Published in final edited form as:
Pediatr Blood Cancer. 2010 October ; 55(4): 678–683. doi:10.1002/pbc.22565.

Education and Employment Status of Children and Adults with Thalassemia in North America

Zahra Pakbaz, MD[1], **Marsha Treadwell, PhD**[1], **Hae-Young Kim, PhD**[2], **Felicia Trachtenberg, PhD**[2], **Nagina Parmar, PhD**[3], **Janet L. Kwiatkowski, MD**[4], **Melody J. Cunningham, MD**[5], **Marie Martin, RN**[4], **Nancy Sweeters, PNP**[1], **Ellis J. Neufeld, MD PhD**[6], **Patricia J. Giardina, MD**[7], **Nancy Olivieri, MD**[8], **Robert C. Yamashita, PhD**[9], and **Elliott Vichinsky, MD**[1] **for the Thalassemia Clinical Research Network**[10]

[1]Hematology Oncology, Children's Hospital and Research Center Oakland, 747 52nd street, Oakland, CA 94609

[2]New England Research Institutes, 9 Galen street, Watertown, MA 02472

[3]Haematology Oncology, Hospital for Sick Children, Toronto, ON M5G 1X8, Canada

[4]Pediatrics Hematology, The Children's Hospital of Philadelphia, 3516 Civic Center Blvd., Philadelphia, PA 19104

[5]Hematology, St Jude Children's Research Hospital Children's Hospital Boston, 332 North Lauderdale Street, Mail Stop 355, Memphis, TN 38105

[6]Boston, MA

[7]Pediatrics-Hematology Oncology, Weill Medical College of Cornell University, 1300 York Avenue, NY, NY 10065

[8]University Health Network, Toronto, Canada

[9]Science & Society, California State University, San Marcos, CA

Abstract

Background—Advances in the management of thalassemia have resulted in increased life expectancy and new challenges. We conducted the first survey of education and employment status of people with thalassemia in North America.

Procedures—A total of 633 patients (349 adults and 284 school age children) enrolled in the Thalassemia Clinical Research Network (TCRN) registry in Canada and the US were included in the data analysis. Predictors considered for analysis were age, gender, race/ethnicity, site of treatment (Canada vs. United States), transfusion and chelation status, serum ferritin, and clinical complications.

Results—Seventy percent of adults were employed of which 67 percent reported working full-time. Sixty percent had a college degree and 14% had achieved some post college education. Eighty-two percent of school age children were at expected grade level. In a multivariate analysis for adults, Whites (OR=2.76, 95% CI: 1.50-5.06) were more likely to be employed compared to Asians. Higher education in adults was associated with older age (OR=1.67, 95% CI: 1.29-2.15), female gender (OR=2.08, 95% CI: 1.32-3.23) and absence of lung disease (OR=14.3, 95% CI:

Corresponding author: Zahra Pakbaz, MD **Phone:** 925-286-7167 **FAX:** 510-450-7953 zpakbaz@yahoo.com .
[10]This is publication number eight of the Thalassemia Clinical Research Network (TCRN). A list of TCRN member institutions and staff appears in Appendix 1.

Conflict of Interest: The authors have no conflict of interest to disclose.

2.04-100). Younger children (OR=5.7 for 10 year increments, 95% CI: 2.0 – 16.7) and Canadian patients (OR=5.6, 95% CI: 1.5-20) were more likely to be at the expected education level. Neither transfusion nor chelation was associated with lower employment or educational achievement.

Conclusions—Individuals with thalassemia in North America can achieve higher education; however, full-time employment remains a problem. Transfusion and chelation do not affect employment or education status of this patient population.

Keywords

Employment; Education; Thalassemia

INTRODUCTION

Thalassemia is a congenital blood disorder requiring chronic blood transfusions and iron chelation therapy to remove excess iron from the body [1,2]. Advances in the management of thalassemia have resulted in increased life expectancy for those affected [3]. This has contributed to the emergence of new clinical complications that include low bone mineral density, pulmonary hypertension, hypogonadism and cardiac failure [4,5]. However, as individuals with thalassemia age, they face challenges not experienced by previous generations of people with thalassemia. These might include poor health-related quality of life [6], maintaining adequate education and employment, experiencing fulfilling relationships, and starting a family. Frequent hospital and outpatient visits required for the management of thalassemia and its complications may create obstacles for maintaining employment and achieving higher education, as with other chronic illnesses [6-10]. However, there are presently no data available on the prevalence of employment and education difficulties for people with thalassemia. What factors might contribute to success or difficulties in these areas have not been studied. Without this information, comprehensive thalassemia centers are not able to develop effective psychosocial support plans for their patients.

Achieving higher education and holding a job not only can improve the quality of life of patients with thalassemia and provide financial stability, but might predict the quality of medical care received. In the United States, the cost and availability of healthcare is partially or completely covered by employers; therefore people with thalassemia who are not employed may not be able to afford healthcare and may not have adequate access to the resources they need for their care. While Canadians with thalassemia benefit from governmental insurance plans, federal and local resources are more limited for people in the United States with chronic conditions, including thalassemia.

Few reports in the literature examine health-related quality of life in thalassemia [6,11,12], with no data on education and employment status. Therefore the aim of this report is to investigate the employment and education status of children and adults with thalassemia in North America and to identify potential factors affecting these.

PARTICIPANTS AND METHODS

Participants

The Thalassemia Clinical Research Network (TCRN) is an NIH/NHLBI-funded clinical research network composed of 5 core centers in North America, 18 clinical satellites, and a data-coordinating center (Appendix 1). The TCRN developed a registry to characterize demographic and clinical features of North American patients with thalassemia, to highlight areas requiring clinical research, and to identify candidates eligible for clinical research

protocols. Patients with thalassemia of all ages who were diagnosed with β-thalassemia (intermedia or major), HbH disease, HbH Constant Spring Disease, Eβ-thalassemia, or homozygous α-thalassemia were eligible for participation. Current and retrospective data for this report were entered once for each participant during the time period from May 2000 through October 2006.

The protocol was approved by the TCRN Data and Safety Monitoring Board and by the Institutional Review Boards of all TCRN institutions. All participants provided signed informed consent. Eight hundred thirty six patients enrolled in the TCRN registry. Observations from thirty participants with successful stem cell transplant were excluded from this analysis, as were 161 children under school age, and 12 children with missing grade level. A total of 633 patients with thalassemia (349 adults and 284 school age children) were included in this analysis. Of the 349 adults, 327 had complete data on employment status and 327 on education level; however, these were not always the same participants with data on both. Information on precise refusal rates for the study and demographics of patients who refused were not collected. On the average, centers estimate that the refusal rate was less than ten percent.

Statistical Analysis

Employment in adults only was defined as employed (full-time or part-time). For those who did not report employment, we use the terms “not employed” and “non-employment” rather then “unemployment” or “unemployed.” Unemployment has a specific US Census Bureau definition, of “people who are jobless, looking for jobs, and available for work” [http://www.bls.gov/cps/cps_htgm.htm]. We did not ask participants who did not report employment if they were looking for a job or were available to work. This precludes any comparisons to census unemployment data.

Education in adults was categorized as less than high school, high school diploma, college degree, or post-college education. Education in children was defined as whether or not the child was at or above expected grade level. Expected grade level was calculated based on the assumption that a child of age 5 years before September 1 would be expected to enter kindergarten. As the cut-off date for kindergarten entry varies by region and public/private schools, this method of calculation was conservative for children with birthdays in the fall. Additionally, parents responding to the question of their child’s grade level in the summer may have answered either as to grade completed or grade to be entered, thereby adding to the conservative nature of the estimation.

Univariate analysis was used to assess the association between education and employment and a number of social and clinical variables. Social variables included age, gender, race/ethnicity (White, Asian, other), site of treatment (United States versus Canada). Education and employment status for the study population was compared with data from the U.S. Census Bureau. Clinical variables included transfusion and chelation status, most recent serum ferritin, hemoglobin level (available only in transfused patients), and clinical complications (heart disease, liver failure or cirrhosis, lung disease, pulmonary hypertension, arthralgia or arthritis, HIV, hepatitis C, osteoporosis/fracture, hypogonadism, hypothyroidism, hypoparathyroidism, diabetes, short stature). Secondary analysis used and expanded geographical location that divided participants in the U.S. into East Coast, West Coast, and Central states. Sub-analyses were performed in transfused patients to assess possible differences in this population. Finally, a sub-analysis excluding children younger than 10 years of age was conducted to assess the significance of complications in children.

Multivariate effects of predictors significant in univariate analysis were modeled using logistic regression for employment and a proportional odds model for education. In the

proportional odds model, the assumption of proportional odds was tested. An α-level of 0.05 was used in the determination of statistical significance. All analyses were conducted using SAS version 9.1.3 (Cary, NC, USA).

RESULTS

Participant demographics are described in Table I. Thirty percent of participants were Canadian. Among patients from the U.S., 40% were from the West Coast, 53% from the East Coast, and 7% from central states. Thirty two percent of patients had at least two complications.

Employment in adults

Forty seven percent of adults were employed full-time, 23% part-time and 30% not employed (Table I). Of those who were not employed, 53% were male, 42% were Asian, 40% were White, 60% were in the U.S., 67% were on regular blood transfusions and chelation, and 48% had at least two complications. Of those who were employed (full-time and part-time), 54% were female, 67% were white, 66% were in the U.S., 72% were on regular blood transfusion, 77% were on regular chelation therapy, and 52% had at least two complications. The most common complications in both the employed and not employed groups were osteoporosis (employed, 49%; not employed, 49.5%), hypogonadism (employed, 42%; not employed, 34%) and short stature (employed, 34%; not employed, 46%).

We compared the employment rate of our study population with the US population 16 years and older using U.S. census bureau data (www.census.gov, access date Feb 2, 2009). We found the employment rate (70%) in this study population to be slightly higher than in the general U.S. population (66.2% in 2003). However, 76% of the employed U.S. population work full-time, whereas 67% of employed individuals with thalassemia were employed full-time. The employment rate in this study population was considerably higher than the employment rate for people with disabilities in the U.S. (20.2% in October 2008). At this time there is no data available in the U.S. census on employment for disabled people before 2008.

In univariate analysis, age, gender, nationality, transfusion and chelation status, clinical complications, and number of complications were not significantly associated with employment status. Multivariate analysis of factors associated with employment (race, lung disease, region) is summarized in Table II. Only race was found to be a significant predictor of employment. The odds of employment for Whites were 2.8 times that of Asians. This finding is similar to the general population census where the rate of employment for Whites is higher than the rate for Asians, (among 20 – 54 year old individuals). Lung disease and patients' geographic location were no longer significantly associated with employment after adjusting for other variables.

The multivariate sub-analysis of transfused adult patients is summarized in Table III. Race continued to be a significant predictor of employment status with Whites more likely to be employed than Asians (OR=2.7). Regular chelation was strongly associated with employment in the general study population. Transfused patients who were receiving regular chelation therapy have 6.7 times higher odds of employment compared to those not receiving regular chelation. Liver failure (n=5; OR=7.9) and HIV positive status (n=6; OR=9.5) appeared to have a significant association with non-employment in transfused adult patients.

Education in adults

Participants' educational attainment is described in Table I. Fourteen percent of adults achieved post college education, 61% had a college degree, 17% had a high school diploma and 8% had less than a high school education. These rates are significantly higher than the educational attainment of people 16 years and older in the general U.S. population in 2000-2006 (www.census.gov, access date Feb 2, 2009), where about 8% had post college education, 25% had associate or bachelor degree and 30% were high school graduates.

Multivariate analysis of factors associated with education in adults (age, female gender, regular transfusion and chelation, lung disease) has been summarized in Table II. Age, gender, regular transfusion, and lung disease remained significant predictors of education level. Regular chelation was no longer significantly associated with education after adjusting for other variables. For each extra 10 years of age, participants had 1.67 times higher odds of having higher education levels. Men were less likely than women to have higher education level, with females having 2.1 times higher odds. This is consistent with U.S. Census Bureau data, with more females attaining higher education compared to their male counterparts since 2002 [13]. Patients who were regularly transfused were more likely to have achieved higher education level (OR=2.1). Adult patients who had lung disease were less likely to have achieved higher education (OR= 0.07).

In the multivariate sub-analysis of transfused adult patients, older age and female gender continued to be significant predictors of higher educational level. Regular chelation also remained a significant predictor of higher education in transfused adult patients, with regular chelation associated with a 3.9 times higher odds of higher education level. Patients' geographic location was not included in the regression model as it was not significant in univariate analysis.

Education in children

Of the 284 school-age children, 86% were at or above expected grade level. Twelve percent of children had at least two complications. In multivariate analysis of factors associated with education in children (younger age, Canadian nationality and not having hypogonadism), only younger age (OR=5.7 for 10 year increments, 95% CI: 2.0 – 16.7) and Canadian nationality (OR=5.6, 95% CI: 1.5 – 20.0) were significantly associated with being at or above expected educational level. Gender, race, transfusion and chelation status, and clinical complications were not significantly associated with education level in children.

In the multivariate sub-group analysis with 160 transfused children, younger age (OR=16.1 for 10 year increments, 95% CI: 3.2-83.3) and Canadian nationality (OR=8.3, CI:1.1-50) remained significant predictors. Additionally, female gender was significantly associated with being at or above expected educational level (OR=2.86, CI:1.07-7.69). Additionally, children on the East Coast compared to West were found more likely (OR=3.85, 95% CI: 1.27-11.1) to be at or above their expected education level.

DISCUSSION

This report evaluated the employment and education status of individuals with thalassemia. We found that adults with thalassemia in North America attained higher education compared to the general U.S. population. Whereas the employment rate for adults with thalassemia was comparable to the general U.S. population, the full-time employment rate was somewhat lower. Transfusion and chelation therapy do not appear to be obstacles to the attainment of higher education or employment, nor to the educational progress of children. Clinical complications also did not appear to negatively impact education level or

employment status, with the exception of patients with liver failure and those who were HIV positive.

The positive relationship between regular chelation and employment rates in transfused adults may be due to improved health and a reduction in complications compared to patients who were not chelating regularly. Alternatively, these patients may be more motivated in general, as evidenced by their ability to adhere to regular chelation therapy. Regardless, effective chelation requires both discipline and commitment, two qualities for which employers look. Patients with HIV or liver failure may have been sicker compared with other patients with thalassemia, resulting in reduced employment.

In contrast to our findings with employment, the educational attainment of study participants was significantly higher than the educational attainment of people 16 years and older in the general U.S. population in 2000-2006 (www.census.gov, access date Feb 2, 2009). This could be explained by the fact that most adult with thalassemia residing in the U.S. and Canada are foreign born immigrants [13]. According to the U.S. census bureau report in 2003 [14], foreign born populations in the U.S. had a lower proportion of high school graduates than the native population. However, at the bachelor's level, foreign born Blacks and non-Hispanic Whites fared better than their native counterparts. Unfortunately, this study did not collect information on county of birth.

Adults who were regularly transfused were more likely to have achieved higher education level. This is difficult to explain as hemoglobin level was not found to be a predictor of education. Nevertheless, it is encouraging that individuals with thalassemia do not appear to have difficulty in achieving higher education. Additionally, children with thalassemia are generally on track with their peers. Regional differences (Canada versus U.S.; East versus West coast) may reflect general differences in education systems. Alternatively, they may reflect the limitations inherent with this analysis, as assumptions were made as to what constituted expected grade level.

A limitation of this study is that it may not be possible to generalize our results from the study population to all patients with these disorders in North America. It is possible that patients able and willing to come to the TCRN network sites are better educated than those who live far from these large urban centers. While the total number of patients with symptomatic thalassemia syndromes living in North America is not known, the total population is not likely to be more than a few times the total in the registry. The largest thalassemia population in Canada is in the Toronto area, and the largest groups in the US are around the large metropolitan areas where TCRN sites are located.

Further studies are needed to investigate the unemployment rate, barriers to employment, and motivations for seeking jobs for adults with thalassemia. Future studies are needed to compare the education and employment status of individuals with thalassemia in North America with those in other countries.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

Acknowledgments

This work was supported by a cooperative agreement with the National Heart, Lung, and Blood Institute, National Institutes of Health (U01-HL-65232 to Children's Hospital of Philadelphia; U01-HL-65233 to University Health Network Toronto General Hospital; U01-HL-65239 to Children's Hospital and Research Center; U01-HL-65244 to Weill Medical College of Cornell University; U01-HL-65260 to Children's Hospital Boston; and U01-HL-65238 to New England Research Institutes) and in part by National Institutes of Health grant M01-RR01271 to the Pediatric

Clinical Research Center. The authors would like to thank the children and adults with thalassemia and their families who agreed to be enrolled in the TCRN registry.

REFERENCES

1. Forget, BG.; Cohen, A. Thalassemia Syndromes. In: Hoffman, R.; Benz, EJ., Jr; Shattil, SJ., et al., editors. Hamtology:Basic Principles and Practices. Fourth Ed. Elsevier, Churchill LIvingstone; Philadelphia, PA: 2005. p. 557-598.
2. Cohen AR, Galanello R, Pennell DJ, et al. Thalassemia. Hematology Am Soc Hematol Educ Program. 2004:14–34. [PubMed: 15561674]
3. Modell B, Khan M, Darlison M, et al. Improved survival of thalassaemia major in the UK and relation to T2* cardiovascular magnetic resonance. J Cardiovasc Magn Reson. 2008; 10:42. [PubMed: 18817553]
4. Borgna-Pignatti C, Cappellini MD, De Stefano P, et al. Survival and complications in thalassemia. Ann N Y Acad Sci. 2005; 1054:40–7. [PubMed: 16339650]
5. Cunningham MJ, Macklin EA, Neufeld EJ, et al. Complications of beta-thalassemia major in North America. Blood. 2004; 104:34–39. [PubMed: 14988152]
6. Payne KA, Rofail D, Baladi JF, et al. Iron chelation therapy: clinical effectiveness, economic burden and quality of life in patients with iron overload. Adv Ther. 2008; 25:725–472. [PubMed: 18704280]
7. Kraut A, Walld R, Tate R, et al. Impact of diabetes on employment and income in Manitoba, Canada. Diabetes Care. 2001; 24:64–68. [PubMed: 11194243]
8. Milton B, Holland P, Whitehead M. The social and economic consequences of childhood-onset Type 1 diabetes mellitus across the lifecourse: a systematic review. Diabet Med. 2006; 23:821–829. [PubMed: 16911617]
9. Robinson N, Stevens LK, Protopapa LE. Education and employment for young people with diabetes. Diabet Med. 1993; 10:983–989. [PubMed: 8306598]
10. Stewart WF, Ricci JA, Chee E, et al. Lost productive time and costs due to diabetes and diabetic neuropathic pain in the US workforce. J Occup Environ Med. 2007; 49:672–679. [PubMed: 17563611]
11. Pakbaz Z, Treadwell M, Yamashita R, et al. Quality of life in patients with thalassemia intermedia compared to thalassemia major. Ann N Y Acad Sci. 2005; 1054:457–461. [PubMed: 16339697]
12. Telfer P, Constantinidou G, Andreou P, et al. Quality of life in thalassemia. Ann N Y Acad Sci. 2005; 1054:273–282. [PubMed: 16339675]
13. Vichinsky EP, Macklin EA, Waye JS, et al. Changes in the epidemiology of thalassemia in North America: a new minority disease. Pediatrics. 2005; 116:e818–e825. [PubMed: 16291734]
14. Stoops, N. Educational attainment in the United States. US Census Bureau; 2004. p. 20-550.

Table 1

Characteristics of study participants

	Adults (n=349)	Children (n=284)	Total (n=633)
Age (years) median, (range)	28 (18-75)	11 (5-17)	19 (5-75)
Male/Female (%)	47.3/52.7	46.8/53.2	47.1/52.9
Race (%)			
White	57.5	26.9	43.7
Asian	29.6	56.2	41.5
Other	13.0	17.0	15.0
Nationality (%)			
American	66.2	74.7	70.0
Canadian	33.8	25.4	30.0
Employment (Adult only) (%)			
Employed full-time	47.4	-	47.4
Employed part-time	22.9	-	22.9
Notemployed	29.7	-	29.7
Education (Adult only) (%)			
< High school	8.6	-	8.6
High school	16.8	-	16.8
College	60.6	-	60.6
Post college	14.1	-	14.1
Education (Children only)(%)			
Grades 1-3	-	31.3	31.3
Grades 4-8	-	41.9	41.9
Grades 9-12	-	26.4	26.4
GED	-	0.4	0.4
≥ 8 Transfusions per year (%)	69.5	56.3	63.6
Regular Chelation (%)	72.3	55.6	64.8
Most recent Serum Ferritin	27.4	9.9	19.7
≥ 2500 ng/ml (%)			
Hemoglobin ≥ 9 g/dL	84.9	78.7	82.4
(Transfused patients only) (%)			
Number of complications≥2 (%)	49.0	11.6	32.2

Table 2

Multivariate Analysis of factors associated with employment and education in 327 adults with thalassemia

	Adjusted OR	95% CI
Non-employment		
Race		
Asian vs. White	**2.76**	**[1.50,5.06]**
Other vs. White	**2.54**	**[1.20, 5.36]**
Lung disease		
Yes vs. No	8.03	[0.78,83.1]
Region		
West vs. East	1.24	[0.57,2.67]
Mid vs. East	0.70	[0.17,2.95]
Canada vs. East	1.16	[0.64,2.10]
Education		
Age	**1.67**	**[1.29,2.15]**
(in 10 year increment s)		
Gender		
Female vs.Male	**2.08**	**[1.32,3.23]**
Transfusion Status		
≥ 8 / year vs. < 8 / year	**2.06**	**[1.03, 4.10]**
Chelation Status		
Regular vs. Not	1.14	[0.56, 2.31]
Lung disease		
Yes vs. No	**0.07**	**[0.01, 0.49]**

Possible predictors were age, gender, race/ethnicity, nationality, transfusion and chelation status, serum ferritin, and clinical complications(heart disease, liver failure or cirrhosis, lung disease, pulmonary hypertension, arthralgia or arthritis, HIV, osteoporosis/fracture, hypogonadism, thyroid disease, parathyroid disease, diabetes, Hep C ,stature). Predictors found significant in univariate analyses were entered into multivariate models. A logistic regression model was used for employment and a proportional odds model for education. OR = odds ratio. CI = confidence interval. The OR models the odds of non -employment (vs. employment) or higher education (post-college vs. college vs. high school vs. < high school). Significance at level 0.05 is indicated in bold type.

Table 3

Multivariate sub-analysis of factors associated with employment and education in 242 transfused adults with thalassemia

	Adjusted OR	95% CI
Non-employment in Adults		
Race		
Asian vs. White	**2.67**	**[1.18,6.04]**
Other vs. White	1.26	[0.42,3.78]
Chelation Status: DFO or L1		
Not Regular vs. Regular	**6.67**	**[1.96, 25.0]**
Liver Failure		
Yes vs. No	**7.86**	**[2.14,28.8]**
HIV		
Positive vs. Negative	**9.45**	**[1.61, 55.5]**
Education in Adults		
Age (in 10 year increments)	**1.61**	**[1.10,2.35]**
Gender		
Female vs.Male	**2.38**	**[1.37,4.17]**
Chelation Status: DFO or L1		
Regular vs. Not regular	**3.87**	**[1.36, 11.0]**
Most recent serum ferritin Level		
>= 2500 vs. < 2500	0.71	[0.40, 1.27]

Possible predictors were age, gender, race/ethnicity, nationality, transfusion and chelation status, serum ferritin, and clinical complications(heart disease, liver failure or cirrhosis, lung disease, pulmonary hypertension, arthralgia or arthritis, HIV, osteoporosis/fracture, hypogonadism, thyroid disease, parathyroid disease, diabetes, Hep C ,stature). Predictors found significant in univariate analyses were entered into multivariatemodels. A logistic regression model was used for employment and a proportional odds model for education. OR = odds ratio. CI = confidence interval. The OR models the odds of non-employment (vs. employment) or higher education (post-college vs. college vs. high school vs. < high school). Significance at level 0.05 is indicated in bold type.