Indirect Costs Among Persons With Hemophilia B: Hemophilia Utilization Group Study Part Vb (HUGS Vb)

Mimi Lou[1], Jiat-Ling Poon[1], Megan Ullman[2], Marion Koerper[3], Brenda Riske[4], Judith Baker[5], Roshni Kulkarni[6], Erin Cockrell[7], Barbara Konkle[8], Kathleen Johnson[1]

[1]University of Southern California, Los Angeles, CA, USA, [2]University of Texas Health Science Center at Houston, Houston, TX, USA, [3]University of California, San Francisco, CA, USA, [4]University of Colorado Denver, Denver, CO, USA, [5]University of California, Los Angeles, CA, USA, [6]Michigan State University, East Lansing, MI, USA, [7]Akron Children's Hospital, Akron, OH, USA, [8]Puget Sound Blood Center, Seattle, WA, USA

INTRODUCTION

- The Hemophilia Utilization Group Study Part Vb (HUGS Vb) is a two-year, prospective, multicenter study designed to evaluate the cost of care, healthcare utilization and burden of illness among a nationally representative cohort of persons with hemophilia B in the United States (US).
- Persons with hemophilia B were recruited from ten US federally supported Hemophilia Treatment Centers (HTCs) that provide care to patients in eleven states: California, Colorado, Indiana, Massachusetts, Michigan, Mississippi, Montana, Ohio, Texas, Washington and Wyoming.
- Indirect costs quantify the productivity loss experienced by persons with hemophilia B and their families associated with (a) impaired ability to work or engage in leisure activities due to morbidity, (b) loss of economic productivity due to death, and (c) caregiver time.

OBJECTIVE

- Examine indirect costs among adults and children with hemophilia B receiving care at ten HTCs in the US.

METHODS

- HUGS Vb launched in June 2009 and recruitment is ongoing. Between June 2009 and May 2012, 130 persons with hemophilia B aged between 2 to 64 years were recruited at participating HTCs.
- Adult participants or parents of children less than 18 years with hemophilia B completed a standardized initial questionnaire about sociodemographics, clinical characteristics and treatment patterns.
- Participants were followed quarterly for two years through mail, internet and telephone surveys. Information was collected on patient-reported outcomes including days missed from work or school, time spent arranging hemophilia care, and caregiver support related to hemophilia received from friends/others.
- A total of 88 participants who completed at least two quarterly follow-up surveys within a one-year time period were included in this analysis.
- Missing school/work days for adults and parents of children, and missing school days for children 5-18 years were defined as excessive if they exceeded the US average missed school/work days for the general population of 8.39 days per year[1] and 11 school days per year[2], in adults and children, respectively.
- Indirect costs were imputed using the human capital approach, which measures productivity loss in terms of lost earnings of the patient or caregiver. This method uses wages as a proxy measure for the output of work time.
- Imputed indirect costs were: (a) lost wages from missed work for those who were employed; (b) lost wages from working part-time or being unemployed due to hemophilia; (c) unpaid caregiver costs.
- Average hourly wage was obtained from the US Department of Labor's Bureau of Labor Statistics "Employer costs per hour worked for employee compensation" published in March 2012. The total employer compensation costs for civilian workers averaged $30.45 per hour worked.
- Part-time work was assumed to be 20 hours/week. Sensitivity analysis was conducted to test the variation of indirect costs by varying part-time work hours from 10 to 30 hours/week.
- All costs are reported in 2012 US dollars.

[1] Yassin A. Cost of lost work and bed days for us workers in private industry--national health interview survey, 2003. J Occup Environ Med. 2007 Jul;49(7):736-47.

[2] National Survey of Children with Special Health Care Needs 2005–2006. http://mchb.hrsa.gov/cshcn05/NF/2healthfs/missed.htm

RESULTS

- The baseline demographics of participants included in the analysis are shown in Table 1. Forty-eight percent were adults and 50% had severe hemophilia. Of the 44 participants with severe hemophilia, 55% of children and 44% of adults used factor prophylactically.
- Thirteen percent of parents and 7% of adults were unemployed or worked part time due to hemophilia (Figure 1).
- Average annual work absenteeism for adults was 3.2 days (range: 0-34), with 2.2 days (range: 0-34) specifically due to hemophilia. For children aged 5-17 years, the average annual school absenteeism was 4.5 school days (range: 0-35), with 2.9 days (range: 0-22) specifically due to hemophilia. Parents missed 2 days (range: 0-21) from work annually, with 1.3 days (range: 0-12) due to their child's hemophilia.
- Twenty-six percent of adults and 20% of parents reported utilizing unpaid caregivers.
- Figure 2 shows the distributions of each category by hemophilia severity. 31% of adults missed work due to hemophilia, and the majority of school children (59%) missed at least one day of school per year due to hemophilia, with 5% missing more than 11 days a year.
- The annual indirect costs of hemophilia for parents of children and adults with hemophilia are shown in Figure 3. The average annual indirect costs for mild to severe hemophilia in parents and adults range from US$47 - US$10,734 and US$132 - US$7,308, respectively. All levels of severity showed a large range of annual indirect costs. Lost wages from missed work and working part time/being unemployed due to hemophilia are the major drivers of total indirect costs of hemophilia.
- Working part-time due to hemophilia occurred only in persons (parents of children) with severe hemophilia. When part-time work hours vary from 10 hours/week to 30 hours/week, the average indirect costs range from US$5,113 - US$8,767 for parents of children with severe hemophilia and from US$5,936 - US$9,455 for adults with severe hemophilia.

Table 1: Baseline Characteristics

Characteristics	Children (N=46)	Adults (N=42)
Age, mean years (median, SD)	9.89 (9.8, 3.9)	40.3 (42.9, 14.7)
Hemophilia Severity (%)		
Mild	8 (17.4)	7 (16.7)
Moderate	12 (26.1)	17 (40.5)
Severe	26 (56.5)	18 (42.8)
Race (%)		
White (Non-Hispanic)	28 (60.9)	33 (78.6)
Black (Non-Hispanic)	4 (8.7)	2 (4.8)
Hispanic	10 (21.7)	4 (9.5)
Other^	4 (8.7)	3 (7.1)
Education (%)†		
≤ 12 years	12 (26.7)	12 (28.6)
> 12 years	33 (73.3)	30 (71.4)
Household income in US$ (%)‡		
≤ 20,000	9 (21.4)	9 (21.4)
Between 20,001 and 40,000	9 (21.4)	6 (14.3)
Between 40,001 and 75,000	6 (14.3)	14 (33.3)
75,001 and above	18 (42.9)	13 (31.0)
Insurance (%)‡		
Both Public and Private	3 (6.5)	6 (14.6)
Public Insurance	17 (37.0)	8 (19.5)
Private Insurance	26 (56.5)	24 (58.6)
No Insurance	0 (0.0)	3 (7.3)
Use factor prophylactically (%)	16 (35.6)	8 (19.1)
Travel time to HTC, mean minutes (median, SD)	95.0 (45.0, 154.7)	87.6 (45.0, 121.9)

† For adults or parents of children age<18;
^ *Other races include: American Indian, Alaskan Native, Asian/Pacific Islander and other;*
‡ *Data do not add up to N=88 because of missing data*

Figure 1: Employment Status

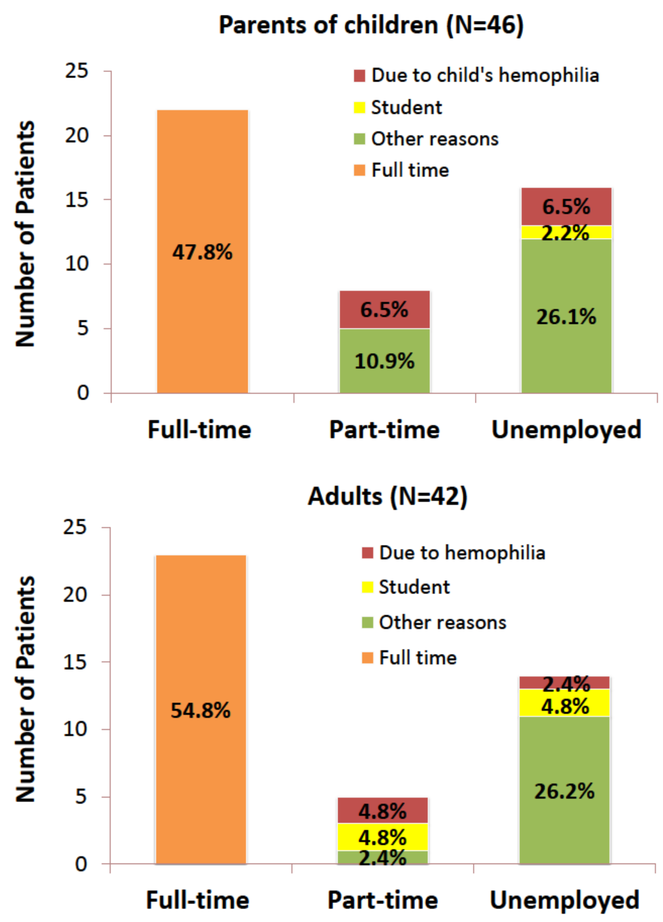


Figure 2: Annual Missed School/Work

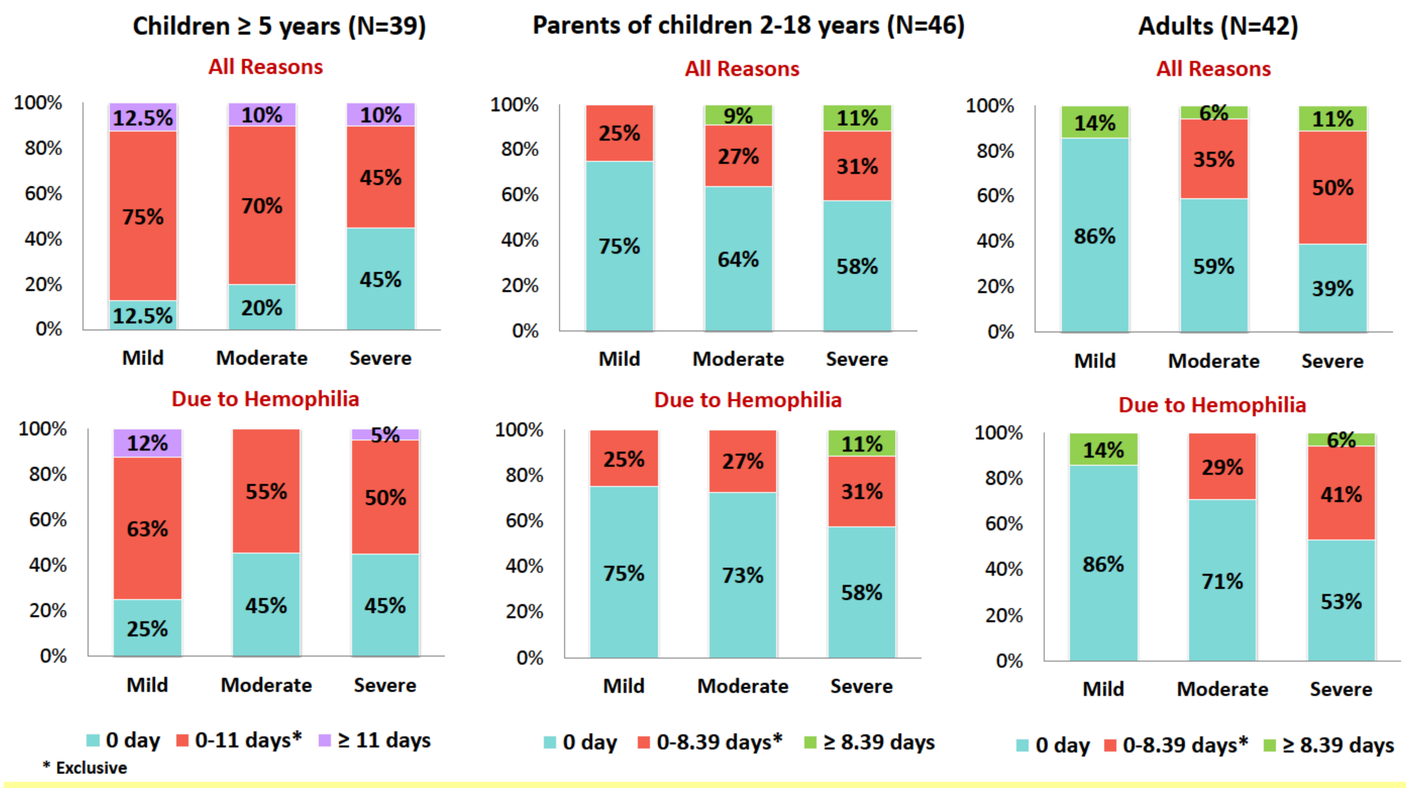
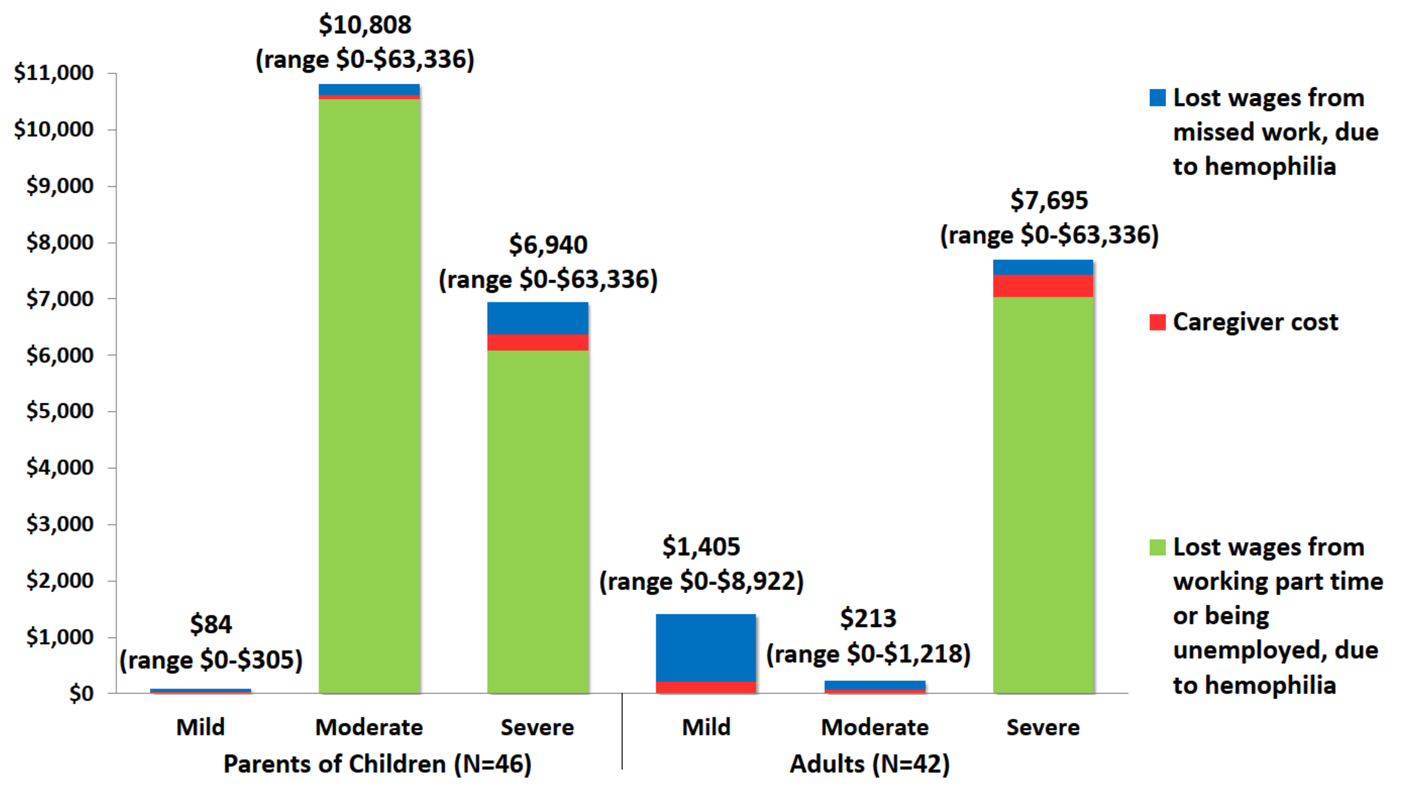


Figure 3: Annual Indirect Costs per Person Due to Hemophilia in US$



DISCUSSION & CONCLUSIONS

- Hemophilia is a costly blood disorder not only because of its high medical expenses, but also for its impact on productivity, such as time lost from work/school due to hemophilia, which incurs indirect costs for patients and their families.
- Accurate measurement of indirect costs will further the development of strategies for treatment approaches to reduce absenteeism and decrease overall cost.
- HUGS Vb is the first study to examine burden of illness in persons with hemophilia B in the US. Further research should compare indirect costs incurred by the HTC-managed hemophilia B population with (a) other chronic conditions, (b) the hemophilia B population receiving care outside the HTC network as well as (c) data collected previously in the hemophilia A population.

ACKNOWLEDGEMENTS

We thank our sponsors: CSL Behring, Baxter Healthcare Corporation, Bayer Foundation, Novo Nordisk for funding the HUGS Va project and Pfizer (formerly Wyeth) for funding the HUGS Va and Vb projects. Additional financial support was obtained from the Federal Hemophilia Treatment Centers/Region IX, Grifols, Red Chip, and CHOC at Home.

The Hemophilia Utilization Group Study Part-Vb (HUGS Vb):
University of Southern California: Kathleen A. Johnson, PharmD, MPH, PhD (Principal Investigator), Mimi Lou, MS, Zheng-Yi Zhou, MS, Jiat Ling Poon, Xiaoli Niu, Jason Doctor, PhD, Joanne Wu, MS, Michael Goode (Programmer);
Children's Hospital Los Angeles, Hemostasis and Thrombosis Center: Robert Miller (Site Principal Investigator), Cathliyn Buranahirun, Jennifer Hanley;
Children's Hospital of Orange County, Hemophilia Treatment Center: Amit Soni, MD (Site Principal Investigator), Heather Huszti, PhD (formerly Site Principal Investigator), James Fabella;
University of Colorado Denver: Brenda Riske, MS, MBA, MPA (Site Principal Investigator), Julie Smith, Deirdre Cooper-Blacketer;
Indiana Hemophilia & Thrombosis Center, Hemophilia Treatment Center: Amy Shapiro, MD (Site Principal Investigator), Natalie Duncan, MPH, Melissa Meyer, Brandy Trawinski, Jayme Harvey;
UMASS Memorial Hospital, New England Hemophilia Center: Ann D. Forsberg, MA, MPH (Site Principal Investigator), Patricia Forand, RN;
University of Texas at Houston, Gulf States Hemophilia and Thrombophilia Center: Megan M. Ullman, MA, MPH (Site Principal Investigator);
Akron Children's Hospital Medical Center, Ohio: Erin Cockrell, DO (Site Principal Investigator), Dawn Ali, Janice Kakish;
Michigan State University, Center for Bleeding and Clotting Disorders: Roshni Kulkarni, MD (Site Principal Investigator), Cheryl Robins, Laura Carlson;
Hemophilia Program, Puget Sound Blood Center, Washington: Barbara A. Konkle, MD (Site Principal Investigator), Sarah Ruuska, Sarah Galdzicka, Steve Grantham;
University of Mississippi Medical Center: Suvankar Majumdar, MD (Site Principal Investigator), Linnea McMillan, RN.
The authors thank the **HUGS Steering Committee:** Randall G. Curtis, MBA, Shelby L. Dietrich, MD and Marion A. Koerper, MD, and **Consultants** Judith R. Baker, MHSA, Kathy Parish, PhD and Femida Gwadry-Sridhar, BSPhm, MS, PhD.

DOI: 10.3252/pso.eu.WFH2012.2012