and SSC. Average utilities for each health state were predicted from the regression coefficients. Utilities were also estimated using individual health state dummy variables in a sensitivity analysis. **RESULTS:** Utilities for health states without BP or SSC were 0.76 for mild, 0.69 for moderate, 0.64 for marked, and 0.60 for severe. The utilities for states with bone symptoms were lower: 0.67 for mild+BP, 0. 68 for mild+SSC, 0.61 for moderate+SSC, 0.56 for marked+SSC, and 0.52 severe+SSC. Sensitivity analysis utilities were similar but less precise and sometimes less clinically plausible. **CONCLUSIONS:** Compared with the norm for the EQ-5D utility index in the UK (0.86), these utility estimates reflect the substantial quality of life impact of GD1, especially with skeletal complications. These utilities can be used in future economic evaluations of GD1 therapies to calculate quality-adjusted life years.

PSY81
SOCIAL UTILITY VALUES FOR PEMPHIGUS VULGARIS AND FOLIACEUS: A COMPOSITE TIME TRADE-OFF STUDY

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OBJECTIVES: In the past few years, a number of biological drugs to treat pemphigus have successfully passed through phase 2 and 3; however, none of the clinical trials in pemphigus has used preference-based outcome measures to generate utilities for economic evaluations, so far. In attempt to fill this gap, the objective of this study is to measure utility weights for hypothetical pemphigus vulgaris (PV) and foliaceus (PF) health states in a general population sample. **METHODS:** Three health state profiles were developed: an uncontrolled PV, an uncontrolled PF and a treated pemphigus state. Health state vignettes combined a description of living with pemphigus including skin and mucosal symptoms, possible food avoidance, bathing/clothing issues, and effects on work and social life. Utilities were obtained from a convenience sample of 106 adults using visual analogue scale (VAS, 0-10 scale) and time trade-off (TTO). Lead-time TTO was applied for health states considered worse than dead. Utilities in this study were ranging between -1 (lower anchor of worse than dead) and 1 (perfect health) **RESULTS:** Mean VAS scores for PV, PF and treated pemphigus were 2.46±1.54, 3.77±1.69 and 6.35±1.60, respectively. The estimated utility weights attached for the health states were as follows: PV 0.34±0.38, PF 0.51±0.31 and treated pemphigus 0.73±0.31. Overall 13% and 5% judged PV and PF as being worse than dead. For both VAS and TTO values, significant difference revealed between each of the three health states (p<0.001). **CONCLUSIONS:** This study provides the first utility weights in pemphigus. Disutility associated with uncontrolled pemphigus was found worse than in uncontrolled psoriasis or atopic dermatitis. Successful treatment of pemphigus may yield significant utility gain (0.22-0.39). Utilities elicited in this study may be used in cost-effectiveness analysis of new pemphigus treatments.

PSY82
"FIRST AND FOREMOST BATTLE THE VIRUS" - PATIENT PREFERENCES IN ANTIVIRAL THERAPY FOR HEPATITIS C

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OBJECTIVES: Treatment options for patients with hepatitis C have made tremendous progress in the last 5 years. Innovative antiviral-therapies promise a huge reduction of burden of disease and side-effects accompanied by an increase of patient's benefit. It is unclear, however, what properties generate the highest benefit for patients. **METHODS:** A systematic literature search was conducted to identify patient-relevant outcomes . Within N=14 semi-structured interviews the decision model was generated. Eight patient relevant characteristics were identified and described by three or six levels. For the discrete choice experiment, an experimental design (3*3+5*6) was generated using Ngene-Software. The design consisted 72 choices, which were divided into 6 blocks. The survey was conducted in august 2014 with computer-assisted personal interviews. **RESULTS:** N=561 hepatitis C-patients (58,1% male) within different treatment states participated. The preference analysis (random parameter logit estimation (with 95%CI)) showed a predominance for the attribute "sustained virological response 6 month after end of treatment"(coef.:1,108). A split second rank is occupied by the possible side effects "anemia"(coef.:0.396) and "rash"(coef.:0.372). The attributes that describe the administration of the treatment "number of interferon injections" (coef.:0.224) and "duration of treatment"(coef.:0.183) share the third place. The attributes "fatigue/exhaustion"(coef.:0.088) and "nausea/diarrhea"(coef.:0.067) follow. Within the treatment decision of hepatitis C-patients the attribute "headache"(coef.:0.036) was not taken into account, as reflected by the lack of significance. **CONCLUSIONS:** In this study, the essential decision criteria for an optimal antiviral therapy of hepatitis C from the Hep C patients' view were obtained by using a DCE. Thereby SVR is ranked highest against all patient relevant side effects and mode of administration characteristics. In the context of this study it could be demonstrated that with the help of the DCE patient-relevant properties can be weighted in terms of possible treatment options. The DCE stands out by a high degree of realism and good handling for the patient.

PSY83
TEN YEARS OF THE INTERNATIONAL POMPE SURVEY: PATIENT REPORTED OUTCOMES AS A RELIABLE TOOL FOR STUDYING TREATED AND UNTREATED CHILDREN AND ADULTS WITH NON-CLASSIC POMPE DISEASE

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OBJECTIVES: Pompe disease is a rare, progressive lysosomal storage disorder for which enzyme therapy (ERT) became available in 2006. Four years earlier, the IPA/Erasmus MC survey, an international longitudinal prospective survey, was established to collect information on the natural course of the disease and its burden on patients. **METHODS:** The survey is a collaboration between Erasmus MC University Medical Center and the International Pompe Association (IPA) and comprises an annual questionnaire that was specifically designed to assess the symptoms and problems of the disease. **RESULTS:** We review our results of over 10 years of follow-up, and discuss the survey's contribution to the field. Tracking 408 Pompe patients between 2002 and 2013, the cumulative data reveals the broad range of clinical manifestations that interfere with patients' lives. The survey allowed us to quantify the rate of disease progression and the positive effects of ERT on patients' quality of life, fatigue, and participation in daily life. Furthermore, it showed for the first time that survival is reduced in adult Pompe disease and improved by ERT. **CONCLUSIONS:** Our results show that a patient survey can serve as a valuable and reliable tool for obtaining quantifiable information on the natural course of a rare disease and on the effects of therapy in a large cohort over a very long time. Most importantly, by working with patient reported outcomes, the survey provides the data that is truly relevant to the patient and complementary to clinical datasets.

PSY84
THE IMPACT OF ORTHOPAEDIC SURGERY ON THE QUALITY OF LIFE OF HAEMOPHILIA PATIENTS WITH OR WITHOUT INHIBITORS: A SYSTEMATIC REVIEW OF THE LITERATURE

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OBJECTIVES: To conduct a systematic review of the literature and assess the impact of orthopaedic surgery on the health-related quality of life (HRQoL) of haemophilia patients with or without inhibitors. **METHODS:** Searches were conducted in the MEDLINE, EMBASE, EconLit, Cochrane library and the Center for Reviews and Dissemination databases from 2000 onwards. A broad search including both Medical Subject Heading (MeSH) terms and key words related to haemophilia and HRQoL was conducted. We included only the publications reporting pre- and post-surgery HRQoL data and all orthopedic interventions were considered. Congress abstracts from 2010 were also reviewed. Abstracts and full text articles were screened by two reviewers and conflicts arbitrated. **RESULTS:** A total of 1,471 articles were retrieved. Following the screening process, 8 full-text articles reporting pre- and post-surgery HRQoL data for patients who received ankle arthrodesis, total knee replacement, ankle replacement or hip arthroplasty were included. Seven studies used patient-reported measures and only one study estimated the EQ-5D using indirect matching techniques. Of the seven studies, four used the visual analogue scale (VAS). Three concluded that post-operative pain was significantly lower and only one study reported that although pain decreased post-surgery, moderate pain remained among patients who underwent multiple joint procedures and received ankle arthrodesis. Of the four studies that reported SF-36/SF-12 scores, significant gains in physical health scores were observed in three and higher mental health scores were reported in one study. Disease-specific measures, Western Ontario and McMaster University (WOMAC) and Oxford Knee Score, were assessed by two studies and improvements in pain and physical functioning were observed on both scales. Evidence from SF-12 summary scores suggests that the improvements in physical functioning are similar for patients with and without inhibitors. **CONCLUSIONS:** Orthopaedic surgery improves the HRQoL of haemophilia patients with severe arthropathy.

PSY85
HEALTH STATES WORSE THAN DEAD IN AN ELDERLY POPULATION – PREVALENCE AND THE SIGNIFICANCE OF CHRONIC PAIN

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OBJECTIVES: When applying accepted health economic methods, it is not uncommon to find valuations of patients' health states that are worse than dead. Hypothetical valuations (the UK value set) of states according to the EQ-5D resulted in 82 of 243 possible states being valued as worse than dead. The presence of health states valued worse than dead risks giving rise to counter-intuitive consequences when incorporated in health economic analyses. The purpose of this study was therefor to investigate the presence of such states in an exposed population, namely people 65 years and older. **METHODS:** Based on a Swedish epidemiological population survey study directed to 10 000 persons 65 years and older the occurrence of states worse than dead were investigated and related to prevalence and intensity of chronic pain. **RESULTS:** Health states worse than dead were found in 1.8 % of the 6618 responders to the questionnaire. Among persons with severe chronic pain the prevalence was 16.7 % and among persons with moderate and no or mild chronic pain the prevalence of states worse than dead were 2.7 % and 0.8 % respectively. Prevalence of these states clearly increased with age and female gender. Of women 85 years and older with severe chronic pain 38.7 % were in states worse than dead. **CONCLUSIONS:** Health states worse than dead are not rare in an elderly population and covariate with chronic pain, age and female gender. The prevalence of states worse than dead are highly dependent on the value set applied, and will have consequences for assessments of treatments for elderly with chronic pain. Based on the results of this study it should be investigated whether and to what extent health states worse than dead exist when using experience based valuation methods, which is the preferred approach in some settings/countries.

PSY86
CLUSTERING HEALTH STATUS AMONG ADULTS WITH HEMOPHILIA USING SF-12 HEALTH SURVEY

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