increased use of prophylaxis. Few studies have analyzed the total costs of treatment, hence we undertook this study. METHODS: To determine total costs and trends of treating children with severe hemophilia-A from 1978 to 1998, at the Hospital for Sick Children in Toronto, one patient chart reviewer identified resource consumption of all patients (n = 17). For each patient, costs for factor concentrate, clinic visits, physicians and healthcare professionals (physiotherapists/social workers), laboratory and other tests (x-ray, MRI, ultrasound) and hospitalizations were determined. Costs in Canadian dollars were taken from standard lists and discounted at 3%. RESULTS: Total average cost (range) was $69,322 ($14,471–$108,294)/year/patient of which the largest part, $65,184 ($8,250–$107,104), 94% (57%–99%) was accounted for by Factor-VIII. Hospitalizations accounted for $2,396 ($0–$57,063)/patient/year including drugs, nursing care and stay. Clinic visits and physician visits were $1290 ($122–$4143) and $177 ($0–$308), respectively. Healthcare professionals averaged $89 ($0–$252) and lab tests and other tests cost $156 ($26–$226) and $31 ($4–$70)/patient/year, respectively. The average number of bleeds was 12.9 (2.0–22.0)/patient/year. Since 1978, the average number of bleeds decreased by 0.71 (r^2 = 0.56)/patient/year. The average number of hospitalizations was 0.21 (0–4)/patient/year, in which patients stayed on average 10.7 (1–135) days. Since 1984, the number of hospitalizations has decreased by 0.5 hospitalizations/patient/year (r^2 = 0.74). Concurrently, the average costs of the treatment of severe hemophiliacs have increased by approximately $3740 (r^2 = 0.62)/patient/year. Clotting factor concentrate cost per patient increased by $4215 (r^2 = 0.66)/year, of which prophylaxis accounted for $1429 (r^2 = 0.60)/year, while on demand Factor-VIII costs decreased by $497 (r^2 = 0.16)/year. CONCLUSIONS: The annual cost of hemophilia care, of $69,322 per patient, is substantial. The number of bleeds and hospitalizations is decreasing while there was a clear trend in increasing costs of treating severe hemophiliacs, primarily associated with increasing use of prophylactic treatment.

HEMOPHILIA ECONOMIC MODEL OF OUTCOMES: CASE STUDY
Evans C1, Roberts HR2, Sagrollikar A3, Gomperts E3, Poulios N4
1Mapi Values, Boston, MA, USA; 2University of North Carolina, Chapel Hill, NC, USA; 3Baxter Healthcare Corporation, Westlake Village, CA, USA

OBJECTIVES: In Asia, individuals with hemophilia are often treated with cryoprecipitate as it is considered a less expensive option. However, the risk of acquiring blood-borne infections is higher with cryoprecipitate than with factor concentrates. Any acquisition cost savings that occur with the use of cryoprecipitate may be offset by greater total healthcare costs to treat transmissible viruses. METHODS: A literature review was conducted to obtain estimates of the prevalence of HIV, HBV and HCV in the blood supply and costs of treating the subsequent diseases. Data from international health organizations, local experts in the treatment of hemophilia, and the local Red Cross Society was utilized. This data forms the basis of an economic and outcomes model for hemophilia treatment in this Asian country. RESULTS: Based on the data identified, the risk of exposure to the blood-borne infections in hemophilia treated with cryoprecipitate is substantial: 3%, 6% and 12% at 5, 10 and 20 years respectively for HIV. Risk for developing hepatitis infection was higher. The discounted cost of treating the three infectious diseases is substantial. For patients treated with cryoprecipitate total treatment costs were $26,575 and $64,291.06 at 10 and 20 years. For patients treated with factor concentrate equivalent costs were $32,200 and $64,401. CONCLUSIONS: Over time, the acquisition cost savings associated with cryoprecipitate use, compared to factor concentrate, disappears as the costs for treating transmissible viruses in infected patients increases.

HEMATOLOGIC/PITUITARY DISORDERS—Quality of Life/Preference Based Outcomes

IMPROVEMENT IN QUALITY OF LIFE AND HEALTHCARE UTILISATION DURING GROWTH HORMONE REPLACEMENT THERAPY IN HYPOPITUITARY ADULTS IN THE NETHERLANDS
den Hartog M1, van Kuijck MA1, Koppeschaar HPP1, Mattsson AF3, Koltowska-Häggström M1
1Pharmacia BV, Woerden, Netherlands; 2Utrecht Medical Centre, Utrecht, Netherlands; 3Pharmacia AB, Stockholm, Sweden

OBJECTIVES: To investigate whether long-term growth hormone (GH) replacement therapy in GH deficient (GHD) adults results in improvements in Quality of Life (QoL), patient-reported outcomes and health care utilization (HCU) in the Netherlands. METHODS: The analysis was based on 74 patients (35 men, 39 women) and all patients were included in KIMS (Pharmacia International Metabolic Survey)—the largest pharmacoeconometric survey of GHD adults on GH therapy. Data were available for all patients for the first year of treatment, and 2-year follow-up data were available for 38 patients. QoL was assessed using the Nottingham Health Profile (NHP) and disease-specific AGHDA questionnaire. Patient reported outcomes and data on HCU were obtained with the Patient Life Situation Form (PLSF). Statistical analyses were performed with repeated measurements technique. RESULTS: Both QoL questionnaires showed a significant improvement after 1 and 2 years of GH therapy (from 20.1 ± 2.6 to 10.1 ± 2.5 for NHP; from 9.3 ± 0.82 to 7.2 ± 1.11 for AGHDA). Data collected
with the PLSF showed a subjective improvement in personal well-being for 65% of the patients after 1 and 2 years of GH treatment. Visits to the doctor in the previous year (from 9.1 ± 1.4 to 3.4 ± 1), days in hospital (from 7.9 ± 2.6 to 1.6 ± 1.5) and days of sick leave (from 26.4 ± 9.8 to 2.2 ± 1.1) significantly decreased during GH therapy. Leisure-time physical activity significantly improved during therapy, whereas satisfaction with physical activity improved only in females. Another gender difference relates to the need for assistance with daily activities, which remained low and constant in males, but seemed to worsen in females. CONCLUSION: Data obtained so far confirm that GH replacement therapy results in a significant long-term improvement of both QoL and general well being and reduction of HCU in the Netherlands. Moreover, after two years of GH therapy both QoL and HCU levels become comparable to the general Dutch population.

DEVELOPMENT OF THE GROWTH HORMONE INJECTION QUESTIONNAIRE (GHIQ) FOR ADOLESCENTS
Cramer JA1, Simeoni MC2, Auquier P3, Brasseur P4, Beresniak A5
1Yale University School of Medicine, West Haven, CT, USA; 2University of Medicine, Marseille, France; 3Serono International SA, Geneva, Switzerland

OBJECTIVES: We report the development of an instrument to assess feelings about self-injection of growth hormone (GH) in adolescents with growth disorders.

METHODS: Adolescents (age 11–19 years) in 2 cohorts (UK N = 74 and USA N = 98) completed a 10-item GH questionnaire, as well as health-related quality of life (HRQOL), functional status scales, self-control, and self-esteem scales to assess external validity. The GH and HRQOL questionnaires were repeated in 2–4 weeks.

RESULTS: The GH questionnaire contains 10 items in 2 subscales (feelings, injection issues) and a total summary score, with higher scores indicating better acceptance of GH. Internal consistency reliabilities were α = 0.77 Summary Score, α = 0.68 Feelings, and α = 0.78 Injection Issues, with overall test-retest reliability 0.72, and no floor or ceiling effects. Significant summary score correlations with external validity scales were 0.42 HRQOL, 0.34 functional status, 0.25 self-control. Mean summary scores did not differ significantly by cohort, age, gender, or diagnosis (range 4.0–4.4 points). The Injection Issues component was significantly associated with all VSP-AM subscales and total score (all p < 0.01), whereas the Feelings component was significantly correlated with self-esteem, well-being, physical health and the total score (all p < 0.01) as well as with teachers (p < 0.05). CONCLUSIONS: The GH Injection Questionnaire is both a reliable and valid measure of attitudes for adolescents with growth disorders. It measures different constructs than other instruments. Adolescents who were more comfortable with GH injections had higher HRQOL, functional capacity, and self-control.