





CHILDREN'S ORTHOPAEDICS

An assessment of the impact of developmental dysplasia of the hip on patients' wellbeing

CALCULATION OF DISABILITY WEIGHT AND COUNTRY-SPECIFIC UTILITY WEIGHTS



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Aims

Within healthcare, several measures are used to quantify and compare the severity of health conditions. Two common measures are disability weight (DW), a context-independent value representing severity of a health state, and utility weight (UW), a context-dependent measure of health-related quality of life. Neither of these measures have previously been determined for developmental dysplasia of the hip (DDH). The aim of this study is to determine the DW and country-specific UWs for DDH.

Methods

A survey was created using three different methods to estimate the DW: a preference ranking exercise, time trade-off exercise, and visual analogue scale (VAS). Participants were fully licensed orthopaedic surgeons who were contacted through national and international orthopaedic organizations. A global DW was calculated using a random effects model through an inverse-variance approach. A UW was calculated for each country as one minus the country-specific DW composed of the time trade-off exercise and VAS.

Results

Over a four-month period, 181 surgeons participated in the survey, with 116 surgeons included in the final analysis. The global DW calculated to be 0.18 (0.11 to 0.24), and the country-specific UWs ranged from 0.26 to 0.89.

Conclusion

This is the first time that a global disability weight and country-specific utility weights have been estimated for DDH, which should assist in economic evaluations and the development of health policy. The methodology may be applied to other orthopaedic conditions.

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Introduction

Developmental dysplasia of the hip (DDH) is a spectrum of hip disease that develops during childhood, including dislocated hips, dislocatable hips, subluxed hips, and dysplastic hips. It is the leading cause of early hip osteoarthritis and the largest single cause of young patients requiring a total hip arthroplasty.1 Infant screening practices for DDH vary considerably between countries,

with universal ultrasound screening for newborns adopted in Austria, Germany, and Mongolia,² selective ultrasound screening in the UK, and no formalized screening within the USA. There is ongoing debate in the literature as to the clinical- and cost-effectiveness of methods of screening.3 In order to elucidate the burden of DDH in the world, evaluate cost-effectiveness of screening and treatment methods, and guide allocation of

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VOL. 4, NO. 3, MARCH 2023 120 healthcare resources, it is necessary to quantify its impact on population health.

Disability-adjusted life years (DALYs) are a method of quantifying the burden of a particular health state by reference to the years of healthy life lost. DALYs are calculated by the sum of the years of life lost due to early mortality and the years of healthy life lost due to disability.4 The DALY uses a disability weight (DW) for each disease, which is a measure of the level of disability between 0 (full health with no disability) and 1 (disability level equivalent to death).5 Murray6 defined six classes of disability severity according to DW, ranging from class one for disability consisting of limited ability to perform at least one activity in the areas of recreation, education, procreation, or occupation (DW of 0.096) to class six, indicating requirement for assistance with activities of daily living (DW of 0.920). As context-independent DWs are not calculated by reference to local context, in this study, the calculated DW is referred to as 'global'.

Quality-adjusted life years (QALYs) are another method to measure a health state, with reference to the duration of time and quality of life in that health state. A QALY is calculated by multiplying the years lived in a health state, by the country-specific utility weight (UW) of the disease.⁷ A UW is a value for each disease representing its effect on quality of life on a scale from 0 (representing death) to 1 (representing life in full health).⁸ The UW for a disease in a specific setting can often be established as one minus the disability weight for that population.⁹ As UWs are context-specific, they highlight populations where patients have a lower quality of life by reason of a particular disease.

Musculoskeletal (MSK) diseases, which typically result in pain and impaired mobility, are the second highest contributor to the global burden of disease,10 with 1.7 billion people suffering musculoskeletal diseases across all age groups. 11 Currently, there are few metrics that allow direct comparison of the health burden for orthopaedic conditions with other health conditions. For example, the Global Burden of Disease (GBD) study published in 2013 does not list the DW for specific MSK lower limb conditions.¹² The GBD study classifies MSK lower limb conditions as mild (DW of 0.023), moderate (DW of 0.079), severe (DW of 0.229), or most severe (DW of 0.304). The study does not offer guidance on how to classify conditions, such as DDH, according to these descriptors. This lack of practical applicability renders these values of little use as a health metric concerning orthopaedic conditions.¹² Given the importance of DWs and UWs within public health and health economics, clarity regarding these metrics for orthopaedic conditions will provide an evidentiary basis for inclusion in the allocation of healthcare resources. In particular, quantification of a global DW and country-specific UWs for DDH is

necessary to provide a basis for cost-effectiveness evaluation of the various infant screening programmes.

The country-specific UWs and global DW for DDH have not previously been calculated. This study aims to calculate the global DW and country-specific UWs for DDH, in order to inform health economic policy relating to the screening and treatment for DDH and allocation of healthcare resources.

Methods

Study design. We conducted a cross-sectional survey to determine the disability weight for DDH through an online international survey of surgeons treating DDH. Approval was obtained from the Institutional Review Board (IRB22-0088).

Setting and survey tool. The online survey (Supplementary material i) was designed using Qualtrics XM (Qualtrics, USA), and was open for completion between 1 March 2022 and 15 April 2022.

Respondents were asked in which country they provide orthopaedic care, whether they were fully licensed in that country, and what percentage of their practice involved managing DDH or its chronic sequela. All responses were anonymous with no identifying information collected regarding individual respondents. Participants were asked to complete the survey only once.

A recent systematic review suggested there was no gold standard for calculation of DW, and the majority of previous studies used more than one technique. Therefore, this study used the preference ranking (PR) series, time trade-off (TTO) exercise, and visual analogue scale (VAS) for the calculation of the DW.⁵ For the calculation of the UWs for DDH, only the results of the TTO and the VAS have been included, as historically the UW calculations have been done with standard gamble or timevaluation techniques, such as TTO, VAS, or validated questionnaires.¹³ The use of vignettes or PR series are not a common practice for UW determinations.¹³

First, respondents were asked to complete a preference ranking (PR) series, in which they were asked to rank untreated DDH among other medical conditions with known DWs in order to produce ordinal data. Five lists were presented consisting of ten medical conditions and their associated DWs, as established in the GBD 2013 study, 12 which was selected due to its inclusion of a broad range of conditions. The lists comprised MSK and non-MSK medical conditions with DWs ranging between 0 to 0.05, 0.1 to 0.2, 0 to 0.2, 0.1 to 0.3, and 0 to 0.4 and recorded as a cardinal value from the ordinal PR methodology.

Second, TTO was used to identify the number of years of full health that a person would be willing to trade for a life lived with DDH. Respondents were asked whether they would be willing to trade a given number of years of healthy life, starting at age 80 years and decreasing in

Table I. Respondent characteristics.

Country	Final number of responses	Greater than 50% of practice is managing paediatric	Percentage of practice involves	Company to 12
		orthopaedic conditions	managing DDH or its sequela	Country level income
Australia	10	8	30.3	High
Austria	2	2	30.0	High
Belgium	2	2	7.5	High
Canada	2	2	20.0	High
Chile	2	1	20.0	High
Czech Republic	1	0	25.0	High
Denmark	1	1	1.0	High
Finland	1	1	5.0	High
France	7	6	17.9	High
Greece	1	1	5.0	High
Hungary	1	1	7.5	High
srael	7	7	10.4	High
taly	6	6	34.2	High
apan	1	1	30.0	High
Saudi Arabia	1	1	70.0	High
Netherlands	4	4	32.5	High
New Zealand	2	2	10.0	High
Poland	6	3	28.3	High
Portugal	1	1	40.0	High
Romania	1	1	40.0	High
Spain	1	0	10.0	High
Switzerland	4	3	10.3	High
JK	6	6	28.0	High
USA	27	26	16.4	High
Costa Rica	1	1	75.0	Upper-middle
Georgia	1	1	60.0	Upper-middle
ordan	1	1	40.0	Upper-middle
Libya	2	0	20.0	Upper-middle
Malaysia	1	0	10.0	Upper-middle
Mexico	1	1	30.0	Upper-middle
North Macedonia	1	1	25.0	Upper-middle
Peru	1	1	40.0	Upper-middle
South Africa	1	1	40.0	Upper-middle
Turkey	2	1	25.0	Upper-middle
ndia	3	2	14.2	Lower-middle
ran	2	2	30.0	Lower-middle
Morocco	1	1	5.0	Lower-middle
Pakistan	1	1	40.0	Lower-middle

increments of five years, for 80 years of life with DDH. At the point a respondent was willing to trade, the resultant value for DDH was calculated as the number of healthy years of life traded (t) divided by 80.

Finally, respondents were presented with a VAS anchored by perfect health (0) and death (100). Using the VAS, respondents were asked to indicate placement of DDH relative to the extremes of perfect health and death with results recorded as continuous data.

Participants. The survey was distributed to surgeons around the world through international paediatric orthopaedic and general orthopaedic societies in seeking broad country representation. Respondents were included if they were fully licensed surgeons in their country

of practice, with clinical experience in managing DDH or its sequelae into adult years. Respondents were excluded if they were not fully licensed, did not have appropriate experience in managing DDH (defined as less than 50% of practice managing paediatric orthopaedic conditions and less than 10% of practice managing DDH or chronic sequelae), or did not complete at least one DW measurement task.

Statistical analysis. Statistical analysis was conducted using Stata version 17 (StataCorp, USA). The survey tool only collected quantitative data of continuous and count nature. Supplementary material ii demonstrates the statistical analysis. Data was summarized per country (Table I).

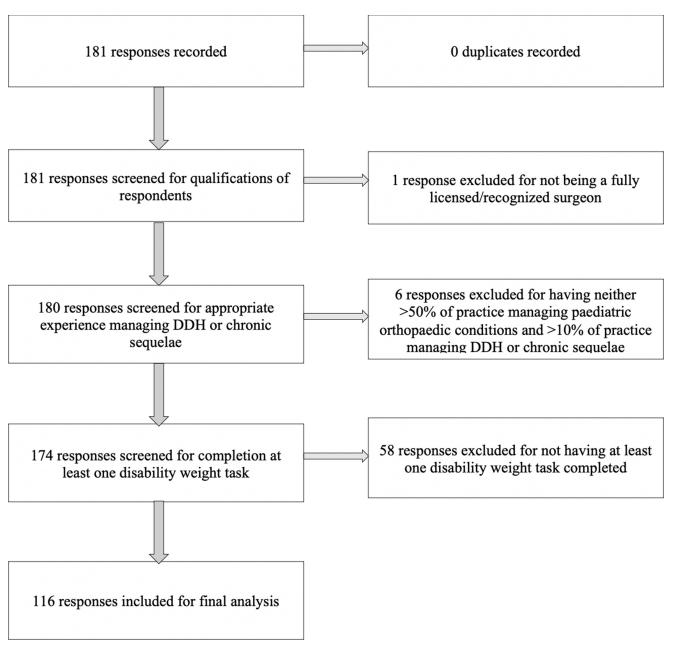


Fig. 1
Exclusion of 181 responses to survey.

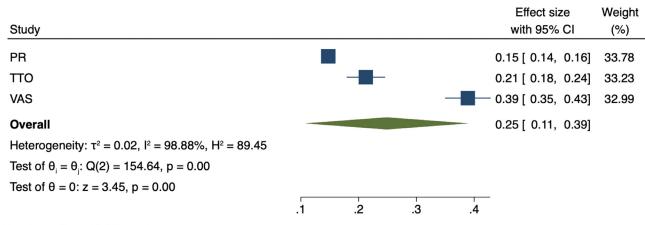
Disability weight calculation. In order to obtain a DW based on the three methodologies, several intermediary steps were necessary:

1) Obtaining intermediate DW based on each methodology

Preference-ranking: For the PR series, each list was scored as the average of the disability weights for the conditions above and below the assigned rank of DDH, and the scores for the five lists were averaged to produce the DW for each respondent. Responses that were less than option one (lowest DW) or greater than option ten (highest DW) were excluded.

Time trade-off exercise: For the TTO exercise, the DW for each respondent was calculated as 1-(t/T), where (t) represented the time in good health traded and (T) was given as 80 years with DDH.

Visual analogue scale: The VAS was converted to a DW for each respondent as (VAS/100).



Random-effects REML model

Fig. 2

Disability weight for developmental dysplasia of the hip using preference ranking (PR), time-trade off (TTO), and visual analogue scale (VAS) calculated using random-effects REML model.

2) Preparing intermediate DWs for usage in inverse-variance technique

The mean and variance for three intermediate DWs was calculated. No weighting was used to adjust for the number of answers given per country or region.

3) Moving from three intermediate DWs to one global DW

An inverse-variance technique was used to obtain a single DW from the three intermediate DWs. The decision to move forward with an inverse-variance technique, rather than a method that allows for weighted averages, was because it gives more weight to variables with less variability. With this methodology, the variability in the end-product is kept to a minimum and more weight is given to values with a higher rate of agreement among participants. A random effects model was used in preference to a fixed-effects model, as it was believed that the survey results are generalizable beyond the included data. Furthermore, the DWs, calculated by each of the three methodologies, are different measurements. 15

Country-specific utility weight calculations. In order to obtain country-specific UWs, several intermediary steps were necessary:

1) Obtaining intermediate UWs based on each methodology

For each country, the corresponding respondents' disability weights according to the TTO and VAS methods described above were converted to UWs by 1-TTO and 1-VAS. This has been shown to be a reliable method of calculating a UW from DWs.⁹ The TTO and VAS were used for the UW calculations as these are direct evaluation methods, while the PR is not considered to be a direct evaluation method.¹³

2) Preparing UWs for use in inverse-variance technique

For each country, the mean and variance for the intermediate UWs were calculated. No weighting was used to adjust for the number of answers given per country.

3) Moving to country-specific UWs

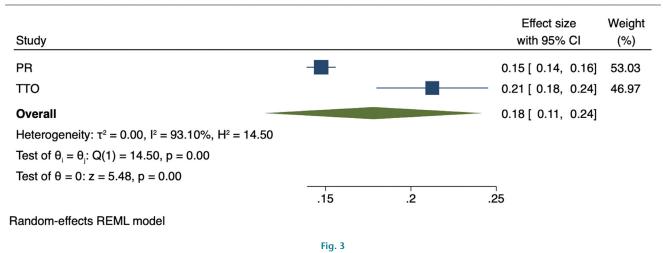
An inverse-variance technique was used to obtain a single UW for each country.

A p-value < 0.05 was considered significant in all calculations.

Results

Overall, 181 survey responses were received between 1 March 2022 and 15 April 2022; 116 respondents met inclusion and exclusion criteria (Figure 1). The study comprised respondents from 38 countries (Table I). Of these, 24 were higher-income countries, ten upper-middle-income countries, four lower-middle-income countries, and no low-income countries according to World Bank Classification. Among the respondents, the percentage of clinical practice involving DDH was 22.3% (5% to 80%).

Global disability weight. The preference ranking series gave an estimated disability weight of 0.15 (95% confidence interval (CI) 0.14 to 0.16). The TTO exercise gave an estimated disability weight of 0.21 (95% CI 0.18 to 0.24). The VAS gave an estimated disability weight of 0.39 (95% CI 0.35 to 0.43). The calculated global disability weight was 0.25 (95% CI 0.11 to 0.39), as shown in Figure 2. However, there was large heterogeneity (I² 98.88%; Cochran's Q test 154.64), which can be visually observed in Figure 2. Therefore, the VAS was excluded as an outlier. Using the PR and TTO, the final global disability weight for DDH was calculated at 0.18 (p-value ≤



Disability weight for developmental dysplasia of the hip using preference ranking (PR) and time-trade off (TTO) calculated using random-effects residual maximum likelihood (REML) model.

0.005; 95% CI 0.11 to 0.24; I² 93.10%; Cochran's Q test 14.50), as shown in Figure 3.

Country-specific utility weight. The values calculated as 1-TTO ranged from 0 to 0.94. The values calculated as 1-VAS ranged from 0.05 to 0.96. The calculated country-specific UWs ranged from 0.26 (95% CI -0.25 to 0.77) in Morocco to 0.89 (95% CI 0.86 to 0.93) in Finland (Figure 4).

Discussion

This study calculated the global DW for DDH as 0.18 (95% CI 0.11 to 0.24). Using the 2013 GBD table, this falls within the spectrum of a "severe" lower limb MSK problem (DW 0.112 to 0.232). Compared to the calculated DW of other orthopaedic conditions, this DW falls between severe leg problems with a DW of 0.165 and severe neck pain with a DW of 0.229. The UWs for each country were calculated as ranging from 0.26 to 0.89. To the authors knowledge, this is the first time a global disability weight or country-specific UWs have been calculated for DDH.

According to the six-grade gradient scale described by Murray,⁶ the global DW determined by this study of 0.18 for untreated DDH was greater than class 1 (i.e. diseases limiting the ability to perform at least one activity in one of the following areas: recreation, education, procreation or occupation (DW of 0.096)) and nearer to class 2 (i.e. diseases limiting the ability to perform most activities in one of the following areas: recreation, education, procreation or occupation (DW of 0.200)). This description correlates well with the reality of living with untreated DDH in the authors' clinical experience, which may be considered as an indirect external validation for the value of our DW.

When comparing the countries in Table I, ordered by country income level, with those in Figure 4, ordered by

UW, it is apparent that there was a gradient in countryspecific UWs according to country income level. This may allude to the possibility of an underlying pattern suggesting that the subjective experience of DDH differs according to the healthcare resources available. A lower UW indicates greater loss in quality of life for the person affected by this disease. It is known that children with delayed diagnosis and treatment experience a reduced quality of life compared to those who earlier diagnosis and treatment. 16 Therefore, the disparity in UWs may serve to identify jurisdictions in which there would be a greater impact from investing in screening and treatment of DDH to minimize the loss of quality of life related to DDH. Furthermore, this disparity in UWs highlights the importance of local context when undertaking economic evaluation, such as a cost-effectiveness analysis (CEA), of population-wide screening programs, treatment methods, or public financing schemes for DDH treatment.

The common practice in orthopaedics and other surgical specialities was to use surrogate endpoints or proxies when executing cost-effectiveness analyses, for example, the disability weight for untreated cleft palate was used in a CEA for unilateral congenital talipes equinovarus.¹⁷ However, the concern with using proxies or surrogate endpoints like "successful treatment" is that it introduces an increased level of uncertainty within cost-effectiveness models. These high uncertainty levels may lead to inaccurate conclusions being drawn about the cost, effect or cost-effectiveness of surgical care,18 leading to exclusion of orthopaedic care from healthcare packages in lower- and middle-income countries. 19 The methodology used in this paper demonstrates that determining a global DW or country-specific UW for an orthopaedic condition may be done in a simple, low-cost and rapid manner. The quantification of additional disability weights for orthopaedic conditions will avoid the

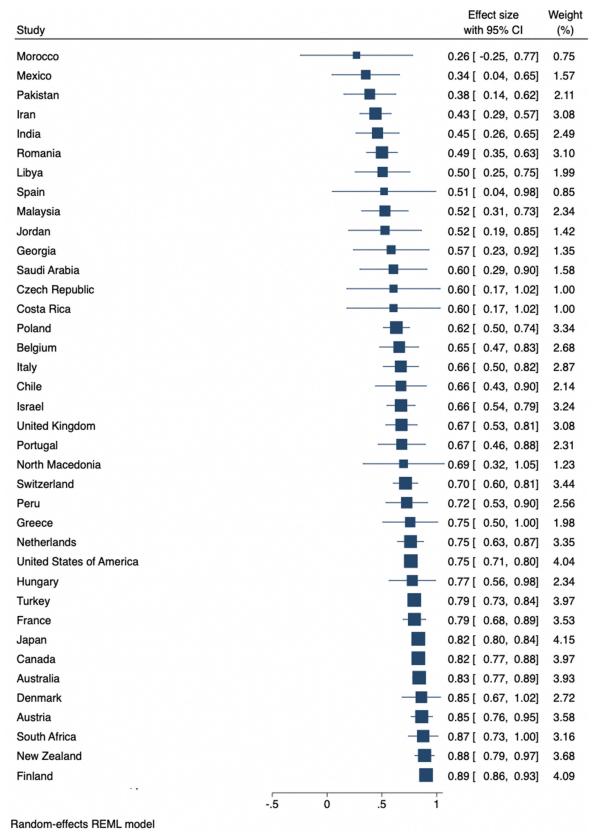


Fig. 4

Country-specific utility weight calculations using time-trade off (TTO) and visual analogue scale (VAS) using random-effects residual maximum likelihood (REML) model.

necessity to use proxies when undertaking CEAs and may facilitate appropriate prioritization and agenda setting for orthopaedic conditions in research and policy agendas.³ With differing national approaches towards screening of infants for DDH, the quantification of the global DW and country-specific UWs of DDH will enable context-specific CEAs of various local screening programmes.

The standard population used to determine a DW, as developed by the GBD team, is a group of members from the general population.¹² However, in more recent years, researchers have adopted different strategies including surveying patients affected by a certain disease or healthcare professionals with considerable experience in treating the disease.²⁰ The decision was made not to interview members of the general population, but to survey orthopaedic clinicians who manage patients with DDH on a regular basis. This decision required a reliance on their accurate understanding by the clinicians on the impact of DDH on patients' daily living. While an alternative would be to rely upon patients' perspective, there is a risk that their estimation of the disease burden would be influenced by their own adaptive strategies.²⁰ The inclusion of patients living with DDH, primary caregivers of children with DDH, or a group of people from the general public without DDH may provide an important additional perspective in future studies.

There are some limitations of this study to note. While the broad inclusion criteria for study participants were intended to maximize responses across the globe, the survey was only distributed in English, and likely limited participation by participants from non-English speaking backgrounds. High-income countries are overrepresented in the responses and no results were obtained from low-income countries. Furthermore, no responses were received from some countries with known high prevalence of DDH,21 and some countries had only one survey respondent, meaning the results must be applied with caution. The survey was intended for distribution by national orthopaedics organizations to suitably qualified and experienced orthopaedic surgeons. As the authors had no control over distribution of the survey by those organizations, and the survey was conducted anonymously, the surgeons' self-declared experience treating DDH cannot be validated and the number of non-responding invitees is not known. Consideration could be given to including other medical practitioners. such as general practitioners or a group of clinicians, who may collectively have a better ability to assess the relative weight of the health burden of DDH. Finally, the survey assumes DDH as a homogenous disease, despite it being a broad spectrum of conditions, though this is a necessary requirement for evaluation of healthcare economics in order to permit comparison across diseases and avoid fragmentation of data.

In conclusion, this study found the global DW for developmental dysplasia of the hip to be 0.18 (0.11 to 0.24), which is approaching the "severe leg MSK problem" classification in the 2013 GBD study. ¹² A UW for each country was calculated, ranging from 0.26 to 0.89. This is the first published literature on DW and UW in DDH, and these metrics will enhance consideration of this disabling condition in public health policy discussions.



Take home message

- First publication that disability weight and utility weights have been estimated in
- developmental dysplasia of the hip (DDH) with global
- This augments health economic understanding for the screening and treatment of DDH and can be used in the development of health policy interventions.

Supplementary material



Survey to calculate the disability weight for developmental dysplasia of the hip, and formulas for three methods to calculate disability weight.

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- Corlew: Conceptualization, Formal analysis, Investigation, Methodology, Resources, Supervision, Validation, Writing – review & editing.

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Data sharing:
 The data that support the findings for this study are available to other researchers from the corresponding author upon reasonable request.

Ethical review statement:

This study was performed in line with the principles of the Declaration of Helsinki. Exemption was received by the Harvard Faculty of Medicine Office of Regulatory Affairs and Research Compliance (IRB22-0088).

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