case 2. The number of key items mentioned in the answers of the fellows to question 1 and 2 of case 1 and question 1 of case 2 was scored by 3 members of the study team, resulting in a total score per open question. Table 3 presents the number and percentage of individual fellows naming these key items. Like the experts, the great majority of fellows identified the importance of a diagnostic plan, but few mentioned the concept of follow up. Table 4 presents the mean number of key items identified by fellows, which was less than 50% of the key elements for case 1, questions 1 and 2, and just over 50% for case 2, question 3. Finally, Figure 3 gives an overview of the percentage of correct answers for both MC and open questions per continent.

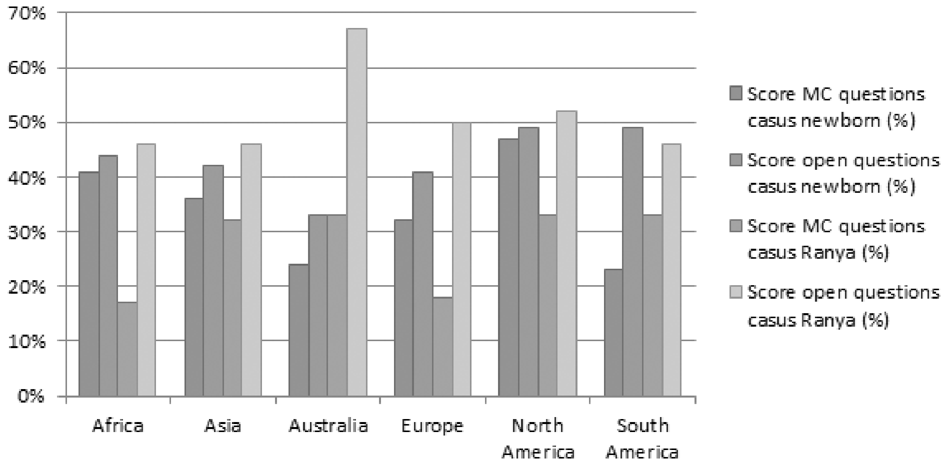

Figure 3. Percentage of correct answers on multiple choice and total score (percentage of maximum score) on open questions for each case, by continent.

Table 4. Mean (SD) number of key items mentioned by fellows compared to total number of key items.

	Total number of key items	Mean (SD) number of key items mentioned	Percentage correct
Case 1, Question 1	7	2.8 (1.4)	40%
Case 1, Question 2	5	2.3 (1.2)	46%
Case 2, Question 1	6	3.4 (1.5)	57%

Personalized feedback by multiple experts

The fellows received personalized feedback to the open questions from 2-3 experts each, one being from the country or region of the fellow and one or two from another country or continent. The experts were free to use a set of response elements prepared by the study team, see supplementary Table 1. The comments by the experts varied in terms of extent of the reply and personal input ("in my experience……."), but no regional differences could be established (data not shown).

Subsequently, the fellows were asked to acknowledge the feedback of the experts. A summary is given of the type of acknowledgement provided by 40 fellows (Table 5).

Examples of positive responses were: "I will definitely use some of the above examples and phrases in clinical practice" and "Feedback is very relevant. It gives a few points which I did not think about (like religion, informing family) and also gives useful information about the different things you have to say to the parents. The text with the 'basic' explanation is very useful and gives a structure for starting the conversation (very practical)"; "Nice to have good comment and to hear that the expert will say more or less the same."; "The suggested text about the mechanism of CAH is amazing, very didactical and clear and very able tool to explain parents about this disease. The comparison with the big river and smaller creeks is very efficient to make parents understand what happens in this disease. Thank you so much for your positive feedback."

There was only one negative reaction, stating: "All in all unfortunately not very useful, relevant or helpful comments".

It was notable that 19% of the fellows mentioned difficulties in understanding the assignment requested in question 3 for both cases, in which the fellow was asked to write a letter to the patient at the age of 17 years to explain her condition. Apparently this question was not sufficiently clear, as most of the fellows wrote a transition letter to an internist – endocrinologist to arrange follow-up of care.

Table 5. Categorization of the acknowledgement of 40 fellows to the feedback provided by the experts. Note that total number of acknowledgements is greater than total number of fellows because fellows responded to feedback from 2-3 experts per question.

Type of acknowledgement	case 1 Q 1	case 1 Q 2	case 1 Q 3	case 2 Q 1	case2 Q 2	case 2 Q 3
Total number of acknowledgements	110	108	109	97	98	98
A = thank you for suggestions; it is useful	54%	43%	38%	52%	53%	56%
A/B = thanks/ little exchange of opinion	6%	5%	4%	2%	3%	2%
B = exchange of opinion regarding content	2%	5%	6%	4%	9%	3%
C = will follow your suggestion in the future	19%	26%	13%	21%	12%	13%
D = I agree with your suggestions	9%	16%	8%	13%	11%	9%
E = need (first) feedback from my superior	1%	0%	0%	0%	0%	0%
F = had difficulty understanding the question	2%	1%	19%	2%	1%	5%
G = not useful/helpful/ relevant comments	1%	0%	0%	2%	1%	2%
H = it is nice that the expert agrees with me	0%	0%	1%	1%	0%	1%
I = No acknowledgement	6%	4%	11%	3%	10%	9%

Fellows' evaluation

Twenty of 68 participating fellows answered our web-based online demographic survey. Most fellows were in training, four participants had longer experience (5, 7, 9 and 12 years). Global distribution was as follows: Africa (2), Asia (5), Australia (2), Europe (6), North America (4), South America (1). Time spent was evaluated by 6 time ranges (0-15; 15-30; 30-60; 60-90; 90-120 minutes or other). The median time range for reading both cases and answering the questions was 90-120 minutes (selected by 8 fellows). Four fellows indicated that they spent much more time, varying from 180 minutes to two days. The median time range for studying case 1, including the answering and discussion with the expert, was between 30-60 minutes and 60-90 minutes. Four fellows indicated that they needed much more time, up to one day. The median time range for studying case 2 was also between 30-60 minutes and 60-90 minutes, with three fellows indicating that they needed much more time. Notably, these reported times are more than double the mean times spent working on the cases online, as registered by the e-learning portal, indicating that fellows undertook additional offline research and study.

Finally, the participating fellows where asked whether they thought it was useful to receive feedback from the experts. All 20 fellows answered positively. For example, one fellow stated "Yes. When all experts provided similar feedback it really sent a message that this feedback was important. It was also useful to receive varying feedback from people with different experiences. Some experts provided very detailed answers which will be helpful in my approach to patients in future." Another interesting response was "It is very useful to receive feedback from various experts because each expert has his one way in dealing with the problem. And when different experts give their opinion you can use a little bit from every answer. Also some experts find that certain part of the explanation has to be better while some find it good, because of the difference in personal interest."

Cultural, religious and regional aspects

Experts were asked to indicate cultural, religious and regional items of relevance in the answers to the open questions. Twelve of 32 experts replied to this request and mentioned these aspects summarized in table 6.

Table 6. Regional, cultural and religious aspects of DSD management reported by 12 experts

Regional, cultural, religious aspects	Number of *experts*
Fertility	8 (12)
Role of family	6 (12)
Role of religious official	6 (12
Genital surgery	5 (12)
Sex assignment	6 (12)
Hormonal replacement	4 (12)
Role of karyotype	4 (12)
Sex registration	4 (12)
Follow up	4 (12)
Association with transgender	3 (12)

The most frequently mentioned items were fertility and the role of the family. In some cultures the girl will visit the doctor only with the mother without involvement of the father. In other cultures the family will demand that the patient is not addressed directly - at least not until she is 18 years old. The possibility for delayed sex registration differs per country. Sometimes sex assignment can be postponed, but this may be unacceptable to families. Several experts expressed a strong preference for male sex assignment for instance in 46,XX newborns with congenital adrenal hyperplasia with severe virilization. Some experts mentioned that possibilities for follow up are limited for socioeconomic reasons or due to lack of confidence in the health care system.

Discussion

In this exploratory study we evaluated the applicability and feasibility at a global level of the ESPE e-learning portal in assessing junior paediatric ians (fellows) during their subspecialty paediatric endocrinology training, regarding knowledge and skills for investigation and management of children and adolescents with DSD. In general, learning in practice is regarded as the most powerful way of learning. However, even in major referral Centres the number of patients with DSD is small, thus exposure to the clinical management of newly referred patients is very limited. Therefore, there is also a strong need for alternative learning methods to train the necessary competencies for a medical specialist as described by the CanMEDS framework. [8][9]. E-learning is increasingly being used to train competencies such as 'Scholar' and 'Medical Expert' [7]. The responses to the MC questions provided some insight into the 'Medical Expert' competency. Although not formally validated, these questions refer to current textbook knowledge. The baseline knowledge with respect to relevant history, differential diagnosis, and management of the participating fellows was notably limited judging on the score of the MC questions.

During this study knowledge is gathered by formative questions in the e-learning and by feedback provided by the experts. Concluding from the acknowledgements from the fellows and from the online questionnaire amongst the fellows, this leads to extended skill of 'Medical Expert'. This argues in favour of the integration of formative learning by e-learning in the training for paediatric endocrinologist.

To our knowledge the use of e-learning to train other competencies such as 'Communicator' and 'Collaborator' is not common practice. This study shows, however, that e-learning with personal feedback offers possibilities to train these competencies. In principal the portal system allows direct interaction between fellow and tutor/expert. As the emphasis is on formative assessment the tutor is in a position to correct any misunderstanding of the assignment or to request further details. The management of DSD requires an interdisciplinary approach, and in this study we focused on the role of the paediatric endocrinologist in the team. It is of note that many key items in the open questions considered by the experts to be an essential part of the set of response elements, were not mentioned by the fellows. This can be explained by the low exposure to such complex cases in practice due to the limited number of patients, highlighting the potential role for and value of e-learning.

The results of our study support the use of e-learning for formative assessment, providing direct feedback (assessment *for* learning). The anonymity between the fellow and faculty may have a positive aspect. Fellows may be anxious or uncomfortable discussing patients with their mentors and other senior faculty members. This study allowed them to write their personal thoughts and opinions without the anxiety of presenting to a respected faculty mentor. Indeed, the personalized comments by the experts proved to be very formative, as the fellows indicated that they learned a great deal from the personalized feedback. They were confronted with certain aspects that they were not aware of, such as religious and cultural aspects and addressing sensitive issues. Moreover, it was gratifying to note that most fellows in the subsequent acknowledgement expressed their appreciation. When feedback is provided in a summative context, for instance as a test or exam with a pass/fail score (assessment *of* learning) it is not always used effectively by learners, which pleads for a formative context. [12][13].

The time spent to study the assigned clinical cases did vary. Fellows from South America, Africa and Asia took more time for both cases than fellows from other continents, presumably because of the cases and questions were presented in English, which would not be the native language of those fellows.

This study aimed at demonstrating the applicability of e-learning at a global level, and identifying regional and cultural differences in medical practice. A subset of experts replied to the request to indicate relevant cultural, religious and regional items. The most frequently mentioned item was fertility and the role of the family. In some cultures fertility is extremely important, as it affects 'marriageability' for women. Egg cell donation may not be allowed and adoption might be uncommon [14][15][16]. A religious official may play a crucial role in management in terms of decision making and with respect to openness on informing family and friends [15][16]. Options regarding genital surgery involving gonadectomy, hysterectomy or fertility preservation procedures may be subject to religious recommendations [17]. In some countries management of DSD is seriously hampered by lack of access to essential medications such as hydrocortisone and to specialized (paediatric) urosurgical expertise [14][18]. Furthermore, recommendations for genetic counseling and transition of care to an adult endocrinologist or gynaecologist are lacking. Several experts mentioned the association with transgenderism or homosexuality, which in some cultural/religious background is condemned [15][16]. It is of interest to note that the fellows in their first reply to the open questions did not mention cultural, religious or regional aspects. However, some acknowledged in their second reply to the expert that religious considerations are important. From the acknowledgements given by the fellows we conclude that they achieved new insights on these aspects for future use. It was also highly appreciated that experts from different continents responded to their replies to the open questions. Although preformatted reply text was supplied by the study team, the participating experts indicated that a careful reply to the fellows' answers to the open questions was time consuming.

As a limitation of the study it is acknowledged that English is not the native language of many participating fellows, which may have influenced their understanding of the questions and formulation of their replies. Regional or national application of the e-learning portal may make the best use of the direct interaction between fellow (student) and expert (tutor).

In a separate study the approach and the wording chosen by the fellow informing parents of a newborn with DSD and an adolescent girl with DSD will be assessed in detail.

In conclusion, in this exploratory study we demonstrated at a global level the applicability of the ESPE e-learning portal in assessing junior paediatric ians during their subspecialty training in paediatric endocrinology regarding knowledge and skills for investigation and management of children and adolescents with DSD. The results indicate that there is limited knowledge with regard to the management of newborns and adolescents with DSD. E-learning with formative feedback by experts is not only appreciated by trainee subspecialists but also may have an important role in their clinical training.

References

1. Sleator RD. The evolution of eLearning background, blends and blackboard.... Sci Prog. 2010;93(Pt 3):319-34.
2. Wu B, Wang M, Johnson JM, Grotzer TA. Improving the learning of clinical reasoning through computer-based cognitive representation. Med Educ Online. 2014 Dec 16;19:25940.
3. Grijpink-van den Biggelaar K, Drop SL, Schuwirth L. Development of an e-learning portal for paediatric endocrinology: educational considerations. Horm Res Paediatr. 2010;73(3):223-30.
4. Kranenburg-van Koppen L.J.C, Grijpink-van den Biggelaar K, Drop SL. An interactive E-Learning Portal in Paediatric Endocrinology: Practical Experience. Pharmacy 2013, 1, 160-171
5. Drop Sl, Kranenburg L, Reerds S, Grijpink K, Iotova I. The Espe elearning webportal : a global tool for the instruction and formative assessment of paediatric endocrinology. Scripta Scientifica Medica 2014;46:11-18.
6. Lee PA, Nordenström A, Houk CP, Ahmed SF, Auchus R, Baratz A, Baratz Dalke K, Liao LM, Lin-Su K, Looijenga LH 3rd, Mazur T, Meyer-Bahlburg HF, Mouriquand P,Quigley CA, Sandberg DE, Vilain E, Witchel S; Global DSD Update Consortium. Global Disorders of Sex Development Update since 2006: Perceptions, Approach and Care. Horm Res Paediatr. 2016;85(3):158-80.
7. Busari JO, Stammen LA, Gennissen LM, Moonen RM. Evaluating medical residents as managers of care: a critical appraisal of assessment methods. Adv Med EducPract. 2014 Feb 20;5:27-37.
8. Van der Lee N, Fokkema JP, Westerman M, Driessen EW, van der Vleuten CP,Scherpbier AJ, Scheele F. The CanMEDS framework: relevant but not quite the whole story. Med Teach. 2013 Nov;35(11):949-55.
9. Yip HK, Smales RJ. Review of competency-based education in dentistry. Br Dent J. 2000 Sep 23;189(6):324-6.
10. Nordenström A, Thyen U. Improving the communication of healthcare professionals with affected children and adolescents. Endocr Dev. 2014;27:113-27.
11. Muscarella M, Kranenburg-van Koppen L, Grijpink-van den Biggelaar K, Drop SL. Global application of disorders of sex development-related electronic resources: e-learning, e-consultation and e-information sharing. Endocr Dev. 2014;27:268-83.

12 Schuwirth L, Ash J. Assessing tomorrow's learners: in competency-based education only a radically different holistic method of assessment will work. Six things we could forget. Med Teach. 2013 Jul;35(7):555-9.

13 Harrison CJ, Könings KD, Schuwirth L, Wass V, van der Vleuten C. Barriers to the uptake and use of feedback in the context of summative assessment. Adv Health Sci Educ Theory Pract. 2015 Mar;20(1):229-45.

14. Raza J, Mazen I. Achieving diagnostic certainty in resource-limited settings. Endocr Dev. 2014;27:257-67.
15. Ediati A, Juniarto AZ, Birnie E, Drop SL, Faradz SM, Dessens AB. Gender Development in Indonesian Children, Adolescents, and Adults with Disorders of Sex Development. Arch Sex Behav. 2015 Jul;44(5):1339-61.

16 Ediati A, Faradz SM, Juniarto AZ, van der Ende J, Drop SL, Dessens AB. Emotional and behavioral problems in late-identified Indonesian patients with disorders of sex development. J Psychosom Res. 2015 Jul;79(1):76-84.

17. Al Herbish AS, Al Jurayyan NA, Abo Bakr AM, Abdullah MA, Al Husain M, Al Rabeah AA, Patel PJ, Jawad A, Al Samarrai AI. Sex reassignment: A challenging problem - current medical and islamic guidelines. Ann Saudi Med. 1996;16(1):12-5.

18. Juniarto Z, van der Zwan YG, Santosa A, Ariani MD, Eggers S, Hersmus R, Themmen AP, Bruggenwirth HT, Wolffenbuttel KP, Sinclair A, White SJ, Looijenga LH, de Jong FH, Faradz SM, Drop SL. Hormonal evaluation in relation to phenotype and genotype in 286 patients with a Disorder of Sex Development from Indonesia.Clin Endocrinol (Oxf). 2016 Mar 3.

Appendix I

This appendix contains the information prepared by study team for the expert paediatric endocrinologist to be used in his/her reply to the fellow(s).

It contains the three open questions of the two provided cases with the answer model when prepared. Furthermore, suggestions for the approach to sharing diagnostic information with parents is provided for question 2 of case 1 A newborn with ambiguous genitalia (congenital adrenal hyperplasia) and additional information is included regarding wording and a communication model for a dialogue which can be used for the open question 2 from case 2 Ranya, a 17-year old girl with delayed puberty.

Case 1. A newborn with ambiguous genitalia (congenital adrenal hyperplasia)

Question 1.

You are the pediatric endocrinology fellow in charge: you have obtained the medical history information and you have examined the baby. What do you tell the parents at this stage?

Elements to include in the answer:

- The setting: private room; no interruption by colleagues, personnel etc.; no beepers, mobile phones. Jointly with medical team psychologist, if available?
- Time allotted: 30-45 minutes
- Introduction of DSD team; one spokesperson
- Explanation of initial findings:
  a. your baby looks healthy and is in no distress. The skin is nicely pink; the baby is breathing normally, the heart beat and blood pressure are normal.
  b. your baby's genitals do look not typical and therefore we cannot see from the outside whether your baby is a boy or a girl.
  c. there are a number of medical conditions that can be associated with this type of difference in development. We will need to do a number of tests to get some information, but we while we are doing so we will make sure that your baby remains healthy, by watching things carefully.
  d. sex of rearing: no sex of rearing assignment until results of investigations are available; registration at municipal office? In some countries it is now possible to register an infant's birth with an "indefinite" or "unknown" sex.
  e. informing extended family; only closest relatives and friends. It is your decision regarding who of your family and friends you decide to tell about this aspect of your baby's development and that your baby required further medical assessment/tests. We understand that this uncertainty is very difficult to manage

and that this time is a very difficult time for you. The people close to you might be able to support you at this difficult time. However, because you are dealing with aspects of your baby's development that are personal (i.e. regarding the genitals) and difficult to understand, you may choose to limit this knowledge to those people in your life that you trust and that you think your child would learn to trust.

f. diagnostic plans: echography; hormone determinations, karyotype, genetic testing, possibly cystoscopy or other radiography of lower urogenital tract
g. other considerations

Note: Give time for questions and concerns of parents

Note: consult religious officials?

Question 2

You have just made the diagnosis of congenital adrenal hyperplasia (CAH), most likely as a result of 21-hydroxylase deficiency. Give a detailed account of how you will inform the parents of the diagnosis

Elements to be included in explanation of the diagnosis:

a. adrenal disorder termed congenital adrenal hyperplasia, abbreviated as "CAH" with prenatally increased levels of "male-type" hormones, androgens
b. uterus and ovaries present
c. corticosteroid treatment (life-long) will stop virilisation.
d. salt loss requires fludrocortisone treatment (life-long)
e. Hormonal treatment is lifelong; normal life expectancy; fertility is possible.
f. Follow up: regular follow up for adjustment of dosage of medication and reminder of instructions for administration of stress medication is essential; discuss importance of adherence to prescribed treatment and consequences of non-adherence.
g. Long-term implications: the clitoris is expected to become less noticeable as your daughter grows; we will make a joint appointment with the surgeon (or urologist or gynaecologist) to check on this in about 3-4 months to discuss whether or not genital surgery is an option.
h. Explanation of the genetics of CAH and risk for future pregnancies, also options for pre-implantation genetic diagnosis.
i. Other considerations

Note: in some institutions this will be done by genetics consultant or counsellor rather than endocrinologist. Need to raise the issues that must be discussed, and list options for who will undertake this discussion.

Note: provide time for parents' questions and concerns

Note: how will you check whether the mother and father understand the main messages, such as the diagnosis and therapeutic consequences, adjustment of medication dosages, and stress medication?

Note: will you provide a written summary of the discussion?

Note: will you offer to arrange a meeting between this family and another family in your clinic who are caring successfully for a child with CAH?

Note: will you provide any reading materials published by support groups, or contact information for local or online support groups?

Suggestions for approach to sharing diagnostic information with parents

"As you know, we did a number of tests on your baby and we have some results to share with you. The first test we did was an ultrasound of the groin area to look at what kind of reproductive structures are present, and we found that your baby does have a uterus and ovaries, as are usually found in girls. So although your baby's external structures developed differently, she has the internal structures of a girl.

The reason that the external genitals developed differently is because your daughter most likely has a condition called congenital adrenal hyperplasia, abbreviated as "CAH" and I will show you with these diagrams what this means for her [show drawings of hypothalamic-pituitary-adrenal axis and describe]. The adrenal glands are a pair of triangular-shaped structures present in both boys and girls that sit up above the kidneys in the back of the abdomen about here (point to location on diagram or body) and are responsible for making 3 types of important hormones. (Using a picture of one adrenal gland, demonstrate adrenal zones and describe).

One part of the gland makes the hormone cortisol – you have probably heard of this; this hormone is very important for helping a person keep healthy, maintaining adequate energy supply for the body, and responding to physical stress such as illness, infection or injury.

Another part of the adrenal gland makes a hormone called aldosterone which is critical for helping the kidneys to keep the right level of salt in this body and keeping the blood pressure normal.

The 3rd part of the adrenal gland makes usually fairly small amounts of mild "male-type" hormones called androgens in both boys and girls.

All of these hormones in the 3 parts of the adrenal gland are made by a number of enzymes within the cells causing a series of chemical reactions; it's a bit like carving a sculpture, where you start with the raw material – a block of wood or stone – then step by step you work on it with the right tools, until you have the finished product. In this condition of CAH that your baby has, one of the enzymes required for production of cortisol is reduced or absent, so the pathway to make cortisol is obstructed. Think of this like what happens if water is flowing down a big river, with smaller creeks running off to the side. If a dam is put up across the big river, the water has to go somewhere, so the path it will take will be down the smaller creeks off the side, making those creeks bigger and bigger over time. This is like what happens in CAH: when the cortisol-making pathway is blocked the hormones have to take another pathway, so this results in low levels of cortisol, and increased levels of the male-type hormones called androgens. The increased amounts of male-type hormones caused your baby's external genitals to develop some more male-type features during the early stages of her development before birth. CAH can vary in how much of each of the hormones are made, so some children/individuals with CAH are affected more severely than others. The more severe form of CAH is called "salt-wasting" because the individual passes lots of extra salt into the urine, and the less severe form is called "simple" CAH, because salt balance is not affected to a significant extent.

CAH is a fairly common genetic condition, affecting about 1 in 15,000 individuals. So this means that in [city or town name], there are about [NN; calculate number based on population] other people with this condition. In fact we see quite a few children with the condition in our clinic here at the hospital, so could introduce to another family at some point if this would be helpful to you. CAH affects girls and boys, women and men equally, although it is usually more noticeable in girls because of the effects of the androgen hormones. It is a life-long condition, but we have experience with treating this. You daughter will need to take hormones to replace the ones she can't make enough of for herself, and we will need to see her regularly in our clinic to check that the hormones are balanced to keep her healthy.

Question 3

Many years later, when Eve is 17 years of age, you prepare a letter for transition of her medical care to an internist-endocrinologist and gynaecologist. You provide her with a written summary of her medical history. Specifically you explain once more in understandable terms:

- what kind of condition she has;
- what happened before she was born;
- why she had operations;
- why she is being referred to an internist-endocrinologist / gynaecologist for ongoing care
- whether she is required to take medication all her life
- whether she will be able to bear children in the typical way and if so, the risk is that one of her children will also be affected

Prepare this letter.

No answer model provided by the study team for this question.

Case 2: Ranya, a 17-year old girl with delayed puberty

Question 1

You have made the diagnosis 46,XY gonadal dysgenesis. Give a detailed account of how you will arrange the sharing of information to Ranya and her parents. Consider the following elements: the setting; allotted time; key elements to be discussed regarding the diagnosis; relevant therapeutic issues; long-term implications; follow up; other considerations

Elements to be included:

- The setting: make an appointment with Ranya and both parents; private room; no interruption by colleagues, personnel etc.; no beepers or mobile phones. Possibly jointly with medical team psychologist or social worker if one is available with experience with DSD.
- Please note: Parents should be invited by arrangement with Ranya. Most often teenagers will agree to one or both parents being present but it may be that she would prefer to be spoken to either alone or with one parent present. If Ranya elects to receive new information alone, then the Doctor should arrange to invite the parents into the consulting room afterwards to present a summary of the information and discussion that Ranya has agreed to.
- Time allotted: 45-60 minutes; ensure that patient/family is aware of the time allotment. If necessary, follow up in following days/week.

- Please note: The important aspect regarding timing is that the patient is aware of how long the consultation will be. If they usually have 15 minutes they may inhibit questions unless they know that this is timetabled as an extended consultation.
- Explanation of the diagnosis. Key elements to be discussed (not necessarily in specific order):
  a. Typical development of ovaries/gonads and atypical development in gonadal dysgenesis
  b. Genes and chromosomes
  c. Hormones and hormone replacement
  d. Female reproductive anatomy
  e. Fertility/infertility
  f. Potential tumor risk
- Therapeutic issues: Hormone replacement during the teenage years, (estrogen, and later progesterone) will help Ranya to go through the physical changes of puberty in a similar pattern to other girls. These hormones can be provided in many ways (pills, patches, sprays, creams, etc.) and have to be continued into adulthood to provide Ranya with the hormonal environment of a typical young woman.
- Implications: sexual relationships; infertility; options to have a partner, a family.
- Follow up: outpatient clinic appointment for initiation of estrogen treatment; subsequent joint appointment with urologist/surgeon or gynecologist.
- Other considerations

Please note: In many countries women worry about risks associated with long-term use of estrogens (following media coverage of adverse events associated with long term use of the oral contraceptive). Therefore, it may be necessary to explain that in Ranya's case, therapeutic hormone use replaces typical hormone secretion, and does not constitute higher than usual level of long-term estrogen exposure.

Note: Provide ample time for discussion and response to questions and concerns of patient and parents.

Note: How will you check whether Ranya, her mother and her father understand the main messages?

Note: Will you provide a written summary of the discussion?

Note: Will you provide any reading materials published by support groups, or contact information for local or online support groups?

Question 2

Give a detailed account of how you will inform Ranya and her parents of the increased risk for development of a gonadal germ cell tumor and the therapeutic options. Consider the following elements: relative risk; time frame; therapeutic procedures and timing; follow up; other considerations.

Answer model as provided below is presented in the form of a monologue, as if the physician is speaking directly to Ranya. A subsequent version (see page 9) provides the information in the form of a dialogue.

- Risk: In girls with a 46,XY karyotype, there is an increased risk for the development of a tumor of the gonads over time. The relative risk for tumor development varies with the specific diagnosis. If this tumor develops, this occurs usually in early adulthood, before the age of 40 years. Currently, no one can tell exactly how high this risk is, but estimates, based on recent reviews of cases reported over many years, are around 30%, maybe even a bit more. Unfortunately, there are no reliable methods, such as ultrasound or MRI or specific markers that can be used to reliably detect this tumor at an early stage. However, one clinic in the United Kingdom has been using MRI to check the status of the gonads in women with a different DSD called complete androgen insensitivity syndrome (Nakhal RS et al. 2013).
- Procedure: This surgery is called "gonadectomy"; it is a short procedure with limited risks, which will be discussed with you in more detail when you meet with the surgeon (urologist, gynecologist or general surgeon). The surgery can usually be performed via a small, flexible instrument called a laparoscope, which is inserted through a small incision in the side of your abdomen; this typically requires just short hospital stay of a day or so.
- Timing: You don't need to decide right away, but because these tumors typically occur soon after puberty or in young adults, it is best not to postpone this decision for many years. Should you decide to have this surgery done, then perhaps you can indicate us when would be good timing for you within the coming year, taking into account your school and holiday plans, so we can arrange things to be as convenient as possible for you.
- Other considerations:
  a. Give time for questions and concerns of patient and parents
  b. How will you check whether Ranya, her mother and her father each understand the main messages?
  c. Will you provide a written summary of the discussion?
  d. Will you offer to put parents / patients in direct contact with other families in similar circumstances, or to provide help through a support group?

Question 3

Ranya is now 19 years of age. Hormone replacement with estrogen and progesterone is resulting in regular withdrawal bleedings. A laparoscopic bilateral gonadectomy was performed at age 18, revealing the presence of a streak gonad on the left side and a streak with gonadoblastoma on the right side. The excision of the gonadoblastoma was complete and no further treatment was indicated.

Some months after surgery, Ranya sends you an email message stating that she has a boyfriend and she asks you to summarize in understandable terms what kind of condition she has. Specifically, she asks you to explain:

- What kind of condition do I have?
- What is the cause?
- Why did I have to undergo surgery?
- Do I have to take medication all my life?e. Are there limitations to be ecxpected regarding my general health?
- Other considerations

No answer model provided by the study team for this question.

Additional information - Some overarching comments regarding general approach

1. Focus the conversation on Ranya, explaining in the beginning to her parents (if present in the room) that you will be talking directly to their daughter, but of course they are welcome to ask questions, as is Ranya herself. Offer Ranya the opportunity for private discussion with her parents out of the room.
2. Stop after each concept to recheck understanding and allow Ranya and the family to ask questions.
3. Explain at the beginning that you will have 60 minutes (or whatever time limit is set) of uninterrupted time focused just on Ranya, but that it will be necessary to set up an additional appointment to follow up, as there will no doubt be some questions that will come up in the family’s minds after the discussion.
4. Begin with reassuring her that she is healthy, but that her situation is complicated, and that is why it will take some time to explain and answer questions. She should be reassured that there is no pressure to make any decisions about any of the information she receives, and she will have time to think about things and ask any questions she wishes.
5. Explain that a summary of the conversation will be provided, but they are also welcome to take notes if they would like or they are welcome to record the discussion to listen to it again when they get home.

6. Begin by ascertaining what Ranya knows or understands at this point and what she would like to learn from you in this discussion. One assumes that some initial discussion must have occurred during the first consultation, when arrangements were made to perform a number of investigations. One assumes that possible diagnoses were mentioned at the time the tests were initially done. One may also assume that Ranya and her parents have been discussing this in the time since her first visit, and that one or all of them have been looking up information online. Ask what they have read and whether this has helped or confused them.
7. Understand what Ranya's specific concerns may be, and address these in order of her personal priority early in the discussion. For example, she may be most concerned about whether she will have breast development, whether she is actually a girl, whether she will be able to have sexual intercourse, whether she is at risk for cancer, or whether she will be able to have children via the usual mechanisms. If she is significantly concerned about one particular issue, clearing this away first should help her to get most information from the rest of the conversation.
8. Once initial concerns are addressed, you may want to put some structure around how the information is packaged – the "What", "Why" and "Plan" approach. However, in most real-life situations, the conversation becomes quite organic and will probably not strictly adhere to such structure. Do not avoid use of medical terminology, but make sure that any words that are likely to be unfamiliar are explained during discussion.
9. Avoid language that expresses urgency, fear, danger, abnormality or disease or is otherwise scary. Choice of words should be made carefully; she should be made to feel positive about her body and who she is, and that she is not alone in her differences in development. Nevertheless, acknowledge that she is dealing with a lot of difficult information, and it is natural to be concerned or confused.
10. It is important not to dismiss or brush off any concerns she may have, even if they may seem relatively minor in the scheme of things.
11. Whether to include a psychologist at this discussion is a point for further exploration and discussion on an individual case basis. While there is value to this in many cases, not all endocrinologists have access to a psychologist to be present during such appointments. Furthermore, if a psychologist is included in this appointment, it is important to ask Ranya and her parents if they are willing to have the psychologist present, or else to in some way explain why the psychologist is there (for example: "We always include our psychologist Doctor Jones during these discussions, because some of the information may cause you to feel anxious or concerned, and she can help you to understand those feelings).

Additional - Suggestions for how to address Ranya and her parents (monologue scenario)

“Hello Ranya and Mr and Mrs X:

You will remember that you came to see me a few weeks ago to look into why you haven’t had your period. And as you know, we did a number of tests to try to find out the reason. So I have some results of those tests to share with you.

First and most importantly, I want to reassure you that you are quite healthy and there is no serious problem to be concerned about.

What we have learned is that your body was made a bit differently. Everyone has differences – like you have black hair and your dad has blonde hair. Those are common differences that are easily seen, but there can be other differences too, that are less common, and can’t be seen from the outside. Your differences are on the inside, and affect how your ovaries developed. Do you remember studying in biology about ovaries and testes? Ovaries are usually present in most girls, located inside the tummy, down toward the bottom [point to general location]. They are about the size of an olive/ a grape [use alternative example as culturally appropriate] and they carry the egg cells that women need to make babies. They also make chemicals called “hormones” that circulate around in the blood and help girls develop at puberty and cause the uterus to have monthly periods. You didn’t develop ovaries, so you don’t have the egg cells to make babies, or the hormones that are needed to develop breasts.

I expect this comes as a shock to you, however it is really important for you to understand that these differences don’t change the fact that you are a girl, and you will develop into a woman. Probably the most important point in all of this for you, is that because you don’t have ovaries you will probably not be able to have babies in the usual way that most women have babies. But there are many ways for women to have children, and that is something we can provide information on later [when you are older]. But for now, we need to talk about giving you some medicine to help you with development of your breasts like your friends at school, and to develop your uterus, to keep this healthy so that you will eventually have periods. This will all take a bit of time, and we will need to talk again about the steps along the way

.

The first thing to know, is that when the ovaries don’t develop completely, as yours didn’t, they are usually called “streak gonads” or “streaks”. I am mentioning this so you understand the words that you might see in the medical records, or if you look any of this up online.

But to help you understand all of this, first let me explain what chromosomes and genes are. (See also response 2d,). Each person usually has 46 chromosomes; they get half of their chromosomes from each parent (i.e. 23 from mother and 23 from father). We usually get one 'X' chromosome from our mothers, and an 'X' or 'Y' chromosome from our fathers.A person's chromosome pattern can also be called their "karyotype" (e.g. 46,XY, meaning 46 total chromosomes including an X chromosome and a Y chromosome, or 46,XX, meaning 46 total chromosomes including 2 X chromosomes,).There are various combinations of chromosomes in humans: many girls with 46,XX; some girls with 46,XY;some girls with 45,X(meaning that they are missing one whole chromosome);many boys with 46,XY; some boys with 46,XX; some boys and girls with 46,XY/46,XX (meaning that some cells have one chromosome pattern, and other cells have a different pattern);some boys and girls with 45,X/46,XY. In your case, you inherited an X chromosome from your mother and a Y chromosome from your father, so your chromosome pattern is 46,XY.

Genes: Chromosomes are made up of many segments called genes which carry genetic codes (DNA) that influence many things, such as the colour of the eyes. Genes can influence sex development, and changes in specific genes can sometimes be the reason for a DSD.

In girls who have a Y chromosome, there can be an increased risk for the development of a tumorof the gonads over time. If this tumor develops, this is thought to most likely occur usually in early adulthood, before the age of 40 years. Currently, no one can tell you exactly how high this risk is, but it is estimated that this is about a 1 in 3 chance of developing a tumor over a lifetime for women who have gonadal dysgenesis. This means that if you live to be 70 or 80, you might have about a 1-in-3 chance of developing a tumor. At your current age of 17 your risk is not high, but as you get older, into your 20s, 30s and 40s, we think that the risk for developing a tumor will increase. So we need to discuss what to do about this eventually, but there is no rush to make a decision right now. Unfortunately, there are at present no reliable methods, such as ultrasound or MRI or specific markers in your blood that we can use to detect this cancer a tumor at an early stage. But nevertheless, if you choose to keep your gonads for a while we will do the best we can to watch for any changes, with blood tests and ultrasound or MRI scans. There is no guarantee that we will be able to pick up a problem at an early stage, but we will do our best.

Because of this risk of a tumor, at some point you will need to think about whether to have your gonads removed in a procedure called "gonadectomy", which just means "gonad removal". Either way, whether you keep your gonads for a while or remove them soon, you will need hormone treatment to make sure that you have periods to keep your uterus healthy. The hormone treatment is also needed to keep your bones strong, and to help protect your heart, so you will need hormone treatment (which can be given in a variety of ways that we can discuss in more detail at your next visit) throughout your teenage years and adult life.

Because the gonads do not produce estrogens or eggs, meaning that the gonads do not function in your body, you will need to take estrogen, which can be given in various ways, to help you go develop breasts and eventually mature the uterus so you have periods. .

Procedure: This surgery is called "gonadectomy"; it is a short procedure with limited risks, which will be discussed with you in more detail when you meet with the surgeon. It can be performed via a laparoscope (a small, flexible instrument inserted into the abdomen through a small incision) that has a camera so the surgeon can look inside). You will not need to stay in the hospital for a long time, and you will be able to restart your daily activities soon afterward. If/when you decide to go ahead with surgery, we can organize an appointment for you with the surgeon to give you more information about the procedure. Of course then you can think about this for a while, before you make a final decision and we are always available if you have questions.

Timing: You don't need to decide right away, but because these tumors typically occur soon after puberty or in young adults, it is best not to postpone this decision for many years. If you have decide to have this surgery done, then perhaps you can indicate to us when would be good timing for you within the coming year, taking into account your school and holiday plans, so we can arrange things for you to be as convenient as possible for you.

Additional - Suggestions for how to address Ranya and her parents (dialogue scenario)

In this scenario, the doctor offers Ranya the opportunity to ask questions that may be of importance to her and uses her questions to guide the conversation and information sharing.

1. Examples are provided below of possible language to use in response to some particular concerns Ranya may raise:

a. Ranya: I am worried that I am not going to be able to have children.

Doctor: I understand your concern, and will answer you honestly. It is likely that you will not be able to have children in the typical way that most women do. And I will explain why that is so a bit later. However, it is important for you to know that there are many ways to have a family, and we can provide you with more information on what options might be available for you to have children.

b. Ranya: Am I still a girl?

Doctor: I can understand how you might be worried about this, but I want to reassure you that you are indeed a girl, although your road to developing as girl has been a bit different from some of your friends, as I will explain later. Do you have any concerns about whether you are a girl? [Her response to this question may open further avenues for clarification].

c. Ranya: I am worried that my chest is going to stay flat forever and I will always look like a child.

Doctor: I understand your concern, and we can help with this. Our test results showed that your body is not making any of the main female hormone called estrogen. Do you know what hormones are? These are chemicals produced by certain organs in the body that work on other parts of the body to make them grow and develop. Estrogen is the hormone that causes the breasts to develop as a girl goes through puberty. It is also one of the hormones that is responsible for the uterus developing and for having periods, and also has other important roles in keeping the body healthy. Because your body doesn't make estrogen you will need to receive this hormone as a medication. This treatment will make your breasts grow and eventually will also make your uterus develop so that you will have periods. We can talk next time about what kind of options there are for this hormone treatment and what are the best and worst aspects (pluses and minuses) of each kind of treatment.

d. Ranya: I am worried that I have cancer and I am going to die.

Doctor: I understand your concern, and I want to reassure you that you are quite healthy and there is no evidence that you have cancer. We will talk a bit later about what we will do to prevent that from happening.

2. Once Ranya's key question(s)/concern(s) has/have been addressed, the doctor can move on to a more systematic discussion of the diagnosis. Suggested scenario:

a. Doctor: So now that we have talked about some of your most important questions, I would like to try to explain why you have developed differently from most of your friends. But first I want to make sure you know that there are a lot of other girls like you who developed in this way. So first, let's talk about ovaries and what they do and how they develop. Do you know what the ovaries are Ranya, and where they are usually found in the body?

Ranya: I think they taught us about that in biology class last year, but I have forgotten, so can you please explain for me?

Doctor: The ovaries are 2 structures usually shaped like small eggs – maybe the size of a grape - [use alternative example as culturally appropriate], located in most girls and women down here in the groin area, one on each side (point to this location on a drawing or on a person). They usually produce the egg cells that are needed to make babies, and also the hormones – those chemicals that I mentioned earlier – that cause breasts to develop and periods to happen. In some girls the ovaries don't develop completely for one reason or another, and this is what happened in your case. As I mentioned, there are quite a lot of girls who have underdeveloped ovaries; these are sometimes called "streaks" or "streak gonads" because instead of looking like a small egg, they look like a small piece of cartilage or gristle. In fact, about one out of every 2000 girl babies is born with underdeveloped ovaries. This means that here in [name of city] there are about [NNN; calculate number based on population of city/town] other girls who have underdeveloped ovaries, and I see quite a few such girls here in my clinic. If you would like to meet one of the other girls I look after, I can work on arranging this.

b. Ranya: What happened to my ovaries?

Doctor: This is a good question, and I'll explain by first describing how babies typically develop, and why you took a slightly different path. All babies, whether they are going to develop as a boy or a girl, start out during their development inside their mother's uterus, with a pair of structures inside the abdomen called "gonads". Gonads is just a word describing the type of tissue that eventually in most cases develops into ovaries in girls and testicles or testes in boys. You probably heard these words in biology class in 5th grade. Boy and girl babies have gonads that look exactly the same during about the first 2 months of the baby's development. After that time, a whole lot of genes carrying the baby's DNA instruct the gonads to develop as ovaries in most girls, and as testes in most boys. Do you know what genes and chromosomes are?

Ranya: I think so, because we learned that in biology in 6th grade. I think genes are tiny packets of DNA inside our body's cells that carry the body's instruction manual. We have different genes that instruct cells of our body to do a certain job so that we have particular characteristics, like brown eyes or blonde hair or big feet.

Doctor: That's right, and you might also remember that DNA is the chemical structure that basically represents the words in the instructions given by the genes. The genes are carried on tiny thread-like structures called chromosomes inside each cell of the body. When the gonads are developing, a certain set of genes instructs them to develop into ovaries, while a somewhat different set of genes instructs the gonads to develop as testes. In situations when the gonads are underdeveloped, this is usually thought to be because one of the genes in the group required to direct the development is missing or is not working. However, in most cases we don't know which particular gene was not working. When the gonads don't develop fully this is called gonadal dysgenesis. So that is the name of the condition you have.

c. Ranya: Do I have a uterus and a vagina?

Doctor: Yes, you do. But because you are not making any estrogen, your uterus is quite small, and has not grown as usually happens during puberty. However, with estrogen treatment your uterus will eventually grow and you will start to have periods like your friends. This will take a while, as we need to increase the estrogen dose slowly, so your body has a chance to get used to it and mature at a natural rate. Some girls with your condition have a vagina that might be a bit narrower or shorter than is typical. From our examination, your vagina developed like that of most girls. If you ever have any questions you'd like to ask us without your parents, we can ask them to leave. We also have some female colleagues who'd be happy to talk with you any time you come to the clinic

d. Ranya: do you know which gene was not working that caused my gonadal dysgenesis?

Doctor: no, we don't, but we do have a bit more information about your condition from the tests we did a few weeks ago. So first, back to 7th grade biology. Do you remember learning about the chromosomes, and how many chromosomes typical human beings have?

Ranya. Yes, we did learn this, but I thought it was very complicated, so can you tell me again?

Doctor: Of course. I think this picture might help (for this part I would show a picture of 2 fluorescent-stained karyotypes side-by-side – a typical 46,XX karyotype and a typical 46,XY karyotype). This shows a picture 2 sets of chromosomes taken down a microscope, stained with special chemicals used to make each pair show up in a different color. These are arranged by size from the biggest to the smallest, and you can see that there are 22 numbered pairs that are pretty much matching – one of each pair from our mother and one from our father (don't take any notice of the fact that a few of them look bent – this is just the shape that they were when the picture was taken). The last 2 at the end in this set are called "X" chromosomes; they were given a letter instead of a number because scientists originally were confused by the X chromosome, so "X" stood for something mysterious. In the other picture you can see that the last 2 chromosomes don't match each other in size. The one on the left is an "X" and the smaller one on the right is called a "Y" chromosome (just because it's the next letter after X). Although it is smaller, the Y chromosome carries most of the same important genes as the X chromosome, plus one gene that is usually involved in making the gonads develop as testes. For this reason some people refer to the Y as the "male" chromosome. However, this is not really correct, because a human embryo needs the actions of lots of different genes on many different chromosomes for the gonads to develop as either ovaries or testes. Now coming back to why gonadal dysgenesis develops, we know that in many cases it is because one of the X chromosomes is missing, so the girl has only one X (show this on karyotype picture by covering up the 2nd X or Y). In a smaller number of cases we find that the girl has an X and a Y chromosome and in other girls with this condition we find 2 X chromosomes. In your case we found that you have an X and a Y chromosome. Although these are the chromosomes we most commonly find in boys and men, there are quite a large number of girls and women who have the same X and Y chromosomes, including quite a few who come to see me.

Ranya: Does this mean that I'm really a boy?

Doctor: No Ranya, you are a girl, but you just took a slightly different pathway to becoming a girl. The way I sometimes describe this is to imagine you are going on a journey. You start out in [City A]and you are heading to [City E] (use appropriate geographic references). If you take the straightest route, you would head North up highway 65 and go through [City B]. If you took a slightly different route, you might go first through [City D] and [City D] and then come back to the highway to complete the journey to [City E]. We can think of your development as being a bit like this – taking the slightly different route, but coming to about the same point at the end of the journey. Does that make sense?

e. Doctor: Now that you understand about chromosomes and which chromosomes you have, there is another topic we need to talk about, and that is your gonads. Remember earlier in our discussion I mentioned that your gonads are underdeveloped and this is called gonadal dysgenesis. In cases of gonadal dysgenesis where a girl has a Y chromosome as you do, there is an increased risk of some of the cells in the gonad growing too much and developing into a tumor. Based on your MRI scan and your blood tests we see no evidence of a tumor. However, as your gonads do not produce eggs or hormones, and do have an increased risk for developing a tumor, we would generally recommend that your gonads should be removed to prevent you from developing a tumor as you get older. This would be done by a surgeon using a flexible scope inserted through a small incision in the groin area. There is no rush to get this done, so we can talk about this some more at your next visit, and we will also talk then about your options and details regarding hormone treatment.

3. Doctor: I have probably given you as much information as you can take today, but I want to be sure that I haven't confused you with too much information. Can you tell me what pieces of information you think are most important for you right now? Is there anything that confused you that you would like to go over again before you leave today? You will probably think of a number of questions as soon as you get into the car to go home, so just write them down and bring them when you come back to see me next time and we can talk about them again.

7

CHAPTER 7

Global Application of the Assessment of Communication Skills of Paediatric Endocrinology Fellows in the Management of Differences in Sex Development Using the ESPE E-Learning.Org Portal

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Background

Information sharing in chronic conditions such as disorders of/differences in sex development (DSD) is essential for a comprehensive understanding by parents and patients. We report on a qualitative analysis of communication skills of fellows undergoing training in paediatric endocrinology. Guidelines are created for the assessment of communication between health professionals and individuals with DSD and their parents. Methods: Paediatric endocrinology fellows worldwide were invited to study two interactive online cases (www.espe-elearning.org) and to describe a best practice communication with (i) the parents of a newborn with congenital adrenal hyperplasia and (ii) a young woman with 46,XY gonadal dysgenesis. The replies were analysed regarding completeness, quality, and evidence of empathy. Guidelines for structured assessment of responses were developed by 22 senior paediatric endocrinologists worldwide who assessed 10 selected replies. Consensus of assessors was established and the evaluation guidelines were created. Results: The replies of the fellows showed considerable variation in completeness, quality of wording, and evidence of empathy. Many relevant aspects of competent clinical communication were not mentioned; 15% (case 1) and 17% (case 2) of the replies were considered poor/insufficient. There was also marked variation between 17 senior experts in the application of the guidelines to assess communication skills. The guidelines were then adjusted to a 3-level assessment with empathy as a separate key item to better reflect the qualitative differences in the replies and for simplicity of use by evaluators. Conclusions: E-learning can play an important role in assessing communication skills. A practical tool is provided to assess how information is shared with patients with DSD and their families and should be refined by all stakeholders, notably interdisciplinary health professionals and patient representatives.

Introduction

Delivery of optimal clinical care should be patient- and family-centred. A comprehensive understanding by parents and patients of a chronic condition such as disorders of/differences in sex development (DSD) is a necessary prerequisite for ongoing communication and care [1--4]. The objectives of information sharing are multiple: first, it allows the parents and patient to understand the condition with all its short- and long-term implications for management; second, it supports the psychological adaptation required to live well with the condition; and finally, it enables communication within the patient's social environment [4]. All these factors will impact positively on the quality of decisions made by patients and families, and adherence to the demands of ongoing management.

Information sharing in DSD conditions is particularly challenging since not only is the management of individuals with DSD continually evolving, it is also taking place within the context of changing perceptions of sex and gender, with questioning or rejection of the binary nature of both constructs in parallel with corresponding changes in societal regulations [5, 6]. Communicating DSD information requires navigating difficult subjects, such as the stigma anticipated by the parents or patient, sensitive matters including potential future sexual activity or fertility, and perhaps most challenging, awareness of one's own tendencies to perpetuate cultural norms that might oppress or limit an individual with a DSD and negatively affect their developing identity.

The rather complicated information on genital development and puberty should be provided to patients in clear and understandable terms. If the information is explained incompletely or deferred, there is a risk of inaccurate self-diagnosis without the opportunity for discussion with expert care providers [7]. Moreover, it has recently been suggested that the confusion parents of children with a DSD feel regarding the information on their child's condition may lead to the development of posttraumatic stress syndrome [8].

Paediatricians pursuing subspecialty training in paediatric endocrinology (fellows) are taught that the conversations with parents and the patient should be informative, but non-intimidating and sensitive to discomfort and anxiety. These conversations should also be respectful of cultural and psychosexual issues [5].

In a recent paper, we reported results of an exploratory study regarding the suitability of the European Society of Paediatric Endocrinology (ESPE) e-learning portal to assess, at a global level, junior paediatricians (fellows) during their subspecialty paediatric endocrinology training regarding knowledge and skills for investigation and management of children and adolescents with DSD [9]. An additional objective of this study was to evaluate the communication skills in informing patients and parents about the diagnosis and management. So far, there have been various examples of the suitability of e-learning in medical education. The review by Sinclair et al. [10] suggests that e-learning is at least as effective as traditional learning in improving health care professional behaviour. There are some examples of e-learning modules especially dedicated to the improvement of patient-physician communication skills [11--13]. The intent of this study was to explore whether the communication skills could be assessed and taught in a formative way in an online environment – in particular, to learn from online feedback. So far, no other such study has been described.

Here we report on the qualitative analysis of these communication skills, including addressing of key elements in the information-sharing process, with particular emphasis on wording and empathy. In presenting these materials, we also create guidelines for the assessment and teaching of communication between health professionals and parents of newborns and individuals who have a DSD.

Material and Methods

The outline of the study has been described previously [9]. In brief, paediatric endocrinologists in training worldwide (referred to hereafter as fellows) were invited to login to a specially created DSD e-learning course within the ESPE e-learning portal (www.espe-elearning.org). Access was provided to two interactive DSD cases prepared as e-learning exercises in English. The two clinical cases were within the expertise of paediatric endocrinologists; the fellow was asked to play the role of a paediatric endocrinologist in an interdisciplinary DSD team.

Case 1 focused on the stepwise approach to the investigation and management of a newborn referred by a midwife because of atypical genital development secondary to congenital adrenal hyperplasia (CAH). Case 2 addressed the approach to a 17-year-old girl with delayed menarche due to 46,XY gonadal dysgenesis. At crucial stages of the diagnostic process, 3 open-ended questions (i.e., requiring a text response) were included, in which the fellow was invited to describe how (s)he would handle the situation, focusing on how (s)he would inform the parents and/or patient of key information regarding diagnosis, implications and management. The fellows had received no formal training in communication skills over the course of their fellowships.

The replies to the open-ended questions of each fellow were submitted to 2--3 senior paediatric endocrinologists (referred to hereafter as experts) from various parts of the world. At least 1 of the experts worked in the same geographic region as the fellow (excluding the expert at the institution where the fellow was employed). The experts were invited to reply with feedback to the fellows' replies. The experts were provided with an exemplary model answer prepared by the study team, which they were free to use. For each question, key items were identified as critical elements of an adequate reply by the study team. Subsequently, the fellows were asked to acknowledge and reply to the comments of the experts. All procedures were handled anonymously through the e-learning portal.

The present study focused on open question 2 of case 1 and open question 1 of case 2, reflecting communication skills in the field of DSD. A description of the cases and selected questions is provided in Table 1.

Table 1. The cases and the open questions as provided to the fellows. This information was also provided to the assessors in the online questionnaire in order to mark the selected answers of the fellows on these questions.

Case 1 – CAH
Mrs. Johnson is at home and has just given birth to her second child. Her pregnancy was uncomplicated, and her term delivery was uneventful. The infant has a birth weight of 2,940 g, and had an Apgar score of 9/10. The midwife is very concerned about the infant's genital development, as it is unclear to her whether the child is a boy or a girl. The decision is made to transfer the baby to a specialized centre. You are now the paediatric endocrine fellow in charge. Physical examination of the infant at 4 h of age reveals a healthy looking, well-developed, alert, non-dysmorphic newborn. The infant is not in any distress. The baby's genitalia have the following features: the hyper-pigmented labio-scrotal folds are fused posteriorly; the gonads are not palpable; the phallic structure is tethered by a fibrous string (chordee); there is one opening on the perineum through which urine is passed.
Open question provided to the fellows Based on laboratory and imaging studies you have just made the diagnosis of congenital adrenal hyperplasia (CAH), most likely as a result of 21-hydroxylase deficiency. Give a detailed account of how you will inform the parents of the diagnosis. Consider the following elements: the setting; allotted time; explanation of the diagnosis; therapeutic issues; implications for later; follow-up; other considerations.

Table 1. Continued

Case 2 – Gonadal dysgenesis
Ranya, a 17-year-old girl adopted from Sri Lanka, is referred to you because of delayed puberty. She is quite concerned, as she hasn't had her period yet, whereas all her girlfriends are menstruating. She saw a doctor 2 years ago because of delayed breast development, and the doctor told her just to be patient. Ranya was born at term with a birth weight of 2,400 g; her length was not measured at birth. She was adopted at the age of 6 weeks. No family history is available. At the age of 14 weeks, she was diagnosed with viral meningitis without obvious sequelae. Since then, she has been healthy and takes no chronic medications. Her developmental milestones have been normal. She noticed some pubic hair growth at 12 years of age, but no breast development, and menarche never occurred.
Open question provided to the fellows Based on laboratory and imaging studies you have made the diagnosis 46,XY gonadal dysgenesis. Give a detailed account of how you will arrange the sharing of information to Ranya and her parents. Consider the following elements: the setting; allotted time; key items to be discussed regarding the diagnosis; relevant therapeutic issues; long-term implications; follow-up; other considerations. In addition, describe how you will address Ranya and her parents when informing them about the diagnosis and therapeutic options.

There appeared to be a wide variation in the replies of the fellows, not only in the number of key items mentioned, but also in the quality of the wording and in the use of empathy. Therefore, to evaluate all the replies, there was a strong need for a structured approach and a rubric was developed. Rubrics are documents that articulate the expectations for an assignment by listing the assessment criteria and defining levels of quality in relation to each of these criteria. The steps below describe this process, which is also presented in a flow chart shown in Figure 1.

Step 1 – Development of Guidelines for Assessment

First (step 1a), the replies of the fellows were dissected into sentences relating to the identified key items. All passages were analysed by one person (S.L.S. Drop), who rated the passages on a scale of 5 levels (poor-insufficient-reasonable-good-excellent). According to this analysis, descriptors were defined. Descriptors spell out for each key item at each level of performance what performance at that particular level looks like. This resulted in a rubric, providing objective guidelines for the assessment of the fellows' replies. In the next step (step 1b), these guidelines were provided to the study team for discussion. The study team consisted of paediatric endocrinologists, psychologists, educationalists and patient/parent representatives (S.L.S. Drop, M. Cools, J. Alderson, L.J.C. Kranenburg, K. Grijpink, M. Muscarella, E. Magrite). After extensive adjustments, consensus was reached (online suppl. Table 1; for all online suppl. material, see www.karger.com/doi/10.1159/000475992).

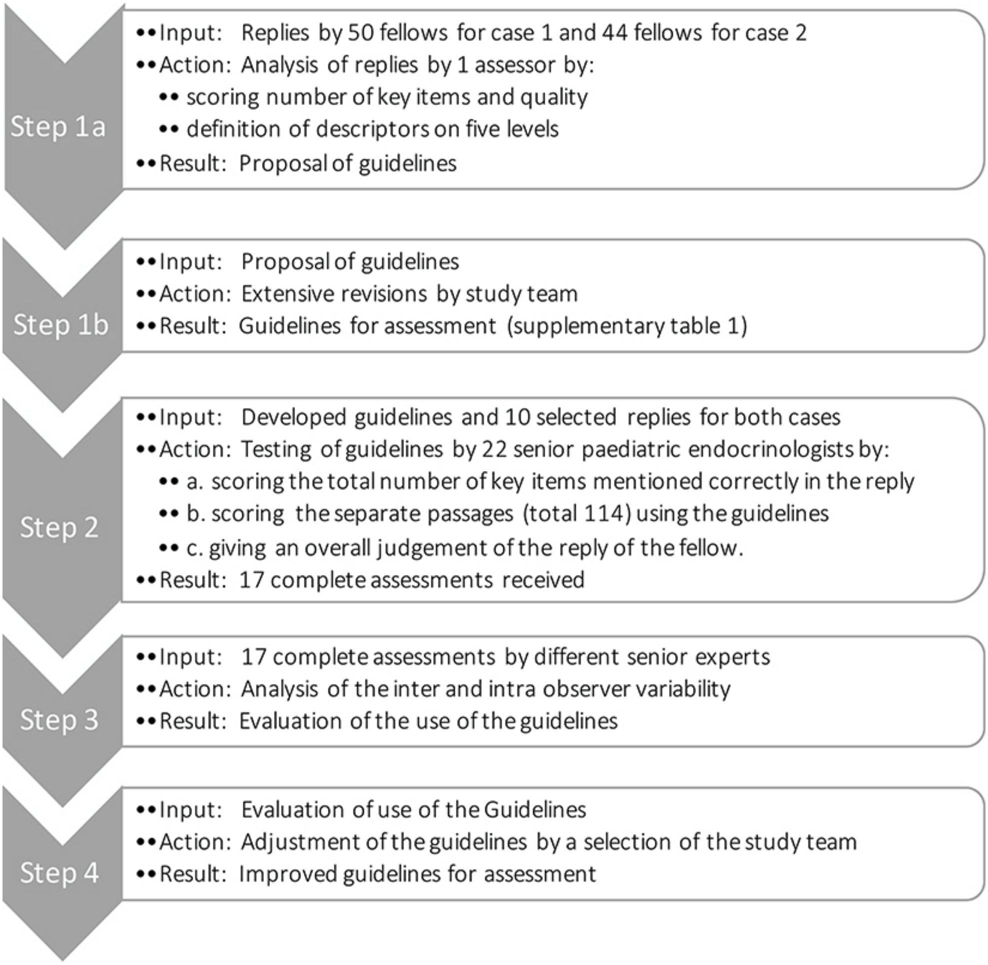

Figure 1. Flow chart describing the steps in development, evaluation and adjustment of the guidelines for assessment of communication skills in DSD. The steps are described in detail in the methods section.

Step 2 – Testing of the Guidelines for Assessment

To validate the guidelines, a selection of the answers from 10 fellows to the open questions of each case was made, based on the first evaluation by one of the investigators. The selection represented a representative sample of all received replies. The selected replies for cases 1 and 2 were not necessarily from the same fellow. The sentences in the replies were cut into passages and grouped according to the identified key items. Twenty-two senior paediatric endocrinologists worldwide (referred to hereafter as assessors) were invited to participate. They were provided access to an online questionnaire with an overview of the two cases and the two open questions (Table 1), the selected replies (provided as distinct passages), and a link to the study design and the guidelines (online suppl. Table 1). Assessors were asked to score each of the passages based on the provided guidelines on a 5-point scale from poor to excellent. To assess the complete reply, assessors were also asked to score the total number of key items described correctly in the answer, and to score the overall reply on a 5-point scale from poor to excellent, taking into account completeness, quality of the answer, and the use of empathy.

Step 3 – Evaluation of the Guidelines for Assessment

As this study aimed to create globally applicable guidelines, one assessor with minimal intra-observer variability compared with the other assessors was identified as an outlier and excluded from further evaluation. Assessors who did not complete all replies were also excluded. The scoring of the remaining assessors was evaluated for each key item separately, as well as combined together as an overall assessment, using an intraclass correlation coefficient (ICC). For further details, see the statistics section. This resulted in an evaluation of the interobserver variance for the assessment of the separate key items and the overall assessment. This analysis was first performed using a 5-level rating (poor-insufficient-reasonable-good-excellent) and subsequently using a 3-level rating (poor-reasonable-good). For the 3-level rating, the two lowest levels (poor and insufficient) and the two highest levels (good and excellent) were combined.

To measure the agreement between the assessors of the competency score of the separate text passages, we calculated the standard deviation for the score of the assessment (1 poor – 5 excellent) of (i) the separate text passages and (ii) the overall reply. The standard deviation was used to identify text passages with a high level of agreement/low standard deviation and passages with a low level of agreement/high standard deviation. These text passages were studied in detail, taking into account the geographic background of the assessors and the provided comments.

Step 4 – Adjustment of the Guidelines for Assessment

The above analyses were used to adjust, where necessary, the guidelines for communication to parents of newborns with CAH and for girls and young women with gonadal dysgenesis. The number of key items was evaluated and optimized, as were the number of levels describing the performance of the key items, in order to create the final guidelines.

Statistical Analysis

To determine the level of agreement between assessors represented by the interrater reliability (IRR), an ICC(2,k) statistical analysis was performed ("icc" command from "irr" package for R, settings: model = "twoway", type = "agreement", unit = "single"). All analyses were performed using R (version 3.2.2, R foundation for Statistical Computing, Vienna, Austria). A p value ≤0.05 was used as a cut-off for statistical significance.

Results

Step 1 – Development of Guidelines for Assessment

Fifty fellows responded to the open question for case 1 and 44 fellows to the open question for case 2. The global distribution of the participating fellows has been previously described [9]. As described in the methods section, these replies were analyzed by scoring the number and quality of the predefined key items on a 5-level rating scale. The results for both cases are presented in Figure 2.

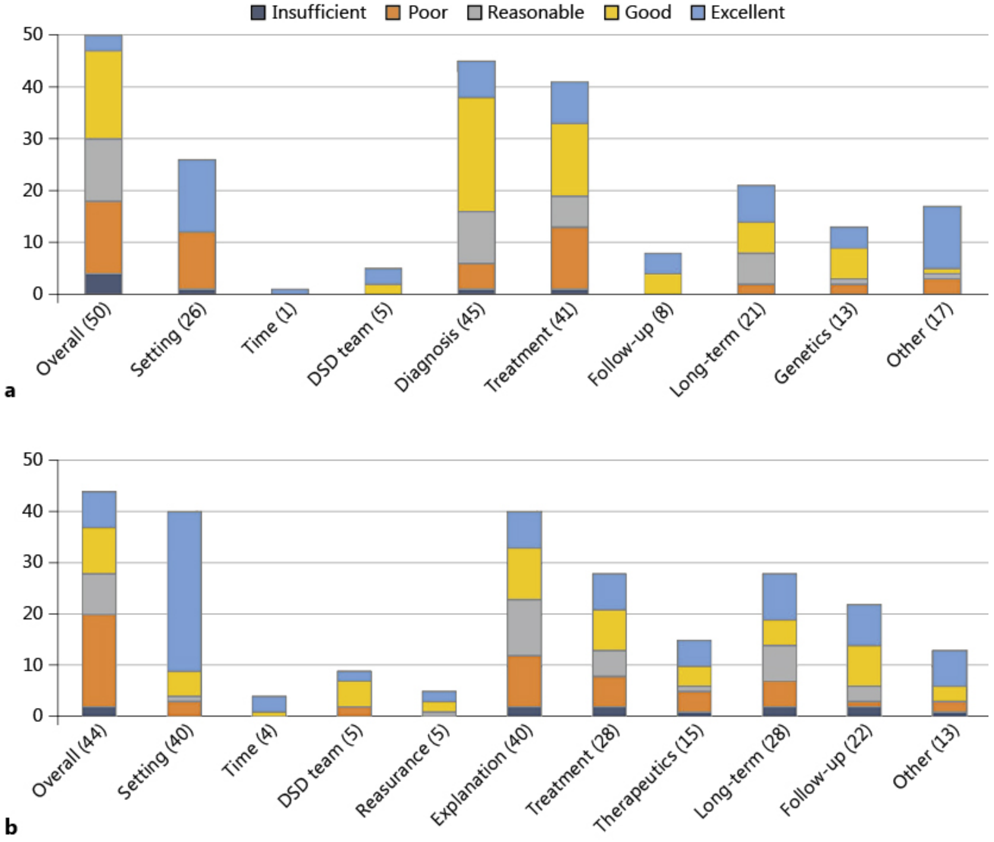

Figure 2.The vertical axis represents the number of replies by the fellows and the frequency with which the different key items were identified in the reply for case 1 (top) and case 2 (bottom).

The horizontal axis shows the overall judgement and the identified key items (number of assessments in brackets). Colours represent the marking on a 5-point scale. This analysis shows that items "diagnosis" and "treatment" are the items most frequently mentioned (45/50 and 41/50 fellows for case 1 and respectively by 40/44 and 28/44 fellows for case 2). "Setting" is mentioned by 26/50 fellows for case 1 and 40/44 times for case 2. The quality of the text fragments specifying "setting" ranged from "poor," when no information on the setting was provided, to "excellent," when the importance of a private ambiance was recognized. Remarkably, "allotted time," explanation of "the role of the DSD team" and the relevance of providing long-term "follow-up" was rarely mentioned. Items mentioned in the category "other" were, for example, handing out written explanations, figures and drawings, and time for questions.

We found that there was substantial variation not only in the number of key items mentioned, but also in the quality of the responses. Examples of text phrases that qualified for the different rating categories are presented in online supplementary Table 2. From this analysis, descriptors were defined, which described each key item at all levels of performance. This allowed the creation of guidelines to score (into 5 categories) the fellow's explanations of the 2 conditions for accurateness, clarity, and empathy. These guidelines were then passed to the study team for refinement and consensus. The resulting product was the guidelines used in this study for assessing communication with parents of newborns with CAH and with girls with complete gonadal dysgenesis (online suppl. Table 1).

Step 2 – Testing of the Guidelines for Assessment

We randomly selected the replies of 10 fellows to each of the 2 open questions. These answers were divided into passages according to the defined key items and sent to 22 experts to score, as outlined in the methods. The demographic background of the experts was as follows: Africa (1); Asia (4); Australia (1); Europe (8); North America (5); and South America (2). The interrater variability, the standard deviation and the distribution of the assessment categories are represented in online supplementary Table 3. We received 21 replies from the experts. Two incomplete replies and 1 which was regarded as an outlier (due to low variation and only high scores in contrast to other experts) were excluded from further evaluation, resulting in 17 complete expert evaluations of the fellows' replies.

Step 3 – Evaluation of the Guidelines for Assessment

The assessments of the fellows' responses by the experts showed considerable interrater variation. As shown in Table 2 using the 5-level rating by the provided guidelines, the ICC for all 134 assessments was 0.37 (95% confidence interval 0.31–0.45), indicating a low level of agreement between the assessors. The highest agreement was reached on the assessment of passages related to "setting," with an ICC of 0.58. The ICC on the overall assessment of the complete answer was higher than the assessment of the separate text passages referring to specific key items. However, statistical significance was never reached. Some categories such as "the role of the DSD team" or "checking of understanding" were mentioned too infrequently to calculate a reliable ICC. When adjusting for a 3-level rating scale by combining ratings “poor” and “insufficient” in the lowest rating and combining “good” and “excellent” as the highest rating, the ICC for “setting” was significantly higher than for all assessments together.

Table 2. Intraclass coefficient (ICC) of the ratings provided by the assessors (excluding outliers and incomplete data

	5-level rating		3-level rating	
	ICC	95% CI	ICC	95% CI
All assessments (134)	0.37	0.31 < ICC <0.45	0.41	0.34 < ICC <0.48
Overall (20)	0.42	0.28 < ICC <0.62	0.47	0.32 < ICC <0.67
Setting (17)	0.58	0.42 < ICC <0.78	0.68*	0.53 < ICC <0.84
Follow up (5)	0.34	0.13 < ICC <0.82	0.38	0.15 < ICC <0.84
Explanation (44)	0.32	0.22 < ICC <0.44	0.35	0.25 < ICC <0.48
Treatment (20)	0.31	0.19 < ICC <0.51	0.31	0.19 < ICC <0.51
Long-term implications (11)	0.28	0.13 < ICC <0.56	0.27	0.13 < ICC <0.56
Genetics (5)	0.23	0.06 < ICC <0.74	0.20	0.05 < ICC <0.71
DSD team (2)	0.12	–0.02 < ICC <0.99	0.11	–0.03 < ICC <0.99
Check understanding (6)	0.09	0.02 < ICC <0.43	0.06	0.008 < ICC <0.35
Other considerations (2)	0.06	–0.01 < ICC <0.99	0.08	–0.006 < ICC <0.99

Parentheses in the first column represent number of responses. The first row represents ICC for all assessments together (ICC 5-level rating 0.37); the second row represents ICC for overall judgement of the replies (ICC 0.42); and the rows following indicate ICC for the ratings of the evaluated text passages referring to the indicated key items. The columns provide ICC values and 95% confidence intervals (CIs) for the used 5-level rating scale and for a 3-level rating scale when the lowest and highest two scores are taken together. When adjusting for a 3-level rating scale by combining ratings “poor” and “insufficient” as the lowest rating and combining “good” and “excellent” as the highest rating, the ICC for “setting” is significantly higher than for all assessments together. *

There was remarkable assessor disagreement in rating the fellows’ replies for both cases, as illustrated in Figure 3, which shows the assessments for the complete replies of each fellow.

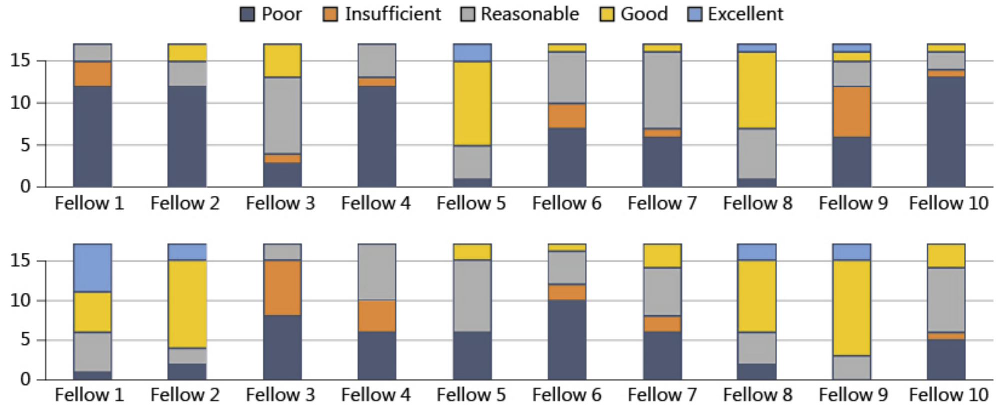

Figure 3. Assessment of the complete answer of the selected fellows by 17 assessors for case 1 (top) and case 2 (bottom).
Horizontal axis represents the fellows, vertical axis the number of assessors. Colours indicate the rating. Outliers and incomplete data were excluded. For example, this shows that there was considerable variation in the rating of reply of fellow 9 for case 1 which is rated as: poor, 6 times; insufficient, 6 times; reasonable, 3 times; good, once; and excellent, once.

The numerical rating of correct key items mentioned in the communication/explanation also showed a wide range, as illustrated in Figure 4. Phrases with a high level of rating agreement or a low level of rating agreement were identified, as described in the methods section. Examples of these passages are provided in online supplementary Table 4.

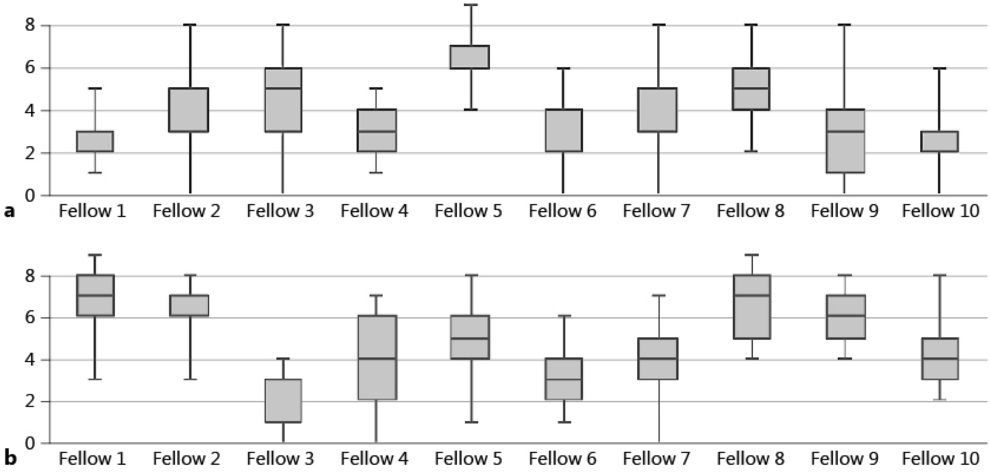

Figure 4. Number of key items correctly mentioned in the replies of the fellows for case 1 (top) and case 2 (bottom) as scored by 17 assessors, represented in box-and-whisker plots.
Horizontal axis represents the separate fellows; vertical axis represents the number of key items. Whiskers represent minimum and maximum values; boxes represent first and third quartiles and the median; outliers and incomplete data were excluded. For example, for fellow 9 of case 1, the minimal number of items mentioned was none, the maximum number of items was 8 and the median was 3.

Table 3. Guidelines for the assessment of communication skills

Key item	Good	Reasonable	Insufficient
When informing parents of a newborn with CAH			
Setting	Private room; no interruption by colleagues, personnel etc.; no beepers, mobile phones	Private room; interruption policy not arranged	No privacy provided
Time	Approx. 30–45 min; ensure that parents are aware of the time allotment. If necessary, follow-up in the following days/week	Approx. 30–45 min	Limited time (<15–20 min)
Reassurance	Emphasizing: "Your baby will be fine." Normal life expectancy; fertility possible	Mentioning: normal life expectancy; fertility possible	Not mentioned
One spokesman on behalf of DSD team	One spokesman on behalf of the DSD team or jointly with medical team psychologist (if available)	One spokesman on behalf of the DSD team	DSD team not mentioned
Explanation of diagnosis	1. Adrenal disorder termed congenital adrenal hyperplasia, abbreviated as "CAH" with prenatally increased levels of "male-type" hormones, androgens 2. Uterus and ovaries present 3. Shows explanatory pictures or drawings	Relevant items mentioned but limited explanation	Relevant items not or incompletely mentioned
Treatment	1. Corticosteroid treatment (life-long) will stop virilization 2. Salt loss requires fludrocortisone treatment (life-long) 3. Instruction for administration of stress medication	Life-long and stress medication mentioned but with limited explanation	Life-long medication not or incompletely mentioned stress medication not mentioned
Follow-up	Provides information regarding regular follow-up for adjustment of dosage of medication and reminder of instructions for administration of stress medication is essential; discusses importance of adherence to prescribed treatment and consequences of non-adherence	Some but not all items mentioned but limited explanation	Not mentioned
Long-term implications	The clitoris is expected to become less noticeable under medical treatment; joint appointment with surgeon (or urologist or gynaecologist) to check on this in about 3–4 months to discuss whether or not genital surgery is an option	Some but not all items mentioned but limited explanation	Not mentioned
Genetic counselling	Explanation of the genetics of CAH and risk for future pregnancies, also options for pre-implantation genetic diagnosis *Note – in some institutions this will be done by a genetics consultant or counselor rather than endocrinologist. Need to raise the issues that must be discussed, and list options for who will undertake this discussion*	Genetics mentioned but not explained	Genetics not mentioned

Table 3. Continued

Empathy	*Questions:* time for parents' questions and concerns *Checks understanding:* checks whether the main messages are understood, e.g. the diagnosis and therapeutic consequences, adjustment of medication dosages, and stress medication *Written information:* written summary of discussion *Arranges meeting with parent/patient:* offers to arrange in clinic a meeting with another family who are caring successfully for a child with CAH *Reading material:* reading materials published by support groups, contact information for local and/or online support groups	Items mentioned in part	Items not mentioned
When informing a patient with gonadal dysgenesis and her parents			
Setting	Make an appointment with patient and both parents; private room; no interruption by colleagues, personnel etc.; no beepers or mobile phones	Private room; interruption policy not arranged	No privacy provided
Time	45–60 min; ensure that patient/family is aware of the time allotment. If necessary, follow-up in the following days/week	Approx. 30–45 min	Limited time (<15–20 min)
Reassurance	Emphasizing: "First and most importantly, I want to reassure you that you are quite healthy and there is no serious problem to be concerned about"	Mentioning: no serious health problem	Not mentioned
One spokesman on behalf of DSD team	Possibly jointly with medical team psychologist or social worker if one is available with experience with DSD	One spokesman on behalf of the DSD team	DSD team not mentioned
Explanation	Provides extensive explanation regarding the following key elements: 1. Typical development of ovaries/gonads and atypical development in gonadal dysgenesis 2. Genes and chromosomes 3. Hormones and hormone replacement 4. Female reproductive anatomy 5. Fertility/infertility 6. Potential tumour risk	Most items mentioned but limited explanation	Some relevant items not or incompletely mentioned; explanation from viewpoint chromosomal error: patient "being a girl by default"
Therapeutic issues	Hormone replacement during the teenage years (oestrogen and later progesterone); these hormones can be provided in many ways (pills, patches, sprays, creams, etc.) and have to be continued into adulthood	Some but not all items mentioned but no or limited explanation	Relevant items not or incompletely mentioned

Long-term implications	Sexual relationships; infertility; options to have a partner, a family Advice to share the condition only with close relatives or friends Offering psychological expertise	Some but not all items mentioned but no or limited explanation	Questioning female gender identity; relevant items not or incompletely mentioned
Follow-up	Outpatient clinic appointment for initiation of oestrogen treatment; subsequent joint appointment with urologist/surgeon or gynaecologist to discuss gonadectomy	Some but not all items mentioned but no or limited explanation	Not mentioned
Empathy	*Questions:* provides time for patient and parents' questions and concerns *Checks understanding:* checks whether patient and parents understand the main messages *Written information:* provides a written summary of the discussion; provides any reading materials published by support groups *Support group:* provides contact information for local or online support groups		

Consensus was reached when answers were very short or blunt, and rated as poor or insufficient. Rating was good or excellent when an answer was complete, mentioning relevant details and expressing empathy. The variation in the ratings by expert assessors was as extensive as the variation in answers provided by respondent fellows. However, no relationship between assessment rating pattern and demographic background of the assessors could be established.

Step 4 – Adjustment of the Guidelines for Assessment

Based on these findings, the guidelines were adjusted to a 3-level assessment, with empathy as a separate key item. The revised guidelines better elucidated the qualitative differences in the replies. The final guidelines are presented in Table 3.

Discussion

In this exploratory study, we evaluated the applicability and feasibility at a global level of the ESPE e-learning portal in assessing junior paediatricians (fellows) during their subspecialty paediatric endocrinology training. We examined the quality of communication skills for information sharing with patients with DSD and/or their parents. So far, there has been little experience in the evaluation of formative assessment within the widespread utilization of online learning in the education of medical specialists [14]. However, it has been shown that e-learning is at least as effective as traditional learning approaches, and superior to no instruction at all in improving health care professional behaviour [10].

Aim: Assessment for Learning

We showed that e-learning which incorporates feedback can play an important role in assessing and learning communication skills for information sharing of DSD. We aimed to evaluate the assessment during the learning process where direct feedback to the learner is provided by a tutor in the context of e-learning. This is termed formative assessment, or assessment for learning [15]. It should be noted that fellows are in training to develop communication skills, and feedback from assessors can be used to enrich communication skills. In addition, we explored whether we could formulate generally applicable qualitative guidelines for formative assessment using a rubric method [16]. It is important to note that these guidelines were developed to stimulate discussion between fellow and expert and thereby improve learning. The questions were certainly not intended to be used for fail/pass decisions at the end of the learning process (summative assessment, or assessment of learning) [15].

Need for Structured Assessment and Applicability

In our study, fellows worldwide showed a broad range of communication skills. In the fellows' replies to the open question to accurately and empathically inform the parents of the newborn and the 17-year-old girl about each condition and its long-term consequences, we noted that many relevant items were not mentioned. These included reassurance, follow-up, long-term implications, and the role of the DSD team. In 15% (case 1) and 17% (case 2), the overall assessment of the replies was considered poor/insufficient by the experts. We were disappointed that very few fellows (5 out of 44) provided reassurance about general health and quality of life to patients and their families in these conversations [17].

Specific DSD conditions require certain considerations. When communicating with parents of a newborn baby with CAH, it should be realized that they are overwhelmed facing the prospect of caring for their infant. Their ability to absorb information is limited [17, 18]. We provided guidelines that are in line with recent suggestions from parents on how to improve the initial information provided after their child's diagnosis [17].

In addition, direct communication with an adolescent means that the doctor must be aware of developmental issues that may be present. In communicating with an adolescent with DSD, it is important to realize that there are many concerns. These include the possibility of emotional distress and feelings of isolation at the time of diagnosis, the prospect of infertility, and the difficulty of sharing this information with their peers and romantic partners [19]. We noticed that these concerns were not addressed adequately in many replies from the fellows.

We have no detailed information on the previous exposure of the fellows to patients with DSD. We recognize that even in a large teaching centre, the number of patients with DSD is small. Furthermore, these fellows had not received formal teaching in their fellowships on communication in DSD. Our study identified a need in this area. Thus, providing e-learning material consisting of structured guidelines and providing a stepwise approach to virtual patients may be a welcome adjunct in fellowship training programs. Virtual patients can readily provide learners with multiple and varied case examples, while offering the opportunity to receive feedback, not only on the accuracy of the learner's diagnosis and treatment plan, and the information-gathering and decision-making path, but also on the quality of information sharing and communication with patients and families [20].

As a limitation of the study, we acknowledge that English was not the native language of many participating fellows, which may have influenced their understanding of the questions and formulation of their replies. It was not possible to make an analysis of the impact of language on the assessment, as no information on their fluency of English was available. Regional or national application of the e-learning portal may best be used with direct interaction between fellow (student) and expert (tutor).

Use of Guidelines by Assessors

We noted considerable diversity in the application of the developed guidelines by the participating senior endocrinologists. Differences between assessors is a well-known phenomenon: first, panels of experts may show differences in their personal performances [21]; second, differences between experts frequently exist in scoring and weighing assessments [22]; and third, differences in scoring may also be due to cultural and geographical differences. However, in our study, no correlation between geographic origin and rating could be established. Obviously, non-verbal communication that may have nuanced the interpretation of the text passages was lacking. The voluntary time commitment asked of the assessors may have also contributed to the varied results. Experts were provided with model answers to the open questions to help reduce variability in their responses. However, they were not formally trained in the use of the guidelines prior to the study, which may be helpful to do in future studies to see to what extent consensus can be reached.

One conclusion is that the participating assessors in this study may differ in their opinions on what the best words to use are during this nature of consultation. Another possibility is that assessors themselves may differ in their communication skills. The selected assessors were not chosen as those specialized in communication skills, but were senior paediatric endocrinologists selected from amongst the most experienced in contemporary DSD practice in specialist centres. In practice, the paediatric endocrinologist often fulfils the role of spokesman in a DSD team, sometimes in the presence of a psychologist. Moreover, the endocrinologist is also responsible for offering face-to-face training of fellows in hospital practice. We would like to express our hope that senior paediatricians might become aware of these differences and work together to develop a (gold) standard for optimal communication in DSD. For future use of this learning method, we would like to make a plea to train the participating assessors, for example, by virtual group discussions on to how to best respond to the scenarios given. This would both enhance the skills of the "experts" and may improve the interrater scoring. Furthermore, this learning method may itself prove helpful for educating established physicians, and not just trainees, in the future.

Creation and Ongoing Evolution of Guidelines for Assessment

We created guidelines with input from international fellows and experts in paediatric endocrinology that can be applied for e-learning and assessment, and complement clinical experience, to improve communication about DSD. In order to prepare guidelines to assess communication skills of the fellows by the experts in the study, a rubric was developed as a structured approach for the development of scoring guidelines [23] (online suppl. Table 1). As the evaluation of the 5-level and 3-level assessment tool gave comparable results, we suggest that it is more practical to use the simplified 3-level rating scale: insufficient-reasonable-good (Table 3).

As we reported previously [9], it is of interest to note that the fellows in their first reply to the open questions did not mention cultural, religious or regional aspects. However, some acknowledged in their second reply to the expert that religious considerations are important, providing new insights for future use. Another important aspect not covered in these guidelines is the communication about strategies for sharing information (education and disclosure) with extended family and close friends. Finally, while the development of these guidelines has focused on the paediatric endocrinologist on the DSD team (an important starting point given the central role of the endocrinologist), we recognize that specialists from other disciplines on the team are (at least) as in need of assessment and learning of communication skills in DSD. It will be important to continue further refinement of these guidelines in close collaboration with all stakeholders, notably interdisciplinary health professionals and patient representatives, as evidence for best practice in DSD care continues to evolve.

Conclusion

It is well established that interactivity, practice exercises, repetition, and feedback improve learning outcomes, and also that interactivity and online discussion improve satisfaction in relation to online learning (or e-learning) for health professionals [23].

In general, online learning has traditionally been used to foster skills in the knowledge domain (e.g., clinical reasoning) rather than in practical skills such as communicating with service users where face-to-face contact is required. However, online learning can be very useful when preparing for face-to-face experiences. Ideally, educators will be best served by blending online learning and face-to-face instruction in individual or small group discussions [24--26]. In this context, e-learning can play an important role, not only for assessing medical knowledge, but also for assessing and learning communication skills [27] – of trainees, as well as physician educators. This method allows for practice in management and communication in DSD. While case vignettes and management advice in text books will quickly date, this method allows for evolving up-to-date practical advice and meaningful feedback from active practitioners. It is interesting to note that whereas communication training is routine for specialties like oncology, there is no requirement for practitioners and researchers in DSD to receive such support [17]. Based on recent guidelines [4--6], we provide a practical tool to assess how information is shared with patients with DSD and their families. This model may also be applicable to other chronic medical conditions.

References

1 Siminoff LA, Sandberg DE: Promoting shared decision making in disorders of sex development (DSD): decision aids and support tools. Horm Metab Res 2015;47:335--339.

2 Karkazis K, Tamar-Mattis A, Kon AA: Genital surgery for disorders of sex development: implementing a shared decision-making approach. J Pediatr Endocrinol Metab 2010;23:789--805.

3 Kogan BA, Gardner M, Alpern AN, Cohen LM, Grimley MB, Quittner AL, Sandberg DE: Challenges of disorders of sex development: diverse perceptions across stakeholders. Horm Res Paediatr 2012;78:40--46.

4 Nordenström A, Thyen U: Improving the communication of healthcare professionals with affected children and adolescents. Endocr Dev 2014;27:113--127.

5 Lee PA, Nordenström A, Houk CP, Ahmed SF, Auchus R, Baratz A, Baratz Dalke K, Liao LM, Lin-Su K, Looijenga LH 3rd, Mazur T, Meyer-Bahlburg HF, Mouriquand P, Quigley CA, Sandberg DE, Vilain E, Witchel S; Global DSD Update Consortium: Global disorders of sex development update since 2006: perceptions, approach and care. Horm Res Paediatr 2016;85:158--180.

6 Meyer-Bahlburg HF, Baratz Dalke K, Berenbaum SA, Cohen-Kettenis PT, Hines M, Schober JM: Gender assignment, reassignment and outcome in disorders of sex development: update of the 2005 consensus conference. Horm Res Paediatr 2016;85:112--118.

7 Liao LM, Green H, Creighton SM, Crouch NS, Conway GS: Service users' experiences of obtaining and giving information about disorders of sex development. BJOG 2010;117:193--199.

8 Pasterski V, Mastroyannopoulou K, Wright D, Zucker KJ, Hughes IA: Predictors of posttraumatic stress in parents of children diagnosed with a disorder of sex development. Arch Sex Behav 2014;43:369--375.

9 Kranenburg L, Reerds S, Cools M, Alderson J, Muscarella M, Grijpink K, Quigley C, Drop S: Global application of assessment of competencies of paediatric endocrinology fellows in the management of differences of sex development (DSD) using the ESPE e-learning.org portal. Med Sce Educ 2016;26:679–689.

10 Sinclair P, Kable A, Levett-Jones T, Booth D: The effectiveness of Internet-based e-learning on clinician behaviour and patient outcomes: a systematic review. Int J Nurs Stud 2016;57:70–81.

11 Wlodarczyk D, et al: Enhancing doctors' competencies in communication with and activation of older patients: the promoting active aging (PRACTA) computer-based intervention study. J Med Internet Res 2017;19:e45.

12 Daetwyler C, Cohen D, Gracely E, Novack D: eLearning to enhance physician patient communication: a pilot test of "doc.com" and "WebEncounter" in teaching bad news delivery. Med Teacher 2010;32:e381--e390.

13 Blödt S, et al: A consultation training program for physicians for communication about complementary medicine with breast cancer patients: a prospective, multi-center, cluster-randomized, mixed-method pilot study. BMC Cancer 2016;16:843.

14 Gikandi JW, Morrowa D, Davis NE: Online formative assessment in higher education: a review of the literature. Comput Educ 2011;57:2333--2351.

15 Schuwirth L, Ash J: Assessing tomorrow's learners: in competency-based education only a radically different holistic method of assessment will work. Six things we could forget. Med Teach 2013;35:555--559.

16 Reddy YM, Andrade H: A review of rubric use in higher education. Assess Eval High Educ 2010;35:435–448.

17 Boyse KL, Gardner M, Marvicsin DJ, Sandberg DE: "It was an overwhelming thing:" parents' needs after infant diagnosis with congenital adrenal hyperplasia. J Pediatr Nurs 2014;29:436--441.

18 Wisniewski AB, Sandberg DE: Parenting children with disorders of sex development (DSD): a developmental perspective beyond gender. Horm Metab Res 2015;47:375--379.

19 Ernst ME, Sandberg DE, Keegan C, Quint EH, Lossie AC, Yashar BM: The lived experience of MRKH: sharing health information with peers. J Pediatr Adolesc Gynecol 2016;29:154--158.

20 Brydges R, Manzone J, Shanks D, Hatala R, Hamstra SJ, Zendejas B, Cook DA: Self-regulated learning in simulation-based training: a systematic review and meta-analysis. Med Educ 2015;49:368--378.

21 Ericsson KAC N: Expert performance. Am Psychol 1994;49:725–747.

22 Swanson DB, Norcini JJ, Grosso LJ: Assessment of clinical competence: written and computer-based simulations. Assess Eval High Educ 1987;12:220–246.

23 Cascallar A, Cascallar E: Setting standards in the assessment of complex performances: the optimized extended response setting method; in Segers M, Dochy F, Cascallar E (eds): Optimizing New Methods of Assessment: in Search of Qualities and Standards. Dordrecht, Kluwer Academic, 2003.

24 Cook DA, Levinson AJ, Garside S, Dupras DM, Erwin PJ, Montori VM: Instructional design variations in internet-based learning for health professions education: a systematic review and meta-analysis. Acad Med 2010;85:909–922.

25 Cook DA, Erwin PJ, Triola MM: Computerized virtual patients in health professions education: a systematic review and meta-analysis. Acad Med 2010;85:1589–1602.

26 Triola MM, Huwendiek S, Levinson AJ, Cook DA: New directions in e-learning research in health professions education: report of two symposia. Med Teach 2012;34:15–20.

27 Grijpink-van den Biggelaar K, Drop SL, Schuwirth L: Development of an e-learning portal for paediatric endocrinology: educational considerations. Horm Res Paediatr 2010;73:223--230.

Supplementary table 1 – developed guidelines

Table 1. Table provided to the assessors to score the answers on the open question from Case 1 regarding CAH. For each qualification from insufficient to excellent a description is provided for the identified key elements.

KEY ITEM	POOR	INSUFFICIENT	REASONABLE	GOOD	EXCELLENT
Setting	not mentioned	On the ward/ no privacy	mentioned	private room; no interruption by colleagues, personnel etc.; no beepers, mobile phones.	**All issues are as in the category GOOD, and to perform EXCELLENT the fellow shows empathy and…** **Reassurance;** Reassures a normal life expectancy and that fertility is possible. **Questions:** provides time for parents' questions and concerns **Checks understanding**: checks whether the mother and father understand the main messages, such as the diagnosis and therapeutic consequences, adjustment of medication dosages, and stress medication
Time	not mentioned	Limited time (< 15-20 min)	mentioned	Approx. 30-45 min	
Reassurance	Not mentioned	Not mentioned	Mentioned without details	Mentioned with details	
One spokesman on behalf of DSD team:	no prior feedback with DSD team	DSD team not mentioned	mentioned	One spokesman DSD team or jointly with medical team psychologist (if available)	
Explanation of diagnosis:	Incorrect info or info literally copied from medical textbooks	Relevant items not or incompletely mentioned	Most items mentioned but limited explanation	a. adrenal disorder termed congenital adrenal hyperplasia, abbreviated as "CAH" with prenatally increased levels of "male-type" hormones, androgens b. uterus and ovaries present	
Treatment	Incorrect information	Life-long medication not or incompletely mentioned Stress medication not mentioned	Life-long and stress medication mentioned but with limited explanation	a. corticosteroid treatment (life-long) will stop virilisation. b. salt loss requires fludrocortisone treatment (life-long) c. instruction for administration of stress medication	

KEY ITEM	POOR	INSUFFICIENT	REASONABLE	GOOD	EXCELLENT
Follow up:	not mentioned	Relevant items not or incompletely mentioned	Some but not all items mentioned but limited explanation	Regular follow up for adjustment of dosage of medication and reminder of instructions for administration of stress medication is essential; discuss importance of adherence to prescribed treatment and consequences of non-adherence.	**Written information**: provides a written summary of the discussion **Arranges meeting with parent/patient**: offers to arrange in clinic a meeting between this family and another family who are caring successfully for a child with CAH **Reading material**: provides any reading materials published by support groups, or contact information for local and/or online support groups
Long term implications:	not mentioned	Relevant items not or incompletely mentioned	Some but not all items mentioned but limited explanation	The clitoris is expected to become less noticeable as your daughter grows; we will make a joint appointment with the whole team including the surgeon (or urologist or gynaecologist) to check on this in about 3-4 months to discuss whether or not genital surgery is an option.	
Genetic counselling :	Incorrect information	Genetic aspects not mentioned at all.	Genetics mentioned but not explained	Explanation of the genetics of CAH and risk for future pregnancies, also options for pre-implantation genetic diagnosis. *Note – in some institutions this will be done by genetics consultant or counsellor rather than endocrinologist. Need to raise the issues that must be discussed, and list options for who will undertake this discussion.*	
Other	blunt approach, lacking empathy	blunt approach, lacking empathy		shows explanatory pictures or drawings	

Table 2. Table provided to the assessors to score the answers on the open question from Case 2 regarding Gonadal dysgenesis. For each qualification from insufficient to excellent a description is provided for the identified key elements.

KEY ITEM	POOR	INSUFFICIENT	REASONABLE	GOOD	EXCELLENT
Setting	not mentioned	- no privacy - parents not invited by arrangement with Ranya - the parents are addressed rather than Ranya - Ranya is addressed as 'she' rather than 'you'	mentioned	make an appointment with Ranya and both parents[1]; Private room; no interruption by colleagues, personnel etc.; no beepers or mobile phones.	**All issues are as in the category GOOD, and to perform EXCELLENT the fellow shows empathy and…** **Reassurance:** you are healthy **Time for questions:** Provides ample time for discussion and response to questions and concerns of patient and parents. **Checks understanding:** checks whether Ranya, her mother and her father understand the main messages **Implications** • to offer check of BMD • sexuality issues are recognised and to be discussed later plan follow up with psychologist • Because your condition involves matters of a personal nature, you only need discuss it with your close friends and family.
Time	not mentioned	Limited time (< 15-20 min)	mentioned	45-60 minutes; ensure that patient/ family is aware of the time allotment. If necessary, follow up in following days/ week	
Reassurance	Not mentioned	Not mentioned	Mentioned without details	Mentioned with details	
One spokesman on behalf of DSD team:	no prior feedback with DSD team	DSD team not mentioned	mentioned	Possibly jointly with medical team psychologist or social worker if one is available with experience with DSD.	
Explanation:	- Incorrect info; - Info literally copied of medical textbooks; - Suggesting gender change.	- Relevant items not/ incompletely mentioned - explanation from viewpoint chromosomal error - thus Ranya being a girl by default	Most items mentioned but limited explanation	**Contains the following key elements:** 1. Typical development of ovaries/ gonads and atypical development in gonadal dysgenesis 2. Genes and chromosomes 3. Hormones and hormone replacement 4. Female reproductive anatomy 5. Fertility/infertility 6. Potential tumour risk	

KEY ITEM	POOR	INSUFFICIENT	REASONABLE	GOOD	EXCELLENT
Therapeutic issues:	Incorrect information	Relevant items not or incompletely mentioned	Some but not all items mentioned but no or limited explanation	Hormone replacement during the teenage years, (oestrogen, and later progesterone) will help Ranya to go through the physical changes of puberty in a similar pattern to other girls. (These hormones can be provided in many ways (pills, patches, sprays, creams, etc.) and have to be continued throughout adulthood to provide Ranya with the hormonal environment of a typical woman.	**Written information:** provides a written summary of the discussion Reading material: provides any reading materials published by support groups, or contact information for local or online support groups Offering follow up with psychologist to discuss private issues such as sexual relationships; infertility; options to have a partner, a family.
Long term implications:	not mentioned	- Relevant items not / incompletely mentioned - Questioning female gender identity	Some but not all items mentioned but no or limited explanation	Sexual relationships are discussed; infertility; options to have a partner, a family.	
Follow up:	not mentioned	Relevant items not / incompletely mentioned	Further steps not mentioned and/or not explained	Outpatient clinic appointment for initiation of oestrogen treatment; subsequent appointment with whole team including surgeon/urologist/ or gynaecologist	
Other	blunt approach, lacking empathy	blunt approach, lacking empathy	Empathy	empathy	

Supplementary table 2

Illustrative examples of text fragments used to explain condition or treatment in case 1(congenital adrenal hyperplasia)

Poor	The child has a disease named Congenital Adrenal Hyperplasia, which is mostly caused by CYP21A1 enzyme deficiency marked with elevation of 17 OH progesterone and compensated by testosterone enhancement that may cause the genital organ of the girl look like a boy.
Insufficient	Information will be provided repeatedly in small portions, but according to my opinion the most important point is to inform the parents about the sex (in this case female) of the child. They will be informed that it is crucial that the girl will need lifelong therapy in order to prevent health crises, but there is a very good chance that the baby will be able to develop completely normal.
Reasonable	The results have shown us that baby has a disorder of the adrenal gland called congenital adrenal hyperplasia where one of the key enzyme of the machinery is missing causing the adrenal to function in overdrive and to compensate for the missing enzyme by producing androgenic or masculinising hormones. These hormones cause the female baby to have such external genitalia The prevalence of the disease is 1 in 16000 and it can affect both the sexes equally though it is more easily picked up in females The ultrasound has shown us that the baby has a uterus and ovaries.
Good	The adrenal glands produce hormones that help to regulate the blood pressure, salt levels, blood sugar, and sex hormones. Your baby is missing an enzyme that is needed for normal production of the hormones that regulate salt and blood sugar, and subsequently more sex hormones, including testosterone, are being produced. This is a treatable condition, and we expect your child to live a full life if following up and taking medication appropriately.
Excellent	*Treatment* When we treat with the missing product that is cortisol or steroid the overdrive will stop and the male hormones will go down. A sheet explaining the symptoms of cortisol deficiency as well as excess will be given in easy language and graphic format. Stress cover and medical bracelet will be given

Illustrative examples of textfragments explaining the condition in case 2 (46,XY complete gonadal dysgenesis)

Poor	She does not have the internal genital parts such as a normal uterus, or ovaries that produce the female hormones based on the information that we have from the ultrasound. This means that if she wishes to remain a girl she would need assistance in order to get through puberty and fully develop into a woman with oestrogen. She should be able to have menstrual periods with hormone replacement but most likely she will not be able to have children of her own (she is missing the genetic cells-"eggs" that you would normally find in ovaries). She might be able to carry a child but we are not sure of that yet. *The passage above was regarded poor by suggesting gender change and by providing incorrect information*
Insufficient	The normal development of the gonads will be discussed, and it will be explained how Ranya has XY karyotype but (probably) an uterus and female external genitalia. It will be explained that gonads containing y material are at risk of developing malignancy and that gonadectomy is indicated. *The passage above was regarded insufficient as it is not understandable for a seventeen year old girl.*
Reasonable	General information about the development of sexes, the multitude of factors which determine sex and fragility of the sex determining factors. We will talk about the differences of sex and gender, about the differences between internal and external sex. We will tell her, that one of these sex determining factors is altered in her case and that there is a discrepancy between these two aspects in her case. *Marked as reasonable explanation but rather brief and not detailed.*
Good	Your body was made differently. (trying to get her background about ovaries) Each girl has 2 ovaries inside her tummy at the size of a grape, the ovary is important in making eggs so women can make babies, also it gives female hormones "oestrogen" which make changes for girls during puberty, so it make breasts and make uterus grow to bleed every month. Your body was made differently; so you don't have ovaries and they were replaced by non-functioning tissues.
Excellent	Contents: Simple and understandable explanation of genital development in general as a complex coaction of different genes and hormones coming from the same background in both males and females. Due to a yet unknown reason (several possible gene defects) Ranya's gonads haven't developed the usual way and therefore don't produce hormones, which can also be seen from the laboratory data. In utero, if there aren't any "male" hormones (androgens), the baby develops an uterus and vagina and female external genitalia. Later in life, some androgens are also produced in the adrenal gland, so that Ranya has developed pubic hair. However, hormones from the gonads are missing which means that she won't be able to go through puberty by herself. Furthermore the gonads have not developed as usual which would be necessary for fertility. Explanation that with a 46,XY-karyotype, the gonads would usually be testes, should be given..

Supplementary table 3 – ratings of assessors

Supplementary table 3. Overview of all assessors with representation of respectively the lowest and the highest score given; the average score and the standard deviation on a scale of 1-5. The last five columns summarizes the total assessments given per category. One expert (nr 20 in this table) was regarded as an outlier with an average rating of 4.46 on a scale of 1-5 and standard deviation of 0.68. This assessor and the assessors who did not score all responses (respondent 18 and 19) were excluded from further analysis.

					Assessment (1 poor – 5 excellent)				
Assessor	**MIN**	**MAX**	**AVERAGE**	**SD**	**1**	**2**	**3**	**4**	**5**
1	1	5	3,74	1,25	15	4	24	49	42
2	1	5	3,43	0,98	10	1	61	45	17
3	1	5	2,92	1,17	26	12	51	37	8
4	1	5	2,86	1,28	26	28	33	33	14
5	1	4	2,81	1,20	33	13	35	53	0
6	1	5	3,25	1,17	20	9	33	62	10
7	1	5	3,04	1,42	35	5	36	35	23
8	1	5	3,04	1,32	29	12	33	45	15
9	1	5	2,42	1,16	38	33	37	21	5
10	1	5	2,69	1,31	38	15	46	21	14
11	1	5	2,26	1,16	54	11	52	14	3
12	1	5	2,86	1,28	30	17	42	32	13
13	1	5	3,53	1,29	16	10	32	39	37
14	1	5	3,72	1,22	14	6	22	53	39
15	1	5	2,92	1,19	26	15	45	40	8
16	1	5	2,80	1,20	34	9	42	48	1
17	1	5	2,40	1,16	45	16	50	20	3
18	1	5	3,20	1,21	20	11	36	49	14
19	1	5	3,64	0,92	7	4	33	70	15
20	3	5	4,46	0,68	0	0	14	45	75

Supplementary table 4 – rating of text fragments by assessors

Table 1. Text fragments for case 1 (CAH) with examples of text passages which are respectively rated by assessors as poor/insufficient for the first row; as good/excellent for the second row and for the third row with great variation in the ratings given by the assessors.

Text fragments	Rating by assessors
Explanation – part 1 fellow 4 case 1 This medical condition implies that: your baby is genitically a girl with a karyotype of 46 XX but with a genetic disorder in a protein that drives to a virilisation intrauterus by male hormones increase.	Poor (6) Insufficient (5) Reasonable (6) Good (0) Excellent (0)
Explanation – part 3 fellow 5 case 1 I would make a drawing of the adrenals and explain what the adrenals do. " the adrenals is a small triangular organ, located above the kidneys. There are a small organ, but responsible for three hormones. The mineralocorticoids (which is responsible for are salts in our blood), the glucocorticoids (cortisol, which we need for waking up and stress situations) and the androgens (which are the masculine hormones). All these tree hormones are made from cholesterol which comes from the blood into the adrenals and is than covered in the 3 hormones. The conversion to these three hormones is done by many different steps (I will draw many arrows so that they can visualize it). What we know now is that in your girl, one of the steps is blocked. This is because of the enzyme is not formed yet and the step is blocked. The step of forming cortisol is blocked, so like you see (I draw it on the picture), the hormone before is accumulated, which we call 17-OH progesterone. And this is what we see in the blood of your daughter that 17-OH progesterone is high. Cortisol level is lower and because our body wants to make sure that there is enough cortisol in the blood, the body will stimulate the adrenals more and more for producing more cortisol. But because there is a block, it does not work, but instead of that the masculine hormones are produced more and that make that the clitoris will grow.	Poor (0) Insufficient (0) Reasonable (1) Good (10) Excellent (6)
Follow up – fellow 3 case 1 We have to start treatment today with these two drugs. During the first weeks you come to our hospital quite often to check the medication. Later you will see us everything 3 month.	Poor (6) Insufficient (0) Reasonable (7) Good (1) Excellent (3)

Table 2. Text fragments for case 2 (Gonadal Dysgenesis) with examples of text passages which are respectively rated by assessors as poor/insufficient for the first row; as good/ excellent for the second row and for the third row with great variation in the ratings given by the assessors.

Passages	Rating by assessors
Follow up – fellow 1 case 1 The girl most likely will have a female gender identity at adulthood, and therefore we do not doubt about assigning the female sex. The girl will need at least one (probably more) operations to make the external genitalia also look female.	Poor (11) Insufficient (2) Reasonable (3) Good (1)
Explanation - part 2 fellow 9 case 2 The dysfunctional testis are of no use to Ranya and need to be removed as they will be a high risk for cancer to the tune 12-25 %	Poor (0) Reasonable (4) Good (10) Excellent (3)
Explanation – part 1; fellow 1 case 2 I tell her that we found the reason why she is not getting into puberty. That she doesn't make 'female hormones'. I ask her permission to further explain this to her.	Poor (5) Insufficient (0) Reasonable (6) Good (2) Excellent (4)

CHAPTER 8

Discussion

Effective doctor-patient communication is a central clinical function in building a therapeutic doctor-patient relationship, which is the heart and art of medicine[1]. Studies on doctor-patient communication have demonstrated that patient discontent can be present even when doctors considered the communication adequate or even excellent.[1 2] A large majority of patient dissatisfaction and ensuing complaints arise from problems in doctor-patient communication.[1 3 4] Communication skills for future medical specialists are mostly taught and acquired during undergraduate training and further optimized during postgraduate medical training.

The main question of this thesis is how can we best support the postgraduate training of communication skills for future medical specialists. Prior research on this theme has been performed in other medical fields, such as general practice, surgery and oncology.[5-7] This thesis looks into the field of rheumatology as well as rare diseases.

Rheumatology is a field of expertise with long-term doctor-patient relationships. Changes in disease activities and life events ask for frequent treatment adjustments.[8-11] Good doctor-patient communication and patient involvement are associated with better outcomes and care satisfaction.[12-14] Our first research questions were how are communication skills incorporated into postgraduate training in the field of rheumatology and how does this relate to the evidence-based principles of communication training. This was followed with further questions on how rheumatologists in training think about Shared Decision Making, whether they use it in daily practice and which barriers they perceive.

Doctor-patient communication training in rare diseases is challenging due to the low exposure of relevant clinical cases and the limited number of experts for feedback. Therefore, a specific question of this thesis was how to improve doctor-patient communication in rare diseases and how online learning can be used for this. The first question was how electronic forms of learning, consultation and patient information can be applied to improve doctor-patient communication. The second question was to what extent online learning can be used to improve postgraduate training in rare diseases. The third question was how can online learning be used to train communication competences in rare diseases by the use of formative assessment and feedback. The fourth question was if formative feedback on communication skills can be optimized by using a rubric format addressing various aspects of communication.

Details of the performed research and the results in this thesis are discussed in the summary. This discussion will first reflect on the findings in the field of rheumatology and secondly on the findings regarding the training of communication skills for doctors that have patients with rare diseases. Finally, findings will be discussed related to the three main aspects from postgraduate communication training. These are ‘Awareness and

Reflection', 'Deliberate practice with repetitive practice and good feedback' and the role of a 'Supportive Environment'.

Postgraduate communication training in the field of rheumatology

The backbone of communication training is workplace learning. Our scoping literature review showed three main groups describing evidence-based principles of postgraduate communication training, which are 1) Awareness and reflection, 2) Deliberate practice and 3) Environment. Key elements related to the first group 'Awareness and reflection' are the confrontation with and awareness of own behaviour[15], debriefing and reflection[16-19] and personalized, well-defined learning goals[6 20]. Key elements related to the group 'Deliberate practice' are personalized, informative, structured, constructive feedback by supervisor, peers, self or patients[6 7 16-28], deliberate practice[7 16 20 28-30] and sufficient time spent on repeated experiential training and practicing skills[5 18-21 23 26 28 31-34]. Key elements related to the third group 'Supportive environment' are a rich clinical context with opportunities for immediate feedback, reflection and corrections[30 32 34 35], a trained and competent facilitator/teacher[6 17 18 20], a working environment that supports the teaching and learning of communication for staff and residents[7 15 31 35] and safety[15]. Rheumatologists-in-training spent most of their time being in attendance of rheumatologists and trial-and-error in order to improve their communication skills. Key elements of evidence-based communication training are rarely represented in these methods. Trial-and-error often lacks active reflection and adjustment of the communication approach. Observation by a supervisor with direct feedback, video-on-the-job followed by feedback and training with actors were experienced as most valuable by rheumatologists-in-training. Although highly valued, these methods were rarely used during postgraduate training, which was largely attributed to organizational barriers.

Rheumatologists-in-training report that doctor-patient communication in general and more specifically Shared Decision Making (SDM) are important. The possibility to improve adherence to therapy and the doctor-patient relationship was mentioned repeatedly as an advantage. Furthermore, they stated that SDM is indispensable within the field of rheumatology and should be obligatory. Less than a third of the rheumatologists-in-training indicated that they use SDM on a daily basis. Discussing patient's preferences is reported only by 33% of the rheumatologists-in-training on a regular basis. A need is expressed for more training, regardless of the level of experience. The main interest is for feedback about own performance on SDM and concrete and practical advice on how to apply SDM in daily practice. SDM in rare diseases is experienced as more difficult, which means specific attention is needed for performing this skill. Finally, identified barriers in the implementation of SDM in daily practice are 1) Unclearness of the concept of SDM, 2) Lack of ability and skills, 3) Misjudgment of patient preferences, 4) No recognition that SDM is not applied and 5) Uncertainty in treatment decision and outcome. Barriers which were found in other studies, such as a potential negative attitude towards SDM and time constraints, were not mentioned in our study.

Postgraduate communication training in rare diseases

To study postgraduate communication training in rare diseases we looked into the field of disorders of sex development (DSD), a subspecialty within the field of paediatric endocrinology. To support distance learning in this domain, the European Society for Paediatric Endocrinology (ESPE) online learning portal is used, which was developed to support clinical reasoning in DSD (providing chapters with background information and real-life cases). If (evidence-based) information is really scarce, a physician needs e-consultation on a global level to be sufficiently equipped for SDM in rare diseases. I-DSD is an example of a diagnostic consultation platform for rare diseases and disorders involving a network of centers of expertise. In addition, various examples of e-information for patients are provided, which are the result of a shared enterprise of clinicians and patient/family stakeholders. These e-information resources aim to help individuals to understand DSD diagnoses comprehensively, to support self-management of patients and enhance individual confidence.

The ESPE Online portal was evaluated by different user groups, varying from undergraduate to postgraduate, general paediatricians and paediatric endocrinologists. Various CanMED competencies were trained in the online learning module, including the role of Communicator. It was appreciated by users to not only receive feedback on medical aspects, but also on communication aspects. Personalized feedback on communication issues by various medical specialists was highly valued. Medical specialists in training specifically valued feedback on cultural aspects, as they felt it raised their awareness. Analysis of the replies on open ended assignments on communication aspects showed a wide variation in the replies of medical specialist in training. Important aspects such as the reassurance of patients and relatives were often lacking. In order to standardize assessment and feedback on various important communication aspects a rubric model was developed. The assessment of communication aspects by medical experts worldwide by use of the developed rubric model showed great intra-rater variability. This illustrates that there is a great variation between medical specialists' view on effective communication. It raises the question on whether it is possible to achieve consensus on how to assess communication skills. Therefore, it is important to be aware of communication differences, to reflect on these differences and to discuss differences between colleagues and also with our patients. All these different views on how communication aspects are perceived can help us to become better communicators. Therefore we plea for the ongoing training of communication skills with repetitive feedback by various assessors.

Awareness and Reflection

Confrontation with and awareness of one's own behavior starts with insight into the performance on communication skills. This thesis shows that residents' insight into the actual performance on doctor-patient communication requires improvement. This is illustrated by the barriers in the implementation of SDM in the field of Rheumatology, which can be related to a lack of awareness (chapter 3). Medical specialists in training indicate that some patients might not be 'fit' for Shared Decision Making, whereas every patient might benefit from a personalized approach taking into account their values and preferences. Another barrier is that medical specialists in training indicate they already practice the principles of Shared Decision making, although recent studies show differently[36]. It is striking that the self-rating for all the steps of Shared Decision Making is moderate to good, while patient preferences are not discussed on a regular basis when starting new treatments (chapter 3).

Shortcomings in awareness are not unique for the field of rheumatology, but were also signaled amongst medical specialists in training in the field of paediatric endocrinology. In the communication with patients and parents in an online assignment regarding a newborn with ambiguous genitalia, key items such as reassurance, sex assignment and informing family and friends were mostly missing (chapter 7). This illustrates the lack of awareness on these items, which can be explained by limited experience. Considering the fact that these conditions are rare, this is not surprising. Targeted feedback on these aspects raised awareness of cultural, religious aspects and sensitive issues (chapter 6). Paediatric endocrinologists in training were glad to receive feedback on these items and indicated that they would act differently in future situations due to increased awareness. This illustrates that reflection on shortcomings leads to improvement.

The issues described above illustrate a common issue, which is seen in many disciplines. Learning begins with the confrontation and awareness of one's own behaviour[15], implying that awareness is essential to improve communication skills. Awareness resulting in a clear learning question and reflection is a factor for success during communication training amongst rheumatologists-in-training (chapter 2). Video-on-the-job, observation and feedback and training with actors are helpful to raise awareness, but are not frequently applied during the training of rheumatologists in the Netherlands (chapter 2). These kind of training methods should be implemented on a regular basis to increase awareness of own behavior and to improve reflection on communication aspects.

Video-feedback based on performance in real-life settings seems to be associated with a significant increase in self-perceived empathy[17]. It appears to be an opportunity for a deeper level of self-assessment, peer-feedback and reflective practices[17 24 37]. Video feedback from real-life settings is very rare during the training of rheumatologists and mostly takes place during the first three years of Internal Medicine (chapter 2). Barriers mentioned by educators and medical specialists in training are mainly organizational and technical (chapter 2). This thesis confirms the importance of personal awareness in regards to patient communication. Various methods are available to support this process, but these are used sporadically. No one seems to be responsible for the initiation and scheduling of video-taping consultations. In order to improve this, there needs to be a combined responsibility of staff and medical specialists in training, both responsible for the frequent scheduling of consultation video-taping followed by self-reflection and multisource feedback. The frequency should be at least several times a year. Regular observations of real-life consultation and feedback on communication aspects should be implemented also several times a year. Finally, a training with actors should be implemented at least once to raise awareness of unknown behavior and stimulate reflection on communication competencies.

Deliberate practice with repetitive practice and good feedback

Deliberate practice is the principle of improving skills by continuous practice, feedback on performance, reflection on shortcomings and searching for possibilities to refine[27 28]. This is an important basis for optimizing all sorts of skills, from playing chess to racing and from making music to doctor-patient communication[7 20 30 38]. Medical students usually achieve a basic level of communication skills early on in their studies. However, after achieving these basic skills little progress is seen during the rest of their studies and in their careers as doctors[30]. In order to develop into a skilled communicator, deliberate practice is needed. This paragraph discusses two important aspects of deliberate practice, which are repetitive practice and feedback. Reflection is also important and has been addressed before in the section on Awareness and Reflection.

The first condition for repetitive practice is sufficient time spent on repeated experiential training and practicing skills.[5 18-21 23 26 28 31-34] The cornerstone of postgraduate medical training is workplace learning in clinical practice. Seeing patients, reflection and improving skills form the basis of learning. However, the effect of workplace learning on improving communication skills is limited. Rheumatologists in training indicate that during workplace learning they rarely pay attention to improving and refining communication skills (chapter 2). Furthermore, feedback after observation is mostly medical and rarely addresses communication aspects (chapter 2). As a result, important aspects of 'deliberate practice' are missing, resulting in only 'practice'. Therefore, there is a plea to introduce deliberate practice of communication skills in daily practice in postgraduate training. This can be easily achieved by structurally asking patients feedback about perceived communication, by asking them to actively summarize the content of the consultation and by asking them how the doctor can improve. This should be scheduled on a regular basis, for example every first day of the month. Medical specialists in training need to be asked to reflect and state learning goals. Furthermore, feedback on communication aspects by medical experts should be improved by scheduling regular feedback sessions focusing on communication.

An important aspect of deliberate practice is frequent training of skills in various situations. This is especially challenging in the training of communication skills for rare diseases and rare situations. An online learning platform with real-life cases with assignments and feedback was experienced as valuable by medical specialists (in training) (chapter 4-7). Feedback is an important aspect in online learning. The ESPE online learning platform provides multiple choice and open assignments with standard feedback (chapter 4-7). Feedback models were prepared by a team comprised of medical experts, a psychologist, a communication expert, an educationalist and patient representatives. This feedback was perceived as helpful by medical specialists in training. Especially, the detailed answer models for the explanation of various disorders of sex development for patients and parents were appreciated for the clear and explicit explanation and the use of wording (chapter 6). The online platform also provided multi-source personalized feedback by multiple medical experts from various continents on specific communication questions. This personalized multisource feedback was highly validated (chapter 6). Reactions of the medical specialists in training on the received feedback was mostly enthusiastic and the personal approach was appreciated. The feedback on non-medical aspects was especially valued, such as regional and ethical issues and the impact on social aspects for patients. Furthermore, confirmation on their performance was important for the medical specialists in training (chapter 6). This need for confirmation was also expressed by rheumatologists in training (chapter 2).

Online learning in order to train communication skills in difficult issues of rare diseases can be helpful for other medical specialties. Rheumatologists in training experience most problems with communication regarding rare diseases and rare treatment options (chapter 2). Furthermore, SDM is experienced as more difficult in rare diseases and in situations where evidence from large studies is missing. Especially in these situations, Shared Decision Making is of utmost importance as the values of the patient might be the most influencing factor in the final medical decision (chapter 3). Extensive answer models prepared by a team of medical experts, communication experts and patient representatives should be combined with real-life cases discussing communication issues.

An intriguing question is what defines ‘good’ feedback. In literature this is described as personalized, informative, structured, constructive feedback by a supervisor, peers, self or patients[6 7 16-28]. In a search to improve assessment of doctor-patient communication in the field of DSD, guidelines for assessment were developed using a rubric (chapter 7). This study showed that there was a remarkable variation in the responses to communication assignments amongst medical experts in training (chapter 6 and 7) and in the assessment of the responses amongst various medical experts. The rubric model did not result in a uniform assessment (chapter 7). From this we can conclude that it is nearly impossible to give uniform feedback on communication aspects. There is large variation in quality and the use of empathy between medical experts (in training), partly explained by regional backgrounds. This stresses the importance of continuous feedback by various assessors from all kinds of backgrounds, in order to develop a rich source of approaches that can be adjusted to the individual patient.

Supportive Environment

Awareness of shortcomings in communication is the first step in improving doctor-patient communication skills. Deliberate practice and feedback are necessary in order to refine communication skills. However, a safe and stimulating environment are essential in the training of communication skills for future medical specialist.

Safety[15] is of great importance in learning. Educators should be aware that medical specialists in training fear being judged when they ask for feedback. Anonymity was highly valued by medical specialists in training in the online learning platform who received personalized feedback on communication aspects (chapter 5). Despite the advantages of video-on-the-job, educators should be aware of the struggle medical specialists in training experience related to the stress of recording and watching oneself on video[17].

The importance of a well-trained and competent facilitator/teacher[6 17 18 20] deserves special attention. This is supported by the experiences amongst rheumatologists in training, who indicate this as a key factor of success in communication interventions (chapter 2).

The studies in the field of DSD show that there is great variation amongst medical experts in the assessment of ‘good’ communication skills. This pleas for ongoing attention for and reflection of communication aspects. Feedback should not only address medical specialists in training, but also stimulate feedback from medical specialists in training on the communication aspects of their supervisors.

Last but not least, we must be aware that a working environment that supports the teaching and learning of communication skills is essential to the training of future medical specialists in chronic diseases to become skilled communicators. [7 15 31 35] This starts with awareness amongst current medical specialists that there is room for improvement that starts now.

Practice implications

Medical specialists in training, supervising specialists and educators should all be aware that there is room for improvement for the training of doctor-patient communication and techniques such as SDM. As stated before, observation and feedback are already embedded in postgraduate medical training, but are mostly used to improve technical medical skills and rarely used to improve communication skills.

It all starts with awareness, whereby medical specialists and medical specialists in training should regularly ask for feedback from their patients. Both groups should regularly inquire what patients understand and remember from the consultation and how this is perceived. Fixed moments for consultation should be scheduled, for example every first day of the month. Feedback should lead to adjusted learning goals and continuous reflection. This should result in an environment where communication is highly valued and thus, support for improvement is common practice.

Regular moments of observation and feedback should be the cornerstone of postgraduate medical specialist training for communication skills. This should be regularly scheduled, several times a year. Furthermore, at least once during medical specialist training there should be a scheduled training with a training actor to increase awareness and to train skills. Scheduling and discussion of the feedback should be a shared responsibility of a therefore chosen staff member and a medical specialist in training. Observation and feedback can be both in real-time by means of video-taping and afterwards with feedback. There are increasing opportunities to use mobile devices to register videos for online feedback. This enables more frequent possibilities for observation and feedback, also at a distance. This is regarded as safe since videos and feedback are stored in an encrypted environment. This should be supported by hospitals for use in daily practice. Security officers should support this process in order to improve patient safety and quality-of-care.

Specific attention is needed for further implementation of SDM, a specific form of doctor-patient communication. Skills for SDM are not routinely trained during postgraduate medical specialist training. There are various barriers that need to be addressed. Most notably, the concept itself lacks clarity. There are general misassumptions regarding the doctors thinking that they know what's important for their patients and whether the patients are interested in Shared Decision Making. These issues can be addressed using online learning, such as the one developed for the Dutch Society for Oncology (https://elearning.nfk.nl). Furthermore, regular moments of observation and reflection should be implemented, ideally by video-taping followed by feedback in small groups with medical specialists in training, a staff member and a communication specialist. In this way, assumptions about SDM can be discussed, resulting in more profound knowledge and skills.

Online learning platforms can be instrumental in improving communication and decision making in rare diseases and rare situations, such as specialized care during pregnancy or the discussion of treatment options in rare diseases. Online case-based learning including formative assessments dealing with communication issues can improve communication skills. Feedback models should be jointly developed with medical experts, communication experts and patient representatives. A safe network of medical specialists in rare diseases can stimulate an ongoing discussion on how to improve doctor-patient communication and shared decision making, even when evidence on treatment options are scarce.

General considerations

Chronologically, the first studies were performed in the field of training of disorders of sex developments and later studies were performed in the field of rheumatology. This chronologic order also reflects a continuous evolving vision on how to train postgraduate communication skills in daily practice. First study (chapter 4) focussed on the training of various CanMED competencies by use of the ESPE e-learning portal. This gave insight into how the portal was perceived by various user groups and in how feedback can be provided. This was just a first step into the search of how to train skills amongst medical specialists (in training). The study was mainly explorative, involving several small studies. Next step chronologically was to explore the possibilities of e-learning, e-consultation and e-information (chapter 3). This study gave insight into the added value of a joint approach of medical experts, communication specialists and patient representatives. Involving patient representatives and communication specialists in the feedback provided to medical specialists (in training) in online learning raises awareness of certain aspects, such as the importance of reassurance, the right use of wording and also social and cultural aspects. This is not only important for the e-learning of doctors, but also the e-information for patients. The study gives a rich overview of examples. In retrospection, I think that the plea for integration of e-learning, e-consultation and e-information is too farfetched. Next in line are the global studies on the use of the ESPE e-learning portal in order to train the competencies 'medical expert' and 'communicator' (chapters 6 and 7). These studies show that awareness on communication aspects among medical specialists in training can be improved. It shows that multiple personalized feedback by experts from different geographical backgrounds was most valued by medical specialists in training. Furthermore, it showed that the answer model as prepared by a group of medical experts, communications specialists and patient representatives gave insights into other aspects, such as the wording, explanation and cultural and religious aspects. A strength of the studies is the number of participating medical specialists in training and medical experts. Another strength is the diverse study team, involving medical specialists, an educationalist, communication experts and patient representatives. In retrospection, it would be interesting to find out whether the feedback from multiple, various medical experts is of added value compared to a standardized response prepared by a study team. This would require a different study approach. After all experience in the field of disorders of sex development, the question arose how medical specialists in training actually achieve their communication skills and how this relates to the findings in literature. The study in the field of rheumatology combines experiences from medical specialists in training and educators with evidence-based principles from literature (chapter 2). The strength of this study is the combination of these various viewpoints and the involvement of a diverse study group with medical experts, communication experts and a educationalist. A weakness of the study is the response rate, which was estimated to be higher than reported. SDM is indispensable in the treatment of chronic diseases. Prior studies showed various barriers in the implementation of SDM. There was no prior study that looked into the perceptions on SDM by medical specialists in training. The study in this thesis (chapter 3) is therefore of added value and shows the importance of more SDM training in the postgraduate training of rheumatologists. Strengths and weaknesses as mentioned for chapter 2 also apply for this study.

Future training and research

The practice implications as advised in this discussion will be challenging to implement into daily practice. It all starts with raising awareness on the importance of good doctor-patient communication. On a personal level, small improvements can be achieved by frequently asking patients how they perceive the doctor-patient communication and how this can be improved. It is also important to regularly check whether information is understood by asking patients to summarize this in their own words. On an institutional/regional level, there has to be a combined responsibility for the realisation of training by both a medical expert and the medical expert in training. On a national level, there should be an extensive cooperation between the national scientific committee, patient representatives, communication specialists and educationalists. Also, there has to be extensive cooperation with Chief Data Officers of hospitals in order to guarantee that secured video-taping applications for feedback by use of a personal mobile phone can be used. This last aspect unfortunately can take a lot of time.

Future research might focus on the added value of interventions to raise awareness amongst medical specialists in training and educators. Furthermore, I would advise implementation studies in order to search for approaches to implement the findings of this thesis.

References

1. Ha JF, Longnecker N. Doctor-patient communication: a review. The Ochsner journal 2010;**10**(1):38-43.
2. Stewart MA. Effective physician-patient communication and health outcomes: a review. Cmaj 1995;**152**(9):1423-33.
3. Halperin EC. Grievances against physicians: 11 years' experience of a medical society grievance committee. West J Med 2000;**173**(4):235-8.
4. Tamblyn R, Abrahamowicz M, Dauphinee D, et al. Physician scores on a national clinical skills examination as predictors of complaints to medical regulatory authorities. Jama 2007;**298**(9):993-1001.
5. Barth J, Lannen P. Efficacy of communication skills training courses in oncology: a systematic review and meta-analysis. Ann Oncol 2011;**22**(5):1030-40.
6. Stiefel F, de Vries M, Bourquin C. Core components of Communication Skills Training in oncology: A synthesis of the literature contrasted with consensual recommendations. Eur J Cancer Care (Engl) 2018;**27**(4):e12859.
7. van den Eertwegh V, van Dalen J, van Dulmen S, et al. Residents' perceived barriers to communication skills learning: comparing two medical working contexts in postgraduate training. Patient Educ Couns 2014;**95**(1):91-7.
8. Dures E, Hewlett S, Ambler N, et al. Rheumatology clinicians' experiences of brief training and implementation of skills to support patient self-management. BMC Musculoskeletal Disorders 2014;**15**:108-08.
9. Dures E, Hewlett S, Ambler N, et al. A qualitative study of patients' perspectives on collaboration to support self-management in routine rheumatology consultations. BMC Musculoskeletal Disorders 2016;**17**:129.
10. Dures E, Fraser I, Almeida C, et al. Patients' Perspectives on the Psychological Impact of Inflammatory Arthritis and Meeting the Associated Support Needs: Open-Ended Responses in a Multi-Centre Survey. Musculoskeletal Care 2016:n/a-n/a.
11. Elwyn G, Frosch DL, Kobrin S. Implementing shared decision-making: consider all the consequences. Implement Sci 2016;**11**:114.
12. Kjeken I, Dagfinrud H, Mowinckel P, et al. Rheumatology care: Involvement in medical decisions, received information, satisfaction with care, and unmet health care needs in patients with rheumatoid arthritis and ankylosing spondylitis. Arthritis Rheum 2006;**55**(3):394-401.
13. Brekke M, Hjortdahl P, Kvien TK. Involvement and satisfaction: a Norwegian study of health care among 1,024 patients with rheumatoid arthritis and 1,509 patients with chronic noninflammatory musculoskeletal pain. Arthritis Rheum 2001;**45**(1):8-15.
14. Nota I, Drossaert CH, Taal E, et al. Patient participation in decisions about disease modifying anti-rheumatic drugs: a cross-sectional survey. BMC Musculoskelet Disord 2014;**15**:333.
15. van den Eertwegh V, van der Vleuten C, Stalmeijer R, et al. Exploring residents' communication learning process in the workplace: a five-phase model. PLoS One 2015;**10**(5):e0125958.

16. Beaudoin PL, Labbé M, Fanous A, et al. Teaching communication skills to OTL-HNS residents: multisource feedback and simulated scenarios. J Otolaryngol Head Neck Surg 2019;**48**(1):8.
17. Dohms MC, Collares CF, Tibério IC. Video-based feedback using real consultations for a formative assessment in communication skills. BMC Med Educ 2020;**20**(1):57.
18. MacDonald-Wicks L, Levett-Jones T. Effective teaching of communication to health professional undergraduate and postgraduate students: A Systematic Review. JBI Libr Syst Rev 2012;**10**(28 Suppl):1-12.
19. Stigt JA, Koele JH, Brand PLP, et al. Workplace mentoring of residents in generic competencies by an independent coach. Perspect Med Educ 2018;**7**(5):337-41.
20. Maatouk-Bürmann B, Ringel N, Spang J, et al. Improving patient-centered communication: Results of a randomized controlled trial. Patient Educ Couns 2016;**99**(1):117-24.
21. Dacre J, Richardson J, Noble L, et al. Communication skills training in postgraduate medicine: the development of a new course. Postgrad Med J 2004;**80**(950):711-5.
22. Junod Perron N, Nendaz M, Louis-Simonet M, et al. Impact of postgraduate training on communication skills teaching: a controlled study. BMC Med Educ 2014;**14**:80.
23. Weiland A, Blankenstein AH, Willems MH, et al. Post-graduate education for medical specialists focused on patients with medically unexplained physical symptoms; development of a communication skills training programme. Patient Educ Couns 2013;**92**(3):355-60.
24. Hulsman RL, van der Vloodt J. Self-evaluation and peer-feedback of medical students' communication skills using a web-based video annotation system. Exploring content and specificity. Patient Educ Couns 2015;**98**(3):356-63.
25. Greco M, Brownlea A, McGovern J. Impact of patient feedback on the interpersonal skills of general practice registrars: results of a longitudinal study. Med Educ 2001;**35**(8):748-56.
26. Berkhof M, van Rijssen HJ, Schellart AJ, et al. Effective training strategies for teaching communication skills to physicians: an overview of systematic reviews. Patient Educ Couns 2011;**84**(2):152-62.
27. Ericsson KA. Deliberate practice and the acquisition and maintenance of expert performance in medicine and related domains. Acad Med 2004;**79**(10 Suppl):S70-81.
28. Ericsson KA. Deliberate practice and acquisition of expert performance: a general overview. Acad Emerg Med 2008;**15**(11):988-94.
29. Brand PL, Stiggelbout AM. Effective follow-up consultations: the importance of patient-centered communication and shared decision making. Paediatr Respir Rev 2013;**14**(4):224-8.
30. Wouda JC, van de Wiel HB. The communication competency of medical students, residents and consultants. Patient Educ Couns 2012;**86**(1):57-62.
31. Wouda JC, van de Wiel HB. How to attain expertise in clinical communication? Paediatr Respir Rev 2013;**14**(4):213-8.
32. Aspegren K. BEME Guide No. 2: Teaching and learning communication skills in medicine-a review with quality grading of articles. Med Teach 1999;**21**(6):563-70.

33. Razavi D, Merckaert I, Marchal S, et al. How to optimize physicians' communication skills in cancer care: results of a randomized study assessing the usefulness of posttraining consolidation workshops. J Clin Oncol 2003;**21**(16):3141-9.
34. van Dalen J, Kerkhofs E, van Knippenberg-Van Den Berg BW, et al. Longitudinal and concentrated communication skills programmes: two dutch medical schools compared. Adv Health Sci Educ Theory Pract 2002;**7**(1):29-40.
35. Essers G, van Dulmen S, van Es J, et al. Context factors in consultations of general practitioner trainees and their impact on communication assessment in the authentic setting. Patient Educ Couns 2013;**93**(3):567-72.
36. Mathijssen EGE, Vriezekolk JE, Popa CD, et al. Shared decision making in routine clinical care of patients with rheumatoid arthritis: an assessment of audio-recorded consultations. Annals of the Rheumatic Diseases 2020;**79**(2):170-75.
37. Wouda JC, van de Wiel HB. The effects of self-assessment and supervisor feedback on residents' patient-education competency using videoed outpatient consultations. Patient Educ Couns 2014;**97**(1):59-66.
38. Beaudoin PL, Labbe M, Fanous A, et al. Teaching communication skills to OTL-HNS residents: multisource feedback and simulated scenarios. J Otolaryngol Head Neck Surg 2019;**48**(1):8.

CHAPTER 9

Summary

Chapter 1

The main question of this thesis is how we can support postgraduate training of communication skills for future medical specialists. Two main aspects are considered. The first aspect is the postgraduate training of communication skills in daily practice in chronic diseases. Secondly, specific attention is needed for communication skills concerning rare diseases for several reasons. Due to small numbers there is scarce experience with the treatment of these patients in general training centers. This requires another approach in training.

Extensive research has been performed regarding training of communication skills. Doctor-patient communication skills can be taught in courses, are learnt, but are easily forgotten if not maintained by practice. The most important enhancing factor in post-graduate communication training is encouragement to deliberately practice in an environment in which the value of communication skills is recognized and support is institutionalized with appropriate feedback from role models. Communication is a competency that should be longitudinally learned across the whole training continuum in an authentic setting with a lot of practice in many different contexts with regular meaningful feedback.

Question arises to what extent these elements are implemented in the daily practice of postgraduate training in chronic diseases. The field of rheumatology is highly suitable to look into this question. Besides general communication skills specific attention goes out to Shared Decision Making (SDM), supporting patients and healthcare professionals to jointly make a decision based on the best available evidence for treatment options while respecting patient's values and preferences.

There are several challenges in training doctor-patient communication in rare diseases. Online learning is evenly effective as classroom teaching and might offer a solution in rare diseases. In order to gain insight in training of doctor-patient communication skills in rare diseases by use of online learning we look into the field of disorders of sex development (DSD), which is a subspecialty of the paediatric endocrinologist. Care for individuals with DSD is characterized by varying short- and long-term challenges that may emerge throughout one's lifespan. This requires adequate doctor-patient communication skills.

Chapter 2

In this chapter we studied which methods are used to train doctor-patient communication skills amongst residents rheumatology in the Netherlands, how these are perceived by residents and educators and how these relate to evidence regarding postgraduate training of communication skills. We send out an online questionnaire, performed semi-structured interviews among rheumatologists in training and educators and performed a scoping literature search to find evidence-based principles of postgraduate communication training.

We summarize the main perceived needs and provide an overview of key elements that support improvement of communication skills in postgraduate training, based on a literature review. Evidence based aspects of communication training are 1) raising awareness of communication skills and reflection, 2) deliberate practice and 3) a safe and supportive environment.

An inventory among rheumatologists in training and educators in the field of rheumatology showed that the most common used elements in postgraduate communication training are the attending of the doctor-patient contact of different rheumatologists and secondly trial-and-error. Key elements of evidence based communication training, such as active participation and reflection are rarely mentioned. Trial-and-error approaches often lacks active reflection and adjustment of communication approach.

Experienced as most valuable by residents were observation with direct feedback, reflection and feedback on video-taped patient interactions and training with actors. However, these were rarely used during postgraduate training. Key elements that support learning and improvement of communication skills are awareness of shortcomings in communication, formulating a learning question prior to training, deliberate practice, reflection, good feedback and a supportive environment. This is in line with the findings in literature.

Recommendations for further training are regular (formative) assessments followed by feedback to raise awareness and to use deliberate practice to improve communication skills. Rheumatologists in training should be stimulated to ask feedback from patients and to formulate learning goals. Reflection and feedback on video-taped patient interactions can be very supportive. Staff should be trained to give adequate feedback on communication aspects. And last but not least, the environment should be safe and stimulating in order to continuously improve communication skills.

Chapter 3

Shared Decision Making (SDM) is an approach in doctor-patient communication which allows patients and healthcare professionals to jointly make a decision based on the best available evidence for treatment options while respecting patient’s values and preferences. *Chapter 3* looks into perceived competences, needs and barriers related to Shared Decision Making among rheumatologists in training.

We explored opinions on SDM among rheumatologists in training in the Netherlands, their experience with SDM, how they self-assess their skills on communication and SDM and to identify barriers in the implementation of SDM. We used an online questionnaire and performed a literature study.

Rheumatologists in training report that doctor-patient communication in general and more specific SDM is important. The possibility to improve adherence to therapy and the doctor-patient relationship was mentioned repeatedly as an advantage. Furthermore respondents state that SDM is indispensable within the field of rheumatology and should be obligatory. SDM is referred to as an ethical imperative. It is stated that it is necessary to investigate patients' values and preferences related to therapy.

Less than a third of the medical specialists in training indicate that they use SDM on a daily basis. Discussing patient's preferences is reported only by one in three of the rheumatologists in training on a regular basis. A need is expressed for more training, regardless of the level of experience. Rheumatologists in training are mainly interested in feedback about their own performance on SDM and concrete and practical advice on how to apply SDM in daily practice.

SDM in rare diseases is experienced as more difficult, which requires specific attention. Finally, barriers identified in the implementation of SDM in daily practice are 1) Lack of clearness of the concept of SDM, 2) Lack of ability and skills to implement it, 3) Misjudgment of patient preferences, 4) No recognition that SDM is not applied and 5) Uncertainty in treatment decision and outcome. Barriers which were found in other studies such as a potential negative attitude towards SDM and time constraints were not mentioned in our study.

Chapter 4

Doctor-patient communication and decision making in rare diseases is regarded by doctors as challenging for several reasons. First of all, in a general practice there is little experience with diagnosis, clinical variety and treatment options and effects. Secondly, due to small numbers, evidence about treatment options is limited and mostly based on case reports. Thirdly, patients are mostly referred to specialized clinics, which might implicate long-distance communication, especially in rural countries.

In *Chapter 4* we explored possibilities and good practices of e-learning, e-consultation and e-information in the field of disorders of sex development (DSD), a subspecialty within the field of paediatric endocrinology. To support distance learning the European Society for Paediatric Endocrinology (ESPE) e-learning portal is described, which is developed to support clinical reasoning providing background information and real-life cases.

If (evidence-based) information is really scarce a physician needs e-consultation across borders to be sufficiently equipped for SDM in rare disease. I-DSD is an example of a diagnostic consultation platform for rare diseases and disorders involving a network of centers of expertise. Standards of care and security need to be carefully set to ensure that patient data are secure. It has to be clear who is legally accountable for decisions and treatments offered through such a network.

Finally various examples of e-information for patients are provided, which usually are the result of a shared enterprise of clinicians and patient/family stakeholders. These e-information resources aim to help individuals to understand DSD diagnoses comprehensively, to support self-management of patients and enhance individual confidence. Care for individuals with DSD is characterized by varying short- and long-term challenges that may emerge throughout one's lifespan – from urogenital variation to concerns about fertility, gonadal malignancy, adrenal complications, hormone replacement, bone health and psychosocial support, among others. E-information can help patients and families to manage those issues positively and proactively as they are encountered, including age-appropriate material on various subjects.

In conclusion, there is a plea for integration of e-information, e-learning and e-consultation, combining patient experiences, expert knowledge and case examples.

Chapter 5

Chapter 5 gives an overview of experiences with the above described ESPE e-learning portal in different user groups within the field of Paediatric Endocrinology. Two cases were developed with each three extra feedback questions that enabled personalized feedback. Five small-scale pilot tests were conducted to evaluate the developed portal.

Altogether, 8 cases and 4 chapters were studied by a total of 71 people: 18 senior experts, 21 fellows, 10 medical students, 9 regional paediatric ians and 13 residents. Overall, the respondents were enthusiastic about the content of the studied two cases that were evaluated. On average, respondents were satisfied by the number and the quality of open and multiple-choice questions.

One pilot was conducted to evaluate personalized feedback given by experts to medical specialists in training. Each studied case contained three feedback questions, where the answer of the medical specialist in training was sent to an expert for personalized feedback. Medical specialists in training appreciated the opinion of another expert, who often brought in another perspective.

It was also appreciated that feedback was not limited to pure medical aspects, but also included communication skills. Anonymity was valued by students and experts, as no personal details were provided to the expert who gave the personalized feedback. Various CanMEDS competencies seem suitable to address in the portal, including the role of communicator and health advocate, whereby personalized feedback is experienced as useful.

Chapter 6

In *Chapter 6* we evaluated the role of e-learning in the training of the competencies 'medical expert' and 'communicator' among medical specialists in training in the field of paediatric endocrinology in a global setting. Special attention was paid to evaluate the added value of personalized feedback and the feedback by different experts with various backgrounds.

Senior paediatric endocrinologists (experts) and paediatric ians undergoing subspecialty training in paediatric endocrinology (fellows) were approached to participate. Fellows were asked to study two interactive cases on the subject of DSD in English. The two clinical cases are within the expertise of paediatric endocrinologists, which are a case related to a newborn with congenital adrenal hyperplasia and a case related to a girl with delayed puberty due to gonadal dysgenesis.

At crucial stages in the problem-solving process, three open ended questions were included for each case, where the fellow was invited to describe how (s)he would handle the situation, with particular focus on how the fellow would inform the parents and/or patient of the key information regarding diagnosis, implications and management.

The replies to the open-ended questions were submitted to two to three experts from various parts of the world. The experts were invited to reply with a brief formative commentary to the fellows' responses. They were free to use an example answer prepared by the study team.

Thirty-two experts and 68 fellows from various parts of the world (Africa, Asia, Australia, Europe, North America, South America) participated in the study. All fellows appreciated the personalized feedback by multiple experts, which is in line with prior studies. It was perceived useful to receive multiple reactions, illustrating that different viewpoints are possible and can be integrated into the fellows' approach. When all experts provided similar feedback, it was perceived as a message that this feedback was important.

The provided feedback raised awareness about certain aspects such as religion and informing family in the case of the newborn. The prepared answer model provided useful information about the different things you have to say to the parents and a very practical structure for starting the conversation. Also, fellows were glad to receive confirmation on their approach. Acknowledgements from the fellows suggest that participating in the study resulted in extended skills in the role of medical expert and communicator.

Chapter 7

In *Chapter 7* we further evaluated the fellows' replies to the open-ended questions from the global study on assessment and feedback in competency training in pedriatic endocrinology. These questions focus on how the fellow would inform the parents and/or patient of the key information regarding diagnosis, implications and management in a case related to a newborn with congenital adrenal hyperplasia and a case related to a girl with delayed puberty due to gonadal dysgenesis.

Our first aim was to give a thorough analysis of the replies on communication aspects by scoring the different key items such as explanation of diagnosis, reassurance, role of the multidisciplinary team and long-term implications. Secondly, we described the process of the development of a rubric, which assesses communication aspects in the replies. Our third objective was to evaluate the developed rubric.

First, the replies of the fellows were dissected into sentences relating to the identified key items. All passages were analysed and rated on a scale of 5 levels of quality of the answer. We found that there was substantial variation not only in the number of key items mentioned, but also in the quality of the responses. The items "explanation of diagnosis" and "treatment options" were most frequently mentioned. Remarkably, allotted time, explanation of the role of the DSD team and the relevance of providing long-term follow-up were rarely mentioned.

From this analysis, descriptors were defined, which describe how responses will be rated for all key items for different levels of performance. This allowed the creation of a rubric to score the fellow's explanations of the 2 conditions into 5 categories for accurateness, clarity, and empathy. This rubric model was then passed to the study team with paediatric endocrinologists, psychologists, educationalists and patient/parent representatives for refinement and consensus.

To validate the developed rubric twenty-two senior paediatric endocrinologists worldwide were invited to assess 10 selected replies of the fellows for each case, using the developed rubric model. The replies were cut into a total of 114 passages, relating to different key items as mentioned in the rubric model. All separate passages and the complete replies were rated by the experts using the rubric, resulting in 134 assessments per expert.

The assessment by the experts showed considerable variation. Phrases with a high level of rating agreement or a low level of rating agreement were identified. Consensus was reached when answers were very short or blunt, and rated as poor or insufficient. Rating was good or excellent when an answer was complete, mentioning relevant details and expressing empathy. Based on these findings, the rubric model was adjusted to a 3-level assessment, with empathy as a separate key item.

In conclusion, fellows worldwide showed a broad range of communication skills regarding completeness of the response, wording and the use of empathy. Certain relevant items such as long-term consequences, follow up and reassurance were not often discussed. Possibly this is the result of low exposure of fellows to patients with DSD due to the small number of patients even in large teaching centres. E-learning providing training in the approach of patients with DSD may be a welcome adjunct in fellowship training programs.

The study also showed considerable diversity in the assessment of fellows' replies by the participating senior paediatric endocrinologists. Multiple explanations are plausible. First, experts were not formally trained in the use of the rubric. Secondly, participating experts might differ in their opinions on how to communicate these kind of conditions. The third possibility is that experts themselves may differ in their communication skills. Cultural, religious or regional differences could not be related to the variation in assessment.

Chapter 8

In chapter 8, the discussion, we reflected on the findings in the field of rheumatology and secondly on the findings regarding the training of communication skills with patients with rare diseases. Finally, findings are discussed related to three main aspects from postgraduate communication training. These are 'Awareness and Reflection', 'Deliberate practice with repetitive practice and good feedback' and the role of a 'Supportive Environment'. Practice implications that follow from all research were discussed. The chapter ends with general considerations and suggestions for future training and research.

CHAPTER 10

10

Nederlandse samenvatting

Nederlandse samenvatting

Hoofdstuk 1

De onderzoeksvraag van dit proefschrift luidt: hoe kan de training van communicatieve vaardigheden in de medische vervolgopleidingen worden verbeterd voor toekomstige artsen. Er is hierbij aandacht voor twee verschillende onderwerpen. Ten eerste wordt gekeken naar de training van communicatieve vaardigheden bij de behandeling van chronische ziektes. Ten tweede wordt er gekeken naar communicatieve vaardigheden bij zeldzame ziektebeelden. Doordat deze ziektebeelden niet veel voorkomen is er minder ervaring met de behandeling. Dat vraagt om een andere aanpak bij de training en het aanleren van vaardigheden.

Er is al veel onderzoek gedaan naar het aanleren van communicatieve vaardigheden voor artsen. Deze kunnen worden geleerd , maar worden snel weer vergeten als ze niet worden gebruikt in de dagelijkse praktijk. Het continu trainen van communicatieve vaardigheden door middel van 'deliberate practice' is het meest succesvol in het aanleren van communicatieve vaardigheden in de vervolgopleiding wanneer er tevens sprake is van een omgeving waar goede dokter-patiënt communicatie wordt gewaardeerd en feedback wordt gegeven door rolmodellen. Het aanleren van communicatieve vaardigheden is een continue proces, waarbij ervaring moet worden opgedaan in verschillende situaties met regelmatig betekenisvolle feedback.

Het is de vraag in hoeverre deze elementen vertegenwoordigd zijn in de medische vervolgopleidingen gericht op chronische ziekten. Het vakgebied van de reumatologie is zeer geschikt om deze vraag te beantwoorden. Er is gekeken naar het aanleren van algemene communicatieve vaardigheden, maar ook naar het aanleren van 'Samen Beslissen', waarbij arts en patiënt gezamenlijk behandel beslissingen maken, gebaseerd op bewezen effectieve behandelmogelijkheden en de waarden en voorkeuren van de patiënt.

Voor het aanleren van communicatieve vaardigheden bij zeldzame ziektebeelden zijn er een aantal uitdagingen. E-learning is even effectief als klassieke onderwijsvormen en zou bij zeldzame ziektebeelden van toegevoegde waarde kunnen zijn. Om inzicht te krijgen in het trainen van communicatieve vaardigheden voor zeldzame ziektebeelden is gekeken naar een subspecialisatie van de kinderendocrinologie, gericht op differences or disorders of sex development (DSD). DSD is een verzamelnaam voor aandoeningen waarbij de geslachtsontwikkeling atypisch verlopen is. De zorg voor patiënten met DSD en hun ouders wordt gekarakteriseerd door zeer veel lange en korte termijn uitdagingen. Dit vereist goede communicatieve vaardigheden.

Hoofdstuk 2

In dit hoofdstuk hebben we gekeken welke technieken worden gebruikt voor het verbeteren van communicatieve vaardigheden in de medische vervolgopleiding voor de reumatologie. Verder is onderzocht hoe de effectiviteit van de technieken wordt ervaren door reumatologen in opleiding en door opleiders. Er is een literatuurstudie verricht naar bewezen effectieve elementen bij het trainen van communicatieve vaardigheden in de medische vervolgopleidingen. Er is gekeken in hoeverre deze elementen aanwezig zijn binnen de gevonden technieken in de reumatologie.

Bewezen effectieve elementen bij het trainen van communicatieve vaardigheden in de medische vervolgopleidingen zijn 1) bewustzijn en reflectie, 2) deliberate practice en 3) een veilige en ondersteunende omgeving.

Meester-gezel leren en werkplek leren zijn de hoekstenen voor het aanleren van communicatieve vaardigheden in de medische vervolgopleiding tot Reumatoloog. Gezamenlijke spreekuren met een Reumatoloog en trial-en -error zijn de meest genoemde methoden voor het aanleren/verbeteren van communicatieve vaardigheden. Bewezen effectieve elementen bij het trainen van communicatieve vaardigheden zijn hierbij nauwelijks aanwezig. Zo ontbreekt bij trail-en-error vaak reflectie en continue verbeteren van handelen.

Observatie gevolgd door feedback, video-opnames van consulten gevolgd door feedback en trainingen met trainingsacteurs worden als meest waardevol ervaren. Helaas worden deze technieken maar zelden ingezet binnen de medische vervolgopleiding. Kern elementen die werden genoemd door de reumatologen in opleiding en bijdragen aan het verbeteren van vaardigheden zijn bewustzijn ten aanzien van tekortkomingen, een leervraag voorafgaand aan de training, deliberate practice, reflectie op handelen, goede feedback en een ondersteunende omgeving. Dit is in lijn met de gevonden elementen in de literatuur.

Aanbevelingen voor het verbeteren van de medische vervolgopleiding zijn regelmatige beoordelingen gevolgd door feedback gericht op communicatie voor het verbeteren van bewustzijn. Reumatologen in opleiding zouden regelmatig feedback moeten vragen van patiënten en leervragen formuleren voor het verbeteren van vaardigheden. Video-opnames gevolgd door feedback kan hierbij zeer ondersteunend zijn. Ook opleiders zouden meer training kunnen krijgen in communicatieve vaardigheden en het geven van feedback in dit kader. Een veilige omgeving waarbij goed voorbeeld goed doet volgen is hierbij zeer belangrijk.

Hoofdstuk 3

Samen beslissen is het proces waarin de arts en de patiënt gezamenlijk bespreken welk medisch beleid het beste bij de patiënt past, waarbij alle opties, voor- en nadelen, patiëntvoorkeuren en omstandigheden worden meegenomen. In hoofdstuk 3 hebben we gekeken naar de visie van reumatologen in opleiding op samen beslissen, hun ervaring met samen beslissen, hun ervaren vaardigheden, behoeften en barrières in het invoeren van samen beslissen. Onderzoek is uitgevoerd door middel van een vragenlijst onder reumatologen in opleiding en literatuuronderzoek.

Reumatologen in opleiding vinden goede dokter-patiënt communicatie in het algemeen belangrijk, maar zeker ook samen beslissen. Samen beslissen wordt gezien als ethisch correct, wat therapietrouw en een goede arts-patiëntrelatie versterkt. Zij zien Samen beslissen al onmisbaar binnen de reumatologie. Zij vinden het belangrijk om de waarden en voorkeuren van de patiënt te verkennen en mee te nemen in de uiteindelijke behandelkeuzes.

Minder dan een derde van de reumatologen in opleiding gebruikt samen beslissen op een dagelijkse basis. Het is opvallend dat slechts één op de drie reumatologen in opleiding structureel patiëntvoorkeuren bespreekt bij het starten van een nieuwe behandeling. Er is een behoefte aan meer training in samen beslissen, ongeacht de mate van ervaring met samen beslissen en opleidingsduur. Reumatologen in opleiding willen graag feedback op hun huidige functioneren ten aanzien van samen beslissen en vragen om praktische tips.

Samen beslissen bij zeldzame ziektebeelden of behandelmogelijkheden wordt als extra lastig ervaren. Ten slotte is er een aantal barrières op te merken, die verdere implementatie van samen beslissen in de weg staan. Dit zijn 1) Onduidelijkheid van het principe van Samen Beslissen, 2) Onvoldoende kennis en vaardigheden in Samen Beslissen, 3)Verkeerd inschatten van voorkeuren van patiënten ten aanzien van Samen Beslissen, 4) Het niet toepassen van Samen Beslissen, terwijl dit niet als zodanig wordt herkend en 5) Samen beslissen bij onzekerheid ten aanzien van de beste behandeling en uitkomsten. In andere studies werden een negatieve attitude ten aanzien van samen beslissen en problemen ten aanzien van tijdsbewaking gezien als barrières. Deze werden in deze studie niet genoemd.

Hoofdstuk 4

Dokter-patiënt communicatie en samen beslissen bij zeldzame ziektebeelden is om meerdere redenen uitdagend. Ten eerste is er weinig persoonlijke ervaring met de diagnose en behandelopties. Ten tweede is er door lage patiëntaantallen weinig bewijs in de literatuur over effectiviteit van behandelingen. Ten derde worden patiënten vaak verwezen naar gespecialiseerde centra, wat communicatie op afstand soms noodzakelijk maakt, met name in ontwikkelingslanden.

In Hoofdstuk 4 worden de mogelijkheden en voorbeelden onderzocht ten aanzien van e-learning, e-consultatie en e-informatie op het gebied van DSD. Ten aanzien van e-learning wordt de e-learning portal beschreven die is ontwikkeld door de Europese Vereniging voor Pediatrische Endocrinologie (ESPE). I-DSD is een voorbeeld van een diagnostisch consultatief platform voor zeldzame ziektebeelden. Er worden verschillende voorbeelden gegeven van e-informatie voor patiënten. Deze websites zijn vaak tot stand gekomen door nauwe samenwerking tussen artsen en patiëntvertegenwoordigers.

Het hoofdstuk sluit af met de gedachte dat e-informatie, e-learning en e-consultatie in de toekomst zouden kunnen worden geïntegreerd, waarbij gebruik gemaakt wordt van ervaringen van patiënten, medische kennis en praktijkvoorbeelden.

Hoofdstuk 5

Hoofdstuk 5 geeft een beschrijving van vijf onderzoeken onder verschillende gebruikersgroepen die gebruik maken van het beschreven ESPE e-learning portal. Er werden twee casus ontwikkeld met drie specifieke feedback vragen, waarbij de student persoonlijke feedback ontvangt op zijn/haar input. Het antwoord van de student wordt anoniem aangeboden aan een medisch expert, die een persoonlijke reactie geeft op het ingestuurde antwoord.

Er zijn in totaal 8 casus en 4 hoofdstukken bestudeerd en beoordeeld van de ESPE e-learning portal. Er waren in totaal 71 beoordelaars: 18 medisch specialisten, 21 kinderartsen in opleiding tot kinderendocrinoloog, 13 kinderartsen in opleiding, 10 medisch specialisten en 9 regionale algemeen kinderartsen. Deelnemers waren enthousiast over de inhoud en over het aantal en de kwaliteit van de reflectieve vragen die waren opgenomen in het bestudeerde materiaal.

Eén van de vijf onderzoeken was specifiek gericht op het verstrekken van persoonlijke feedback op communicatie gerelateerde vraagstukken. Medisch specialisten in opleiding waardeerden de persoonlijke feedback, hetgeen hen nieuwe inzichten verstrekte.

Verder werd het gewaardeerd dat feedback niet alleen medisch inhoudelijk was, maar ook gericht op communicatieve vaardigheden. De mogelijkheid om anoniem deel te nemen werd gewaardeerd, zowel door artsen in opleiding als door de experts die de feedback verstrekten.

Hoofdstuk 6

Hoofdstuk 6 bekijkt de rol van e-learning in het trainen van de competenties 'Medisch expert' en 'Communicator' binnen het aandachtsgebied DSD van de kinderendocrinologie. De studie was wereldwijd opgezet, waarbij 32 medisch specialisten (experts) en 68 kinderartsen in opleiding tot kinderendocrinoloog (fellows) deelnamen. Fellows werd gevraagd om 2 casus te bestuderen binnen de ESPE e-learning portal en daarbij per casus 3 vragen te beantwoorden. De vragen betroffen communicatieve dilemma s, waarbij bijvoorbeeld werd gevraagd een zeldzame diagnose uit te leggen aan patiënt en ouders. De antwoorden van de fellows werden verzonden naar 3 verschillende experts voor feedback. Hierbij werden experts geselecteerd van 3 verschillende continenten, waarvan één expert afkomstig was uit dezelfde regio, maar uit een ander opleidingscentrum. Experts konden gebruik maken van een door het studieteam opgesteld antwoordmodel, maar waren vrij in hun feedback .

De Fellows waardeerden de persoonlijke feedback door meerdere experts, hetgeen overeen komt met eerdere bevindingen. De meervoudige feedback werd als nuttig ervaren, waarbij werd ervaren dat verschillende gezichtspunten mogelijk zijn. Bovendien gaf de feedback inzicht in aspecten zoals invloed van religie en in ondersteuning voor patiënten bij het delen van informatie over de aandoening met familie en vrienden. Fellows vonden het ook heel prettig om een bevestiging te krijgen dat ze juist gehandeld hadden. Reacties van de fellows geven aan dat de opgedane ervaringen zullen gebruiken in hun toekomstige carrière.

Hoofdstuk 7

Hoofdstuk 7 bouwt voort op de reacties van de fellows in hoofdstuk 6, waarbij hen werd gevraagd een antwoord te formuleren op communicatieve vraagstukken binnen twee casus op het gebied van DSD. Ten eerste werden deze antwoorden geanalyseerd op kern elementen, zoals uitleg van de diagnose, behandelmogelijkheden en begeleiding. De kern elementen waren bepaald door het studie team, dat bestond uit medisch specialisten, communicatie deskundigen, patiënt vertegenwoordigers en een onderwijskundige. Bij de analyse werden de antwoorden van de fellows beoordeeld op kwaliteit, volledigheid en empathie. Er was een grote mate van variatie in de antwoorden, zowel in de volledigheid, als in de kwaliteit en in de mate van empathie. De kern elementen 'uitleg van de diagnose' en 'behandelmogelijkheden' werden het vaakst genoemd. Het was opvallend dat de beschikbare tijd, de uitleg van de rol van het DSD team en geruststelling zelden werden benoemd.

Op basis van de analyse van de antwoorden werd een rubric opgesteld. Een rubric is een instrument om verschillende soorten prestaties te evalueren of te beoordelen. Voor beide casus werd een rubric uitgewerkt, waarbij de kern elementen werden beschreven op vijf verschillende niveaus met daarbij voorbeeldteksten. Deze rubric werd vervolgens geëvalueerd door 22 kinderendocrinologen wereldwijd. Op basis van de rubric hebben zij de antwoorden van 10 fellows beoordeeld, waarbij de antwoorden waren opgedeeld in 114 passages gerelateerd aan de kern elementen zoals beschreven in de rubric. De beoordeling door de medisch experts liep zeer uiteen. Uiteindelijk is de rubric aangepast naar drie verschillende niveaus met empathie als een extra kern element.

Concluderend lieten de antwoorden van de fellow grote variatie in communicatieve vaardigheden zien. Bepaalde aspecten zoals lange-termijn effecten en geruststelling warden nauwelijks benoemd. Dit kan mogelijk een gevolg zijn van de beperkte ervaring met deze ziektebeelden. E-learning gericht op training communicatieve vaardigheden voor patiënten met DSD kan hierbij zeer bijdragend zijn.

De studie toonde ook grote variatie in de beoordeling van antwoorden van de fellows door de deelnemende medisch specialisten. Hier zijn meerdere verklaringen mogelijk. De experts waren niet getraind in het gebruik van de rubric. Het is zeer goed mogelijk dat de medische specialisten van mening verschillen wat goede communicatie is. Derde mogelijke verklaring is dat de communicatieve vaardigheden van de medische specialisten zelf ook sterk varieert.

Hoofdstuk 8

In Hoofdstuk 8 worden alle resultaten beschouwd in de discussie. Hierbij worden eerst de bevindingen ten aanzien van het trainen van communicatieve vaardigheden in chronische ziekte tijdens de medische vervolgopleiding besproken. Vervolgens worden de bevindingen ten aanzien van het trainen van communicatieve vaardigheden bij zeldzame ziekten worden besproken. Alle bevindingen worden gerelateerd aan de bewezen effectieve elementen bij het trainen van communicatieve vaardigheden in de medische vervolgopleidingen. Achtereenvolgens zijn dit 1) bewustzijn en reflectie, 2) deliberate practice en 3) een veilige en ondersteunende omgeving. Aanbevelingen voor de praktijk worden geformuleerd en ten slotte wordt gereflecteerd op alle onderzoeken en aanbevelingen voor medische vervolgopleidingen en voor onderzoek.

CHAPTER 11

PhD portfolio
Dankwoord
Curriculum Vitae
Publications

PhD Portfolio

	Year	Workload (ECTS)
General academic and research skills		
Workshop Systematic Literature Retrieval in Pubmed	2017	0,4
Training didactics (Tweedaagse training basisdidactiek voor docenten (TtT I)	2018	0,4
Training didactics (Eendaagse training begeleiden voor AIOS (TtT III)	2019	0,2
Training didactics (Competentieontwikkeling AIOS door opleidingsgroep)	2019	0,2
Training communication (Conversation in Motion Shared Decision Making)	2019	0,1
Training communication (Conversation in Motion Empathie en vertrouwen)	2019	0,1
Female Talent Class (Erasmus MC, 7 workshops)	2021	0,5
National conferences		
Nederlandse Vereniging voor Reumatologie (NVR) Januarivergadering, Utrecht (oral presentation)	2017	0,5
Nederlandse Vereniging voor Reumatologie (NVR) Najaarsdagen, Papendal (oral presentation)	2017	1,0
Nederlandse Vereniging voor Reumatologie (NVR) Januarivergadering, Utrecht (oral presentation)	2018	0,5
Nederlandse Vereniging voor Reumatologie (NVR) Najaarsdagen, Papendal (2 oral presentations)	2018	1,0
International conferences		
55th Annual Meeting European Society for Paediatric Endocrinology, Paris, France (oral presentation)	2016	1,0
6th International Conference on Medical & Nursing Education, Vienna, Austria	2017	1,0
European League Against Rheumatism (EULAR) annual meeting, Amsterdam (poster presentation)	2018	1,0
European League Against Rheumatism (EULAR) annual meeting, online (poster presentation in tour)	2020	1,0
Workgroups		
Department Rheumatology research meetings (attendance)	2017-2021	4,0
Participation national workgroup improvement Electronic Patient Record	2017-2021	1,0
Participation eRA workgroup shared decision en patient empowerment	2019	0,5
Participation national advisory board eHealth 2.0	2019	0,5
Implementation project shared decision making (multicenter involvement and patient organisation)	2019-2021	4,0
Teaching tasks		
Supervising students Technical University Delft - Reuma App for patients	2020	1,0
Supervising student Psychology - internship chronic disease	2020	0,5
Resident communication training rheumatology	2020-2021	1,0
Communication training Shared Decision Making	2020-2021	0,5
Training Shared Decision Making in pregnancy	2021	0,5

Other		
Development Reuma App for improvement of selfmanagement of patients	2018-2020	4,0
Nederlandse Vereniging voor Reumatologie (NVR), member accreditation comittee	2019-2021	4,0
Total		30,4

Dankwoord

Dankwoord. Dit is het laatste onderdeel van dit proefschrift. Niet alleen een afronding van een boekje, maar ook van een bijzondere en intensieve periode. Een periode, waarbij ik zeer veel heb geleerd. In de eerste plaats heb ik veel geleerd over mijzelf en over het omgaan met tegenslagen. Bij het begin van deze onderneming heb ik mij nooit gerealiseerd dat dit zo intensief kon zijn. Maar, 'if it doesn't kill you, it will make you stronger', zoals een wijs man vaak zegt. Ten tweede heb ik veel geleerd over onderzoek. Al doende leert men. Wat is een goede onderzoeksvraag, wat wil je weten en wat zijn goede methodes om hier een antwoord op te vinden. Ten slotte wil ik benadrukken dat ik me met veel plezier en enthousiasme heb bezig gehouden met dit onderwerp en van plan ben hier voorlopig nog niet mee op te houden.

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Curriculum Vitae

Laura Kranenburg werd geboren op 12 mei 1979 in Rotterdam. Na het cum laude behalen van haar Gymnasium diploma aan de christelijke scholengemeenschap Johannes Calvijn in Rotterdam vervolgde zij haar opleiding Industrieel Ontwerpen aan de TU Delft in 1997. Zij studeerde in 2004 af in de richting ‘Medisign’ met als afstudeeronderwerp “*Ontwerp van een ergonomische bediening van electrochirurgie tijdens minimaal invasieve operaties”*. Vervolgens werkte zij in verschillende functies, onder andere als productontwerper voor de gezondheidszorg en als adviseur voor onderwijsvernieuwing. In 2007 startte zij de opleiding geneeskunde welke in 2012 werd afgerond. Dit werd vervolgd door een specialisatie in de reumatologie. Vanaf 2018 is zij werkzaam als reumatoloog verbonden aan het Erasmus MC en is tevens werkzaam als reumatoloog bij het IJsselland ziekenhuis. Haar aandachtsgebieden bestaan uit het verbeteren van gezamenlijke besluitvorming, inzet van eHealth en begeleiding van zwangerschap bij reumatische aandoeningen.

Publications

- EULAR 2021 POS0063-PARE implementing shared decision making to advance patient centred rheumatoid arthritis care: a role for a patient organization. P. Pennings Ms; Helene R Voogdt – Pruis; B. Maat et al. Annals of the Rheumatic Diseases 80(Suppl 1):238.1-238
- Modern treatment approach results in low disease activity in 90% of pregnant rheumatoid arthritis patients: the PreCARA study. Hieronymus TW Smeele; Esther RöderEsther Röder; Hetty M Wintjes; et.al. Annals of the Rheumatic Diseases 80(7):annrheumdis-2020-219547
- EULAR 2020 AB1197 treat-to-target is feasible in rheumatoid arthritis patients during pregnancy, first results of the precara cohort. H. T. Smeele; ; Esther Röder; H. Wintjes et al. Annals of the Rheumatic Diseases 79(Suppl 1):1889.1-1890
- EULAR 2020 Shared decision making amongst rheumatologists in training in the Netherlands: where do we stand?, L. Kranenburg, M. Dankbaar, N. Basoski, W. Van den Broek, J. Hazes
- EULAR 2018 How do we learn to communicate to our patients? an inventory study amongst rheumatologists in training in the Netherlands how they train communication skills, L. Kranenburg – Van Koppen, M. Hazes, W.V.D. Broek, M. Dankbaar
- Global Application of the Assessment of Communication Skills of Paediatric Endocrinology Fellows in the Management of Differences in Sex Development Using the ESPE E-Learning. Laura J.C. Kranenburg, Stenvert L.S. Drop, et. al. Org Portal. Horm Res Paediatr. 2017;88(2):127-139.
- Global Application of Assessment of Competencies of Paediatric Endocrinology Fellows in the Management of Differences of Sex Development (DSD) Using the ESPE e-learning.org Portal. Kranenburg L, Stenvert L.S. Drop, et al. Med.Sci.Educ. (2016) 26: 679.
- Global Application of Disorders of Sex Development-Related Electronic Resources: e-Learning, e-Consultation and e-Information Sharing. Miriam Muscarella, Laura Kranenburg-van Koppen, Kalinka Grijpink-van den Biggelaar, Stenvert L.S. Drop. Endocr Dev. 2014;27:268-83.
- The espe e-learning webportal: a global tool for instruction and formative assessment of pediatric endocrinology. Stenvert L.S. Drop, Laura Kranenburg, Sam Reerds, Kalinka Grijpink, Violeta Iotova. Scripta Scientifica Medica, vol. 46, No 3, 2014, pp. 11-18
- An interactive E-Learning Portal in Pediatric Endocrinology: Practical Experience. Laura J. C. Kranenburg, Kalinka Grijpink-van den Biggelaar, Stenvert L. S. Drop. Pharmacy 2013, 1(2), 160-171.
- Ergonomics in thoracoscopic surgery: results of a survey among thoracic surgeons. Katrin Welcker, Emeka B. Kesieme, Eveline Internullo, Laura J.C. Kranenburg van Koppen. Interactive CardioVascular and Thoracic Surgery 15 (2012) 197- 200

- Diagnostic Yield Improves With Collection of 2 Samples in Fecal Immunochemical Test Screening Without Affecting Attendance. van Roon A, Wilschut J, Hol L, v Ballegooijen M, Reijerink J, 't Mannetje H, Kranenburg L et al. Clinical gastroenterology and hepatology 2011;9:333–339
- Ergonomic problems encountered during Video- Assisted Thoracic Surgery. Kranenburg L, Gossot D. Minimally Invasive Therapy & Allied Technologies 2004;13(3): 147-155