and SSC. Average utilities for each health state were predicted from the regression coefficients and 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, 0.68 for severe. The utilities for states with bone symptoms were lower: 0.67 for mild+BP, 0.68 for moderate+BP, 0.69 for severe+BP, 0.74 for mild+SSC, 0.76 for 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, our survey estimates reflect the substantial burden of GD1, especially with skeletal complications. These utilities can be used in future economic evaluations of GD1 therapies to calculate quality-adjusted life years.

PSY1

SOCIAL UTILITY VALUES FOR PEMPHIGUS VULGARIS AND FOLIEACEA: 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 been developed. However, disutility associated with the presence of these states clearly increased with age and female gender. Of these states worse than dead were found more frequent in the elderly. We review our results of over 10 years of follow-up, and discuss the survey’s contribution to the field. Tracking 480 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 hospitalization, progression of disease, and 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 incidence rate estimate is consistent with a p-values of less than 0.05. The results are valuable 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.

PSY2

"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 major reduction of disease risk and side-effects. A side-effect that is accompanied by a negative impact on the patient’s benefit is the nausea. It is unclear, however, what properties generate the highest benefit for patients. METHODS: A systematic literature search was conducted to identify patient preferences and priorities. Within N=14 semi-structured interviews a decision-aid model was generated. Eight patient relevant characteristics were identified and described by three or six levels. For the discrete choice experiment, an experimentally determined choice design with optimal D-efficiency and applying the method of truncated normal distributions was 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). The results 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.

RESULTS: Orthopaedic surgery improves the HRQoL of haemophilia patients with severe arthropathy.

PSY3

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 62 of 243 possible states being valued as worse than dead. The prevalence of health states valued worse than dead risks giving rise to counter-intuitive conclusions when incorporated in health economic analyses. The purpose of this study was therefore 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 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 cooccur 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.

PSY4

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 MEDLINE, EMBASE, EconLit, Cochrane library and the Center for Reviews and Dissemination databases from 2000 onwards. A broad search including both Medical Subject Headings (MeSH) and free text words related to haemophilia and orthopaedic HRQoL was performed. Studies were included if they 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). The results 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.

CONCLUSIONS: 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.