Appendix: Selected Published Analyses of Informal Care Time Valuation

1. Applications to specific health problems
	1. Replacement cost and direct measurement of informal care

Most studies of informal care in older adults and adults with chronic diseases utilize a replacement cost or proxy good approach. As previously discussed, much of the peer-reviewed literature assessing the cost of informal care centers around dementia in general or Alzheimer disease (AD) in particular. Dozens of studies of informal care costs for dementia have been published over the past 25 years, almost all of which have used replacement cost methods [1]. In addition to COI studies, AD or dementia is the one disease for which published CEA studies have typically included the costs of informal care [2]. For example, an early CEA of treatment for AD published in 1999 used the replacement cost estimates from the 1993 study by Rice et al. [3] to estimate the avoidable costs of delayed progression [4].

Informal care costs for dementia commonly use some variant of the replacement cost approach. A problematic feature in most of these studies is a lack of estimates of condition-specific informal care hours. Since elderly persons without dementia also require considerable care, it is often difficult to determine how much of the time spent engaging in informal care is attributable to dementia beyond usual caregiving of elderly relatives. Relatively few studies follow the lead of Ostbye and Crosse [5], as summarized in section 2, in empirically estimating incremental informal care time hours relative to what care would have been provided in a counterfactual scenario of the absence of dementia. Accurate estimates of incremental hours of informal care are necessary to assess the attributable burden of informal care. [6]

* 1. Opportunity cost approach and indirect measure of lost hours of work among informal caregivers

A number of CEAs and COI studies, particularly in North America, follow the recommendation of the First Panel in estimating the cost of informal care. In particular, costs have been estimated for child disability of various types resulting from foregone parental earnings comparing samples of families of affected and unaffected children and adolescents [7]. A body of economic research in the United States that has used representative surveys to compare employment and earnings among parents of children of differing health status has found that parents of children with disabling conditions are more likely to either stop working or reduce hours of employment [8-18]. Similar findings have been reported from European countries [19].

One of the worked examples for the First Panel used an unpublished study of foregone parental paid work for a small sample of families with children with spina bifida to estimate preventable costs associated with folic acid fortification [20]. Specifically, Kelly et al. assumed that parents lost 19 hours of paid work per week, which were valued by the combination of paid earnings and imputed market work [20]. Subsequently, Tilford et al. found that the primary caregivers in a US sample of children and adolescents with spina bifida in the US state of Arkansas were approximately one-third less likely to work for pay than a matched control group derived from representative population survey during the same time period (March 2003) [21]. The loss of paid work time averaged 7.5 to 11.3 hours per week over the course of a year, roughly half the reduction assumed by Kelly et al. in their prospective economic assessment. Using an age-sex wage profile derived from the general population and a 3% discount rate, the authors estimated a labor market productivity loss from birth to age 21 of $133,275 in 2002 dollars, which was primarily associated with withdrawal from the labor force. Those estimates of informal care costs have been included in two economic evaluations of folic acid interventions [22, 23]. Parental time cost of $214,900 in 2014 US dollars was equivalent to almost 40% of the lifetime incremental medical cost attributable to spina bifida [22].

Other researchers have assessed survey reports of informal care time associated with autism or autism spectrum disorder (ASD) in children. Variable estimates of hours of informal care have been reported in survey samples. In one UK survey, parents were reported to spend an average of 40 extra hours per week caring for their young child with an ASD [24]. That estimate is very high relative to other surveys; for example, an analysis of data from a representative, national US survey found that 73% of families of a child with an ASD ages 3 to 17 years reported spending <10 hours per week providing care or coordinating care for their child [25]. Similarly, Lavelle et al. found that having a child with ASD raised hours of care by 4 hours per week relative to parents of unaffected children, or 221 hours per year [26]. Those data came from an online US survey and included time spent on activities such as coordinating their child’s therapies, homework help, and travel to appointments and activities during the previous 12 months. Multiplied by an opportunity cost of $23 per hour, the economic value of informal care time per child with ASD was estimated at just over $5,000 per year on average [26]. Finally, Cidav et al. reported that for mothers of children with ASD, paid work was reduced by 7 hours per week and 364 hours per year relative to mothers of children with no health limitations, but no hourly wage was applied [27].

An alternative to estimating hours of time taken away from market work valued by an hourly wage is to estimate directly annual earnings comparing samples of affected and unaffected families [19]. In a 2005 US survey, Montes and Halterman found that mothers and fathers of children with ASD in kindergarten through 8th grade were less likely to be employed and more likely to work part-time than other parents [28]. The mean loss of parental income for each child with an ASD was roughly $6,000 per year in 2003 US dollars, similar to the indirect estimate of Lavelle et al. [26]. In contrast, Cidav et al. reported that average family earnings were lower by roughly $18,000 (currency year not reported) for families of children with ASD than for families of children with no health limitations based on 2002-2008 data from a different survey [27]. That estimate was subsequently incorporated into a broader autism COI study [30]. The authors also reported an annual reduction of $14,755 in maternal earnings [27], which implies an implausibly high hourly wage of $41 if the reduction in earnings was due solely to fewer hours of work.

Informal caregivers may not only forego income from reduced employment but may also pay a penalty in terms of lower hourly earnings. In the Arkansas spina bifida care study, Tilford et al. conservatively assumed that hourly wages were unaffected by care responsibilities [21]. Nonetheless, since on average primary caregivers in the spina bifida sample earned about 25% less per hour, the loss of annual earnings may have been substantially greater than estimated in that study.

* 1. Contingent Valuation

Several studies of informal care valuation have utilized CV [2, 31, 32]. For example, Dutch researchers developed a CV survey to estimate WTP of informal care for two samples of patients and their informal caregivers; one with rheumatoid arthritis (RA) and one with heterogeneous conditions[33] [34]. Among caregivers, mean WTA and WTP for 1 hour of informal care were €7.80 and €9.52 respectively for the RA sample and €8.61 and €10.52 respectively for the diverse sample. A larger differential between WTA and WTP has been reported in a study of caregivers of HIV-infected women in Malawi [35].

CV has also been applied to informal care for dementia and AD. Konig and Weinstein (2002) looked at dementia using CV methods to elicit the preferences of 109 caregivers in Zurich, Switzerland for three hypothetical treatments [36]. Caregivers’ WTP for a reduction of care burden from moderate to low averaged 2,200 Swiss Francs (US$1,500) per year [36]. In another study researchers used dichotomous choice and bidding game methods in four different countries to assess caregivers’ WTP for either a reduction in care burden of AD patients by 1 hour per day or a total elimination of care needs [37]. They found WTP of £105, £121, £59, and £144 per month for a 1-hour reduction in need for care per day, caregivers in the UK, Spain, Sweden, and US, respectively (currency year not reported and there is no indication of the assumed or derived number of days in a month the caregiver worked) [37]. In another study, researchers in France found that WTP for informal care for AD was €12.10 per hour or €298 per month (28 days) (2010 currency) [38]. A Spanish study reported that WTA for informal care for AD ranged by task from €4.5 to €7 per hour, compared with an hourly wage of a domestic worker of €4.7 per hour [39].

Other researchers have focused on methods issues in the application of CV to informal care valuation. For example, de Meijer et al. (2010) conducted a survey of 1,453 informal caregivers and 787 care recipients to investigate feasibility in terms of responsiveness, sensitivity to the individual circumstances of caring, and divergence in WTP and WTA estimates [40]. They found an insignificant difference between WTP and WTA and concluded that feasibility was not an issue, but CV values were sensitive to individual circumstances of caring. In another study, researchers examined informal care preferences (in the form of WTA) of informal caregivers and non-caregivers to determine if it is feasible to elicit the value of informal care from non-caregivers [41]. They found no significant differences between caregivers and non-caregivers, but respondents’ valuations were sensitive to the type of hypothetical care tasks.

* 1. Conjoint Analysis

Although conjoint analysis/DCE methods have become commonly utilized to value health interventions in general, few studies have focused on informal care [42, 43]. In 2005, the same group of Dutch researchers who had administered a CV survey to patients with RA and caregivers [34] also developed and applied a CA survey regarding pairwise choices [44, 45]. Informal caregivers were asked to rate four different hypothetical informal care situations, each with monetary compensation differentiated by care tasks [45]. The researchers found that informal caregivers of patients with RA would require extra compensation of €1 per hour to provide the first hour of care per day, €2 for the second hour, and so on, up to €8 for the 8th hour of care in a day [44]. In a second study, researchers applied a CA survey to a heterogeneous sample of care recipients and caregivers in which caregivers were asked to choose among scenarios in which they would be paid from €9.10 to €13.65 per hour tax-free to provide from 1 to 3 hours per day of care, including heavy housework or personal care, or provide 1 hour per day of light housework without compensation. They found that informal caregivers would require mean compensation of €6.83 per hour for the first hour per day and €12.36 per hour for the next 2 hours per day of care [45].

In a recent study, researchers applied DCE to value three categories of informal care tasks: personal care, supervising, and household tasks [46]. Questionnaires were sent to 1,331 individuals affiliated with caregiver organizations in Scotland, of whom 209 respondents returned complete questionnaires. The design allowed for 48 decisions blocked into six sets of eight choice sets. WTP per hour values ranged from £0.38 to £0.83 for personal care, £0.75 for supervising, and £0.31 to £0.6 for household tasks (currency year not specified).

* 1. Valuing informal care in low- and middle- income countries

Special consideration should be given when valuing caregiver time in low- and middle-income countries (LMIC), where access to the formal health sector is often limited, a sizeable proportion of the population may be employed in the informal sector or may not have constant income, and data on wages by occupation group are often not readily available. In their recommendations for costing Dengue fever cases and outbreaks in Latin America and the Caribbean, Constenia et al. suggested using a caregiver questionnaire to obtain the number of days lost from work due to the illness to estimate productivity loss and country-specific costing to value caregiver time [47]. Several approaches have been employed by researchers conducting economic evaluation in LMIC to value caregiver time. In evaluating the cost-effectiveness of home-based care of tuberculosis patients in Francistown, Botswana, Moalosi et al. used the country’s legal minimum wage for the private and parastatal sector to convert time costs to monetary value [48]. Another study in Botswana used a combination of the caregiver’s self-reported salary (if employed) and the legal minimum wage (if unemployed) [49], an approach that has also been used in Thailand [50, 51]. In Kenya, researchers asked households their perceived monetary value of the time lost due to providing care to a child with diarrhea [52]. All of these methods have their limitations. For example, using the legal minimum wage alone may over-estimate the productivity costs of those working in the informal sector or under-estimate those who do not work in the informal sector. Urban/rural differences may also be overlooked when using minimum wage to value caregiver time. Similarly, the caregiver’s perceived monetary value of time lost may be vulnerable to double-counting, as caregivers may also take into account the care recipients’ preferences and health into the valuation. Researchers should discuss these limitations and their potential effects on analysis results.

References

1. Schaller S, Mauskopf J, Kriza C, Wahlster P, Kolominsky-Rabas PL. The main cost drivers in dementia: a systematic review. Int J Geriatr Psychiatry. 2015 Feb;30(2):111-29.

2. Krol M, Papenburg J, van Exel J. Does including informal care in economic evaluations matter? A systematic review of inclusion and impact of informal care in cost-effectiveness studies. Pharmacoeconomics. 2015 Feb;33(2):123-35.

3. Rice DP, Fox PJ, Max W, Webber PA, Lindeman DA, Hauck WW, et al. The economic burden of Alzheimer's disease care. Health Aff (Millwood). 1993 Summer;12(2):164-76.

4. Neumann P, Hermann R, Kuntz K, Araki S, Duff S, Leon J, et al. Cost-effectiveness of donepezil in the treatment of mild or moderate Alzheimer’s disease. Neurology. 1999;52(6):1138-.

5. Ostbye T, Crosse E. Net economic costs of dementia in Canada. CMAJ. 1994 Nov 15;151(10):1457-64.

6. Van den Berg B, Brouwer WB, Koopmanschap MA. Economic valuation of informal care. Eur J Health Econ. 2004;5(1):36-45.

7. Lamsal R, Zwicker JD. Economic Evaluation of Interventions for Children with Neurodevelopmental Disorders: Opportunities and Challenges. Appl Health Econ Health Policy. 2017 Aug 18.

8. Stabile M, Allin S. The economic costs of childhood disability. Future Child. 2012 Spring;22(1):65-96.

9. Anderson D, Dumont S, Jacobs P, Azzaria L. The personal costs of caring for a child with a disability: a review of the literature. Public Health Rep. 2007;122(1):3-16.

10. Tilford JM, Robbins JM, Hobbs CA. Improving estimates of caregiver time cost and family impact associated with birth defects. Teratology. 2001;64 Suppl 1:S37-41.

11. Wehby GL, Ohsfeldt RL. The impact of having a young child with disabilities on maternal labor supply by race and marital status. J Health Hum Serv Adm. 2007 Winter;30(3):327-51.

12. Loprest P, Davidoff A. How children with special health care needs affect the employment decisions of low-income parents. Matern Child Health J. 2004 Sep;8(3):171-82.

13. DeRigne L. The employment and financial effects on families raising children with special health care needs: an examination of the evidence. J Pediatr Health Care. 2012 Jul-Aug;26(4):283-90.

14. Gordon M, Rosenman L, Cuskelly M. Constrained labour: Maternal employment when children have disabilities. J Appl Res Intellect Disab. 2007;20(3):236-46.

15. Witt WP, Gottlieb CA, Hampton J, Litzelman K. The impact of childhood activity limitations on parental health, mental health, and workdays lost in the United States. Acad Pediatr. 2009 Jul-Aug;9(4):263-9.

16. Okumura MJ, Van Cleave J, Gnanasekaran S, Houtrow A. Understanding factors associated with work loss for families caring for CSHCN. Pediatrics. 2009;124(Supplement 4):S392-S8.

17. Grosse SD. Sociodemographic characteristics of families of children with Down syndrome and the economic impacts of child disability on families. Int Rev Res Mental Retard. 2010;39:257-94.

18. Brehaut JC, Kohen DE, Raina P, Walter SD, Russell DJ, Swinton M, et al. The health of primary caregivers of children with cerebral palsy: how does it compare with that of other Canadian caregivers? Pediatrics. 2004;114(2):e182-e91.

19. Landfeldt E, Lindgren P, Bell CF, Schmitt C, Guglieri M, Straub V, et al. The burden of Duchenne muscular dystrophy: an international, cross-sectional study. Neurology. 2014 Aug 5;83(6):529-36.

20. Kelly AE, Haddix AC, Scanlon KS, Helmick CG, Mulinare J. Cost-effectiveness of strategies to prevent neural tube defects. In: Gold MR, Siegel JE, Russell LB, Weinstein MC, editors. Cost-Effectiveness in Health and Medicine. New York, NY: Oxford University Press; 1996. p. 312–49.

21. Tilford JM, Grosse SD, Goodman AC, Li K. Labor market productivity costs for caregivers of children with spina bifida: a population-based analysis. Med Decis Making. 2009 /;29(1):23-32.

22. Grosse SD, Berry RJ, Mick Tilford J, Kucik JE, Waitzman NJ. Retrospective assessment of cost savings from prevention: folic acid fortification and spina bifida in the U.S. Am J Prev Med. 2016 May;50(5 Suppl 1):S74-80.

23. Grosse SD, Ouyang L, Collins JS, Green D, Dean JH, Stevenson RE. Economic evaluation of a neural tube defect recurrence-prevention program. Am J Prev Med. 2008 12/;35(6):572-7.

24. Jarbrink K, Fombonne E, Knapp M. Measuring the parental, service and cost impacts of children with autistic spectrum disorder: a pilot study. J Autism Dev Disord. 2003 Aug;33(4):395-402.

25. Kogan MD, Strickland BB, Blumberg SJ, Singh GK, Perrin JM, van Dyck PC. A national profile of the health care experiences and family impact of autism spectrum disorder among children in the United States, 2005-2006. Pediatrics. 2008 Dec;122(6):e1149-58.

26. Lavelle TA, Weinstein MC, Newhouse JP, Munir K, Kuhlthau KA, Prosser LA. Economic burden of childhood autism spectrum disorders. Pediatrics. 2014 Mar;133(3):e520-9.

27. Cidav Z, Marcus SC, Mandell DS. Implications of childhood autism for parental employment and earnings. Pediatrics. 2012 Apr;129(4):617-23.

28. Montes G, Halterman JS. Association of childhood autism spectrum disorders and loss of family income. Pediatrics. 2008 Apr;121(4):e821-6.

29. Montes G, Halterman JS. Child care problems and employment among families with preschool-aged children with autism in the United States. Pediatrics. 2008 Jul;122(1):e202-8.

30. Buescher AV, Cidav Z, Knapp M, Mandell DS. Costs of autism spectrum disorders in the United Kingdom and the United States. JAMA Pediatr. 2014 Aug;168(8):721-8.

31. Koopmanschap MA, van Exel NJA, van den Berg B, Brouwer WB. An overview of methods and applications to value informal care in economic evaluations of healthcare. Pharmacoeconomics. 2008;26(4):269-80.

32. Hoefman RJ, van Exel J, Brouwer W. How to include informal care in economic evaluations. Pharmacoeconomics. 2013;31(12):1105-19.

33. Van den Berg B, Bleichrodt H, Eeckhoudt L. The economic value of informal care: a study of informal caregivers' and patients' willingness to pay and willingness to accept for informal care. Health Econ. 2005;14(4):363-76.

34. van den Berg B, Brouwer W, van Exel J, Koopmanschap M. Economic valuation of informal care: the contingent valuation method applied to informal caregiving. Health Econ. 2005 Feb;14(2):169-83.

35. Chiwaula LS, Chirwa GC, Caltado F, Kapito-Tembo A, Hosseinipour MC, van Lettow M, et al. The value of informal care in the context of option B+ in Malawi: a contingent valuation approach. BMC Health Serv Res. 2016;16(1):136.

36. König M, Wettstein A. Caring for relatives with dementia: willingness-to-pay for a reduction in caregiver’s burden. Expert Rev Pharmacoecon Outcomes Res. 2002;2(6):535-47.

37. Gustavsson A, Jönsson L, McShane R, Boada M, Wimo A, Zbrozek AS. Willingness‐to‐pay for reductions in care need: estimating the value of informal care in Alzheimer's disease. Int J Geriatr Psychiatry. 2010;25(6):622-32.

38. Gervès-Pinquié C, Bellanger MM, Ankri J. Willingness to pay for informal care in France: the value of funding support interventions for caregivers. Health Econ Rev. 2014;4(1):34.

39. Pena-Longobardo LM, Oliva-Moreno J. Economic valuation and determinants of informal care to people with Alzheimer's disease. Eur J Health Econ. 2015 Jun;16(5):507-15.

40. de Meijer C, Brouwer W, Koopmanschap M, van den Berg B, van Exel J. The value of informal care–a further investigation of the feasibility of contingent valuation in informal caregivers. Health Econ. 2010;19(7):755-71.

41. Garrido-García S, Sánchez-Martínez F-I, Abellán-Perpiñán J-M, van Exel J. Monetary valuation of informal care based on carers’ and noncarers’ preferences. Value in Health. 2015;18(6):832-40.

42. Clark MD, Determann D, Petrou S, Moro D, de Bekker-Grob EW. Discrete choice experiments in health economics: a review of the literature. Pharmacoeconomics. 2014;32(9):883-902.

43. de Bekker‐Grob EW, Ryan M, Gerard K. Discrete choice experiments in health economics: a review of the literature. Health Econ. 2012;21(2):145-72.

44. van den Berg B, Al M, Brouwer W, van Exel J, Koopmanschap M. Economic valuation of informal care: the conjoint measurement method applied to informal caregiving. Soc Sci Med. 2005;61(6):1342-55.

45. Van Den Berg B, Al M, Van Exel J, Koopmanschap M, Brouwer W. Economic valuation of informal care: conjoint analysis applied in a heterogeneous population of informal caregivers. Value in Health. 2008;11(7):1041-50.

46. Mentzakis E, Ryan M, McNamee P. Using discrete choice experiments to value informal care tasks: exploring preference heterogeneity. Health Econ. 2011;20(8):930-44.

47. Constenla D, Armien B, Arredondo J, Carabali M, Carrasquilla G, Castro R, et al. Costing Dengue Fever Cases and Outbreaks: Recommendations from a Costing Dengue Working Group in the Americas. Value in Health Regional Issues. 2015;8:80-91.

48. Moalosi G, Floyd K, Phatshwane J, Moeti T, Binkin N, Kenyon T. Cost-effectiveness of home-based care versus hospital care for chronically ill tuberculosis patients, Francistown, Botswana. International Journal of Tuberculosis and Lung Disease. 2003;7(9):S80-S5.

49. Ama NO, Seloilwe ES. Estimating the cost of care giving on caregivers for people living with HIV and AIDS in Botswana: a cross-sectional study. J Int AIDS Soc. 2010 Apr 20;13:14.

50. Chatterjee S, Riewpaiboon A, Piyauthakit P, Riewpaiboon W. Cost of informal care for diabetic patients in Thailand. Primary Care Diabetes. 2011;5(2):109-15.

51. Riewpaiboon A, Riewpaiboon W, Ponsoongnern K, Van den Berg B. Economic valuation of informal care in Asia: a case study of care for disabled stroke survivors in Thailand. Soc Sci Med. 2009 Aug;69(4):648-53.

52. Kukla M, McKay N, Rheingans R, Harman J, Schumacher J, Kotloff KL, et al. The effect of costs on Kenyan households' demand for medical care: why time and distance matter. Health Policy Plan. 2017 Dec 1;32(10):1397-406.