

# Self-presentation

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## 1. Introduction

The aim of this self-presentation is to discuss my scientific achievements in the context of my professional career. In the second section, I presented how my scientific and professional career developed. In the third section, I listed a series of monothematic publications that constitute the scientific achievement presented for evaluation and forming the basis for applying for the postdoctoral degree in the field of economic sciences in the discipline of economics; while in the fourth section, I characterized the results obtained in this series. In the fifth section, I discussed my other important scientific publications. Next (in the sixth section), I presented a summary of my scientific achievements. In the seventh section, I collected abbreviations and acronyms used in the self-presentation, while the last section contains references.

My other scientific achievements (including scientific projects and conference presentations) as well as didactic and organizational achievements are described in more details in Appendix 4 attached to the application. The list of published scientific works is additionally presented in a bibliometric analysis conducted by the Library of the SGH Warsaw School of Economics, also attached to the application.

## 2. The course of scientific and professional career

After completing the secondary school, IV L.O. H. Sawicka in Kielce, I studied in the period 1996–2001 at the SGH Warsaw School of Economics (SGH, henceforth) in the field of Quantitative Methods and Information Systems. After the presentation of the MSc thesis "*Rates of return and chaotic dynamics – WGPW*", supervised by prof. dr. hab. T. Szapiro, I graduated, receiving a diploma with honours and MSc degree on 13<sup>th</sup> June, 2001. In the period 1999–2003, I additionally studied at the Faculty of Mathematics, Informatics and Mechanics at the Warsaw

University, where on 24th April, 2003, I obtained BSc degree – the supervisor of my thesis "*Paradoxes in the probability calculus*" was dr. E. Stachowski.

The beginning of my scientific work dates back to the third year of studies at SGH: I participated in the grant by the Scientific Research Committee "*Construction of integrative solutions in hierarchical negotiation problems with noncoherent structures of preferences*" (No. H02D 00510, head: prof. dr. hab. T. Szapiro). In the fourth year at SGH, thanks to the Socrates-Erasmus scholarship, I studied one semester at *Universidade Lusofona de Humanidades e Tecnologias* in Lisbon, Portugal. In the last year of MSc studies, I completed an internship at the Institute of Econometrics, Collegium of Economic Analysis (KAE) at SGH. In 2001, I started my PhD studies at KAE; my tasks, besides participation in scientific research, included being a teaching assistant. During my doctoral studies, my research focused on the issues of supporting and modelling sequential decisions, taking into account the time inconsistency of decisions, in the context of both hyperbolic discounting and multi-criterial evaluation of decision alternatives. In the years 2004–2005, as part of the Marie Curie scholarship, I conducted my PhD research at the *Center for operations research and econometrics* in Louvain-la-Neuve, Belgium. In the years 2006–2013, I worked as an assistant at the Department of Pharmacoeconomics at the Medical University of Warsaw. In 2007, I defended my PhD dissertation at SGH "*On the mechanisms of loss of effectiveness in multi-criteria analysis of sequential decision problems*", whose supervisor was prof. dr. hab. T. Szapiro.

In 2007, I started working as an assistant professor at the Decision Analysis and Support Unit at the Institute of Econometrics at KAE (since the start it has been my main working place, and I continue this work up to the present day). I conduct research on the border of decision theory and its applications in the field of health economics; I give lectures on regular basis in decision analysis and theory in Polish and English, full-time and part-time studies, undergraduate, graduate, and MBA level (I have lectured at four MBA programmes, I also co-organized and co-manage an MBA programme offered by SGH in cooperation with the Medical University of Warsaw), I also gave lectures (as a team member) at doctoral studies. I supervise BSc theses (113) and MSc theses (57), I am the auxiliary supervisor of two PhD candidates.

During my employment at SGH, I did several internships and study visits: at the University of York, York, United Kingdom (2012, one week), University of Glasgow, Glasgow, United Kingdom (2013, one week), Università degli Studi di Milano-Bicocca, Milan, Italy (2013, one week). In 2015–2016, thanks to the Fulbright Senior Award scholarship, I spent six months at the Tippie College of Business at the University of Iowa, Iowa City, Iowa, USA, chosen by me as an opportunity to continue the scientific co-operation established during the Marie Curie scholarship (with prof. R. Amir).

Since obtaining my PhD degree, my scientific interests have focused on methods of supporting and analysing decisions, especially decisions made in the health care market. In particular, my research focused on modelling and estimating societal preferences regarding health states. I characterized the properties of the existing methods and proposed some new, with more favourable properties, methods of decision support in the context of risk and multiple criteria (related to the so-called cost-effectiveness acceptability curves); I used and developed primarily two methods of preference elicitation: the time trade-off method and discrete choice experiments; I also proposed and developed methods for using fuzzy sets to describe preferences for health states—both in axiomatic approach and by proposing empirical methods for eliciting preferences while accounting for the inherent imprecision.

Currently, I continue my research, also as a manager of grants financed by the National Science Center (OPUS10), entitled "*Methods of analysis of decisions in multi-criteria problems and estimation of willingness to pay / accept using fuzzy modelling*") and by the EuroQol

Group scientific association (“A fuzzy approach to time trade-off experiment in EQ-5D-3L valuation” and “Building values sets based on TTO results by averaging model predictions and actually observed means”) and as part of international cooperation as a scientific partner in the POLONEZ project (“Evidence use in the allocation of health care budgets: A comparative study of drug reimbursement in Poland, Canada, and Germany”, manager: Dr. D. Wranik, home university: Dalhousie University, Canada).

### 3. The monothematic series of publications

The result of my research conducted after receiving the doctoral degree is a series of the following publications related thematically, under the collective title "**Modelling preferences and supporting the decisions in the health care market from the economic perspective**"<sup>1</sup>:

1. **M. Jakubczyk** (2009): „**Impact of complementarity and heterogeneity on health related utility of life**”, *Central European Journal of Economic Modelling and Econometrics*, 1, 139–156;
2. **M. Jakubczyk**, B. Kamiński (2010): “**Cost-effectiveness acceptability curves – caveats quantified**”, *Health Economics*, 19 (8), 955–963, **IF = 1.946, MNiSW = 32**;
3. D. Golicki, **M. Jakubczyk**, M. Niewada, W. Wrona, J.J.V. Busschbach (2010): "**Valuation of EQ-5D Health States in Poland: First TTO-based Social Value Set in Central and Eastern Europe**", *Value in Health*, 13 (2), 289–297, **IF = 2.342, MNiSW = 32**;
4. **M. Jakubczyk** (2015a): "**Using a Fuzzy Approach in Multi-Criteria Decision Making with Multiple Alternatives in Health Care**", *Multiple Criteria Decision Making*, 10, 65–81, **MNiSW = 12**;
5. **M. Jakubczyk** (2015b): „**Rekurencyjne i konserwatywne reguły decyzyjne przy wyborze technologii medycznych w warunkach ryzyka**” in: D. Kopańska-Bródka (Ed.) „*Studia ekonomiczne. Zeszyty Naukowe Uniwersytetu Ekonomicznego w Katowicach*”, Wydawnictwo Uniwersytetu Ekonomicznego w Katowicach, ISSN 2083-8611, 235, 85–99, **MNiSW = 10**;
6. **M. Jakubczyk**, D. Golicki, M. Niewada (2016): "**The impact of a belief in life after death on health-state preferences: True difference or artifact?**", *Quality of Life Research*, 25 (12), 2997–3008, **IF = 2.344, MNiSW = 35**;
7. **M. Jakubczyk**, B. Kamiński (2017): “**Fuzzy approach to decision analysis with multiple criteria and uncertainty in health technology assessment**”, *Annals of Operations Research*, 251 (1–2), 301–324, **IF = 1.709, MNiSW = 30**;
8. **M. Jakubczyk**, D. Golicki (2018): “**Estimating the Fuzzy Trade-Offs Between Health Dimensions with Standard Time Trade-Off Data**” in: J. Kacprzyk, E. Szmidt, S. Zadrożny, K. Atanassov, M. Krawczak (Eds.) “*Advances in Fuzzy Logic and Technology 2017. IWIFSGN 2017, EUSFLAT 2017.*”, *Advances in Intelligent Systems and Computing*, vol 642. Springer, ISSN: 2194-5357; ISBN: 978-3-319-66823-9, DOI: 10.1007/978-3-319-66824-6, 266–277, **MNiSW = 5**;

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<sup>1</sup> Some of the research projects were carried out with co-authors due to the interdisciplinary nature of the research. My original contribution consisted in the application of economic methods to the study of preferences in the area of health and did not include purely medical issues. The impact factor (IF) and Ministry of Science and Higher Education (MNiSW) scoring was assigned according to the results of the bibliometric analysis attached to the application.

9. **M. Jakubczyk**, B. Kamiński, M. Lewandowski (2018): “**Eliciting Fuzzy Preferences Towards Health States with Discrete Choice Experiments**”, in: C. Berger-Vachon, A. Gil Lafuente, J. Kacprzyk, Y. Kondratenko, J. Merigó, C. Morabito (Eds.) “Complex Systems: Solutions and Challenges in Economics, Management and Engineering”, *Studies in Systems, Decision and Control*, vol 125. Springer, ISBN 978-3-319-69988-2, DOI: 10.1007/978-3-319-69989-9, 131–148, **MNiSW = 5**;

I designate the above series of publications as a scientific achievement in the meaning of the Act of 14<sup>th</sup> March, 2003, on academic degrees and academic title, and on degrees and title in the field of art (Article 16, paragraphs 1 and 2), which constitutes a significant contribution to the development of the discipline economy. The papers in the series concern the following sequentially-related and complementary research problems:

- defining the societal preferences in Poland for health states (accounting for the quality and longevity of life) in the context of the complexity of health as an economic good and demographic and cultural factors;
- characterization of the existing methods and construction of new methods of supporting the decisions by the public regulator regarding the selection of health technologies in the healthcare market from the economic perspective, while using the measures of effectiveness based on the societal preferences and accounting for uncertainty;
- accounting for the lack of precision while modelling the societal preferences for health states using fuzzy sets theory.

I used the above thematic division in the summary of the main results of the studies in the series. I presented a detailed description of my own contribution to the co-authored papers in Appendix 4 (in agreement with the relevant co-authors’ statements, also attached to the application). Many of the works are interdisciplinary and required referring the obtained results to the area of health care and health-related quality of life, hence the cooperation with co-authors from the Medical University of Warsaw (D. Golicki and M. Niewada). In particular, in Jakubczyk, Golicki, and Niewada (2016) study, it was my contribution to formulate the idea for research, to develop the research plan, to design and carry out the econometric and statistical analysis, and to discuss the results obtained in the context of preference estimation methods. In Golicki et al. (2010) study, it was mainly my contribution to perform the econometric analysis, and it was entirely my contribution to do the statistical analysis of the consequences of increasing the number of experiments per one respondent. In Jakubczyk and Golicki (2018) study, it was entirely my contribution to formulate the research idea, to construct the model of preferences presented in the study, and to develop and implement methods for estimating these preferences, as well as to perform the calculations. Similarly, it was entirely my contribution to develop the model of preferences used in Jakubczyk, Kamiński, and Lewandowski (2018) study.

## **4. Discussion of the publications forming the achievement**

### **4.1. Introduction to the area of research**

Health can be regarded as the subject of economic research in accordance with the standard definitions of economics: as the discipline interested in *how the economic society decides what and for whom to produce* (Begg, Fischer, and Dornbusch, 1999) or as *research about social laws governing the production and distribution of wealth in society, research about rational*

*management* (Dictionary of the Polish language). Health can be treated as a consumption good (being healthy is desirable) or a capital good (being healthy enables working, health depreciates over time, and investments in health can be made; see Grossman, 1972). Improving the health level positively impacts on the functioning of the entire economy (see Suhrcke et al., 2006, and Figueras and McKee, 2012).

From the market point of view, it is the health services (e.g. medical consultation) or medical technologies (e.g. medicines), rather than health itself, that is the subject of transactions. The healthcare market constitutes a significant part of the whole economy in developed countries: in Poland, in 2015, the current expenditures on healthcare amounted to 6.34% of Gross Domestic Product (GDP), with public expenditures accounted for 4.44% of GDP (data from the Central Statistical Office, GUS). In many countries, the share of spending on healthcare in GDP is higher, for example 6.6% in Lithuania, 7.4% in the Czech Republic and Hungary, 8.1% in Slovakia, 9.1% in the United Kingdom, 11.3% in Germany, and as much as 17.1% in the United States (World Bank data from 2014).

The healthcare market has many distinctive features that are the subject of economic research, see Arrow (1963). For example, the asymmetry of information between consumers and suppliers (doctors) is an inherent element of providing health services—medical knowledge requires many years of study and practice. Because the treatment effects are often delayed and subject to stochastic uncertainty<sup>2</sup>, the consumers of medical services are typically unable to verify the quality of the services. Hence, the trust and ethical requirements (e.g. the Hippocratic oath) become of great importance. The demand for health services is stochastic (contrary to, for example, demand for food) and is distributed unevenly between individuals. The need to use medical services on average increases with age and usually concentrates in only a part of the lifetime: for example, about 31% of expenses are incurred in middle age (40–64 years of age), and 48.6% after being 65 (Alemayehu and Warner, 2004); as much as 25% of expenditures are incurred in the last year of life (see Hogan et al., 2001). Due to this concentration, health insurance is an important element of the health care market, allowing for spreading risk both over lifetime and between individuals.

The provision of health services is affected not only by purely market regulations, such as prices; health care is often perceived as an irrevocable right of citizens. For example, the Article 68 of the Constitution of the Republic of Poland states: *everyone has the right to health protection*. This approach to health care is also present in the philosophical systems of social policies. In Sen's (1985) capabilities theory, the assessment of the well-being of the society and policy directions depends on how it enables the citizens to strive for desirable functionings in their life. Nussbaum (2013) derives from that that providing health care is an obligation of the modern state. As a consequence, health (in the form of life expectancy) was included as one of the elements of the society development level indicator, HDI (human development index).

Due to the above justifications, in developed countries, health care is largely regulated by the state and largely financed from public funds. The share of public expenditure on health care in total expenditure amounts to 71% in Poland, and although only to 48.3% in the United States, it again amounts to 66% in Hungary, 67.9% in Lithuania, 72.5% in Slovakia and as much as 77% in Germany, 83.1% in the United Kingdom, and 84.5% in the Czech Republic (World Bank data from 2014). One of the areas of regulation is to determine which medical technologies should be financed by the state. Limited financial resources force such choices, taking into account social preferences expressed through medical and economic criteria, and

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<sup>2</sup> There is a first-order uncertainty: for example, a priori we can only expect a patient to be cured with some probability, see Briggs *et al.* (2012).

reflected in the so-called cost-effectiveness analysis<sup>3</sup> (CEA) or cost-utility analysis<sup>4</sup> (CUA). Developing methods of such analyses requires first the construction of individual and group models of preferences of decision-makers. Importantly, especially from an economic point of view, the analysed medical technologies compete for financial resources from a single public budget, and at the same time generate various types of effects—for example, they may prolong life or improve the health-related quality of life (HRQoL); hence, it is necessary to bring these different effects into a single measure that allows comparisons.

Typically, the attempt to maximize social well-being (the *welfarist approach*) is abandoned due to, for example, practical difficulties and ethical doubts (should health care differ between individuals due to estimated differences in their preferences?). Still, in a pragmatic *decision-maker's approach* (see Brouwer and Koopmanschap, 2000), the measure of health is defined by reference to the preferences of individual members of society for health (the quality and longevity of life), so that the decisions are made based on a measure accounting for society's needs as expressed by preferences. The so-called QALY (quality-adjusted life years) theoretical model is typically used for that purpose; the model's axiomatic foundations were presented by Pliskin, Shepard, and Weinstein (1980), Bleichrodt, Wakker, and Johannesson (1997), and Miyamoto *et al.* (1998). Each health state,  $Q$ , has an assigned number  $u(Q)$  interpreted as the utility of spending one year in this state. The utility is treated as the von Neumann-Morgenstern (1947) utility, i.e. its expected value is maximized in the decision problems under risk. The QALY utilities are normalized (death is assigned the utility 0, and the full health is assigned the utility 1).

The above considerations have an important practical value in many countries, including Poland. The reimbursement act (2011) and accompanying legal acts, in the context of decisions on the reimbursement of drugs, require the health benefits to be expressed within the QALY model and the determination the cost-effectiveness for effects measured in QALYs. When estimating the benefits, additionally, the issue of uncertainty arises as the estimates are based on data from clinical trials, i.e. random samples (these issues are discussed in more detail below).

In the such defined area, I conducted research that aimed at:

- determining the Polish societal preferences for health states (accounting for the quality and longevity of life) in the context of health being a complex economic good as well as demographic and cultural factors;
- characterizing the properties of the existing methods and constructing new methods to support the decisions by the public regulator on the selection of health technologies, from the economic perspective, accounting for uncertainty and when expressing the treatment effectiveness with measures based on societal preferences;
- accounting for imprecision when constructing the models of societal preferences for health, using the fuzzy sets theory.

The results obtained in the above three sub-areas were discussed in sections 4.2, 4.3, and 4.4, respectively.

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<sup>3</sup> In the Polish version of this self-presentation, the corresponding footnote discusses how the term *cost-effectiveness* is and ought to be translated into Polish. Obviously, it is irrelevant in the present version (but was added to preserve the correspondence of footnote numbering between the versions).

<sup>4</sup> This term is sometimes used when the unit of effect can be treated as a utility measure.

## 4.2. Determining the societal preferences for the health states in Poland

The first study in the area of measurement of health preferences in which I participated was **Golicki et al. (2010)**. The study aimed to determine the societal preferences for health states in Poland and to assign to individual health states the population-average utility values under the QALY model, i.e. to construct a so-called *value set*. In addition, an innovative result of the study was the assessment of the consequences of increasing the number of thought experiments used per one respondent in preference elicitation studies.

In the study, we used EQ-5D-3L descriptive system to define and describe HRQoL. The system was developed by the EuroQol Group (see Brooks and De Charro, 1996, Brazier et al., 2017): a health state is described by five dimensions (hence, 5D): mobility (MO), self-care (SC), the ability to perform usual activities (UA), pain/discomfort (PD), and anxiety/depression (AD). Each dimension is valued at one of three levels (hence, 3L): 1 denoting no problems (e.g. with mobility), and 3 denoting extreme problems (e.g. being confined to bed). The EQ-5D-3L questionnaire can be used in various diseases (it is a generic questionnaire) and is the preferred method of measuring HRQoL in applications assessed by the National Institute for Clinical Excellence in England.

The study included 321 respondents with a gender and age structure representative for the adult population in Poland. In the literature, it is mostly suggested that respondents in such studies should recruit from the general population; firstly, because the general population bears the cost of health insurance, and, secondly, for pragmatic reasons (it is difficult to find enough respondents in some rare health states), see Whitehead and Ali (2010) and Drummond et al. (2009). In our study, the respondents were recruited from visitors in eight hospitals in Poland. Each respondent in the main part of the experiment assessed the utility of 23 health states (selected based on Lamers et al., 2006, cf. Table 1 in the original publication). The study used the time trade-off<sup>5</sup> (TTO) method to elicit utility, see Attema et al. (2013). In TTO method, for a given state  $Q$ , the respondent in a series of questions was asked to choose between two options: 10 years in  $Q$  (followed by death) and  $T$  years in full health (followed by death). Within the QALY model, the indifference indicates that  $u(Q) = T / 10$ , for normalized utility:  $u(\text{full health}) = 1$  and  $u(\text{death}) = 0$ . In our study, a modified TTO method was used for  $Q$  states assessed as worse than dead (for which immediate death was preferred as compared to 10 years in  $Q$ ).

After data cleaning, there were 6777 observations obtained from 305 respondents included in the analysis. The utility values elicited were then extrapolated to the complete set of 243 health states using econometric modelling in the panel random effects approach (see Dolan et al., 1996)<sup>6</sup>. The dummy variables were assigned to the dimension-level combinations to determine the additive disutility of the deterioration of individual dimensions, see Table 5 in the original publication.

An innovative element of the study was a significant increase in the number of health states assessed by one respondent (up to 23). Previously, typically 13 states were used (see Shaw, Johnson and Coons, 2005, Dolan et al., 1996, Badia et al., 2001, Greiner et al. 2005) or

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<sup>5</sup> A comment on the Polish translation was provided in the corresponding footnote of the Polish version of the self-presentation.

<sup>6</sup> Modelowania tego nie trzeba przeprowadzać w tzw. badaniach nasyconych (ang. *saturation studies*), w których wykorzystuje się wszystkie stany zdrowia (por. np. Santos et al., 2016). Wadą badań nasyconych jest duży koszt badania (są 243 stany zdrowia w systemie EQ-5D-3L) oraz trudność wyobrażenia sobie przez respondenta niektórych stanów zdrowia (np. bycie przykutym do łóżka i odczuwanie krańcowego bólu bez żadnego przygnębienia).

even as few as 7 states (cf. Jelsma et al., 2003); occasionally the number was increased to 16 or 17 (see Wittrup-Jensen et al., 2002, Tsuchiya et al., 2002, Lamers et al., 2006). The influence of the extension of the study on the reliability of the obtained results was verified by formally testing two statistical hypotheses: (H1) it does not impact the average utility value assigned to the health state whether the state is evaluated in the middle (in positions 6–17) or in the final part of the experiment (in positions 18–23); (H2) the variance of utility values does not differ for evaluations done in positions 6–17 and in positions 18–23. Testing the H1 hypothesis was aimed at determining whether the extension of the TTO experiment may introduce a bias on the results (e.g. respondents getting fatigued during the experiment assign higher utility values, wanting to avoid a more complicated version of TTO for conditions worse than dead). Testing the H2 hypothesis was aimed at determining whether extending the experiment may reduce the precision of the estimators, e.g. by increasing the dispersion of responses given due to the reduced concentration. In the analysis, the results from positions 1–6 were omitted due to the possibility of a learning curve for the decision maker. Due to multiple hypotheses testing (44 health states) and drawing a joint conclusion for the whole family of hypotheses (that the extension of the experiment harms the credibility of the results), the Hölm-Bonferroni correction was used in the testing. Eventually, none of the two hypotheses was rejected, demonstrating the possibility of extending the experiment and confirming the credibility of the obtained results (see Table 7 in the original publication).

The work presented here was the first study in the EQ-5D-3L system in Central and Eastern Europe using a choice-based method to elicit health state utilities.<sup>7</sup> The comparison of the obtained utility values with other countries (Germany, United Kingdom and Slovenia) showed differences in the structure of preferences (e.g. higher relative importance of the PD dimension in Poland, see Table 8 and Figure 1 in the original publication), and thus the legitimacy of applying Polish values when making decisions by a public regulator in Poland to reflect the country's specific preferential structure. The developed value set (see Table 6 in the original publication), i.e. the assignment of utility values to health states, is officially recommended in economic analyses by guidelines of the Agency for Health Technology Assessment (2009) and newer guidelines of the Agency for Health Technology Assessment and Tariff System (AOTMiT, 2016).<sup>8</sup>

I continued the research discussed above in the study **Jakubczyk (2009)**<sup>9</sup>, which aimed, firstly, to understand the heterogeneity of societal preferences, i.e. to learn the demographic factors affecting these preferences, and secondly, to understand the mechanisms that shape the preferential structure of individual respondents—to determine the interaction between attributes characterizing the health condition (EQ-5D dimensions) and affecting the utility, i.e. to understand the economic character of interdependence (whether the attributes are complements or substitutes). The inspiration for undertaking such research was derived

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<sup>7</sup> In an earlier study in Slovenia (Prevolnik Rupel and Rebojl, 2000), the visual analogue scale (VAS) method was used, which is not based on theoretical foundations, e.g. such as the QALY model.

<sup>8</sup> I still continue research in the presented context; currently I participate in a project aiming at constructing a Polish value set using the EQ-5D-5L descriptive system. The preliminary results were presented at the international conferences: D. Golicki, M. Jakubczyk, M. Niewada, K. Graczyk (2017): a poster „Polish Tariff for the EQ-5D-5L”, 34th EuroQol Plenary Meeting, Barcelona, Spain, and D. Golicki, M. Niewada, K. Graczyk (2017): a poster “The First EQ-5D-5L Value Set in Central and Eastern Europe”, International Society for Pharmacoeconomics and Outcomes Research Annual European Congress, Glasgow, Great Britain, and at the national conferences: M. Jakubczyk (2017): a poster „Modelowanie użyteczności stanu zdrowia — aspekty ekonometryczne (czyli piki, triki i wyniki)”, XV Conference of the Polish Pharmacoeconomic Society, Warszawa, Poland.

<sup>9</sup> The earlier data resulted from the publishing process for the study Golicki *et al.* (2010), which was available online in 2009.



from contemporary demographic changes and growing awareness of the significance of these changes for the managing of the economy.

In the study, I used data from the Golicki et al. (2010), however, I used a different data cleaning procedure (eventually, 6,601 individual utility assessments obtained for 295 respondents formed the dataset). I built four linear regression models, denoted M1–M4. In M1, only dimension-specific variables were used (i.e. variables  $d_{i,j}$  indicating whether dimension  $i$  is at level  $j$ ,  $i = 1, \dots, 5$ ,  $j = 1, 2, 3$ ), and neither demographic characteristics nor possible interactions between domains were taken into account. Model M1 was considered a reference point for the other models. In M2, I additionally included variables describing interactions between dimensions in their impact on utility: variables  $N2$ ,  $N3$ ,  $I2$ ,  $I3$ , and  $D1$  (and squares of the last three) proposed in the literature (see Tsuchiya et al., 2002, Greiner et al., 2004, Lamers et al., 2006), as well as the cross-product of the pairs of variables  $d_{i,3}$ . The inclusion of the latter group was an innovative element of the study and aimed to directly determine the interaction between specific aspects of HRQoL (due to the moderate sample size, the study was limited to interaction at the third level of dimension values). In M3, I included demographic variables (e.g. sex, age, or own health)—both individually and in cross-products with variables  $d_{i,2}$  and  $d_{i,3}$ . Models M1–M3 were estimated as using panel random effects approach. The statistically insignificant variables were removed (using the significance level of 0.05), and the joint significance of the variables was tested.

The M4 model was specified as a random parameters model (see e.g. Maddala, 2006), i.e. it was assumed that the impact  $d_{i,j}$  on utility differs between individuals and is a random variable drawn from a population-specific probability distribution (normal distribution was assumed); the expected value of this distribution is subject to estimation. The M4 model was estimated in the Bayesian approach using the MCMC method (the Bayesian approach was tested already in the Golicki et al., 2010, but in the constant-parameters specification).

The most important results of the Jakubczyk (2009) are as follows. Model M1 confirmed the results of study Golicki et al. (2010), that PD dimension has the greatest impact (at level 3) on the utility of health in the Polish society, while the AD dimensions has the smallest impact (the deterioration of this dimension at level 2 did not have a statistically significant impact on utility).

The results of M2 (Table 5 in the original publication) demonstrated interdependencies between the PD and all other dimensions in the impact on health state utility. The negative values of coefficients (the dependent variable was defined as the disutility, i.e. the loss of utility, 1 minus the utility of a given state, where 1 is the normalized utility of full health) indicate the complementarity of dimensions—the deterioration of one domain has a less negative impact on utility when another dimension has been already deteriorated. One of the possible interpretations is that the respondents imagine (or perhaps know from experience) that feeling pain significantly hinders functioning in life, despite the, for example, lack of limitations in mobility.

The results of M3 (Table 6 in the original publication) demonstrated, firstly, that demographic variables affect the perceived utility mainly through interactions with variables  $d_{i,3}$ , i.e. by shaping the impact of dimension deterioration on level 3. The most important conclusions are: men react more intensely to (the worsening of) SC dimension and less intensely to UA and PD dimensions; educated people attach greater importance to SC and PD dimensions; people who themselves experience problems in the MO dimension assign a lower disutility to the deterioration of this dimension, and the opposite effect is observed for the AD dimension (which may explain the low impact of this dimension in M1—respondents from the general population, especially due to the sample selection method in Golicki et al., 2010, underestimate the actual impact of psychological problems).

A novel and unexpected result of Jakubczyk (2009) was to show the influence of faith in life after death (variable *faith* in Table 6 in the original publication)—a proxy for religiosity—on perceived utility. The variable *faith* in the final specification occurred in the largest number of interactions between demographic variables, and one of these interactions was characterized by the largest (absolute value) effect on utility (interaction with the variable  $d_{1,3}$  describing the deterioration of the UA dimension). The religious (more precisely – those who believe in life after death) attributed a smaller loss of utility to health states, especially to states in which the possibility of performing daily activities is deteriorated. An important conclusion from the M3 model is that the preference studies should ensure the representativeness of the sample also with respect to the above-mentioned demographic variables.

Finally, the results of the M4 model indicated (Table 6 in the original publication) that the respondents differ significantly between themselves in the perceived importance of individual dimensions. Using the random parameters approach was an innovative element of the study (I am aware of no earlier publications using this approach to estimate the utility of EQ-5D-3L health states); while currently, random parameter models have become the standard approach to specifying the model (e.g. Rowen et al., 2014, Shiroiwa et al., 2016, Feng et al., 2017).

The results obtained in Jakubczyk (2009) created a motivation to conduct a more detailed analysis of relationships between religiosity and preferences for health states. The purpose of **Jakubczyk, Golicki, and Niewada (2016)** study research was to determine the influence of religiosity on the utility of health states as elicited in TTO experiments and on the final utility value set, as well as to discuss the consequences for the CEA analyses and possible modification of preferences elicitation methods (to what extent should the utility differences be treated as an inherent element of preferences, and to what extent are they the effect of a particular elicitation method).

The data from Golicki et al. (2010) were used. Religiosity was defined based on the faith-in-life-after-death variable on three levels: strong religiosity (*I strongly believe*), moderate religiosity (*I strongly believe* or *I tend to believe*) and lack of religiosity (*probably not, definitely not* and *I do not know*). The study took into account demographic variables to account for the possible influence of another factor (confounding) at the same time affecting religiosity and utility (e.g. age or education).

The most important results obtained in the study are as follows. The religious are more often unwilling to give up any lifetime in the TTO experiment in exchange for improving HRQoL (hence, are *non-traders*): the odds ratio is about 2 for strongly religious people (in comparison to the remaining ones, i.e. moderately religious and non-religious) and about 1.5 for the moderately/strongly religious (as compared to the non-religious). Such results were obtained for a univariate analysis and for multivariate analysis with the Cochran-Mantel-Haenszel test and for a multivariate logistic regression allowing to eliminate the influence of other variables. The religious (both strongly and moderately) less often consider a state worse than dead. The study therefore confirmed the influence of religiosity on the results of the TTO experiment.

The influence of religiosity is manifested mostly in the willingness to trade life duration in the TTO experiment (being a trader), and to a lesser degree in the specific extent of the traded time (among those who decided to give at least part of their time in the first place), see Table 4 in the original publication. Our interpretation of the obtained results is that religious people more often do not accept the thought experiment used in the TTO method altogether (a decision whether to choose a better but shorter life), due to (perhaps subconsciously) deeming such decisions to be the sole domain of some supernatural being, e.g. God in the Christian faith.

Such an interpretation is validated by the qualitative results from other TTO studies, see Au Eong et al. (2012) and Wee et al. (2008) and links between religiosity and the results of TTO and VAS values reported in other studies (see Augestad et al., 2013, and personal communication with K. Rand-Hendriksen). For this reason, the conclusion of the present study was to remove the influence of religiosity on the utility estimates.<sup>10</sup>

On average, the strongly religious individuals assign utility greater by 0.136 (in the additive model) or the disutility lower by a factor of 2.1 (in the multiplicative model). The study constructed a corrected utility value set (see Table 3 in the original publication); the average difference in value was 0.046.

An innovative element of the study was a systematic examination of the influence of religiosity on utility using a variety of analytical techniques (in order to eliminate e.g. the confounding) in a large random sample. Additionally, in the study a modified TTO was presented that would allow examining whether the effect observed in the data is caused by the actual difference in preferences or is the artefactual consequence of using the standard TTO as the elicitation method (illustration in Figure 3 and discussion in the text in the original publication).

### **4.3. Methods of supporting the decisions by the public regulator**

Health preference studies are conducted not only for purely theoretical reasons. As presented in the introduction, the motivation comes also from the need to use the estimates to support the decision on the selection of medical technologies to be financed. The intention is to juxtapose the clinical benefits of using a given medical technology with the costs of the technology (see e.g. Drummond et al., 2015).

As mentioned above, there is first-order uncertainty present in the treatment process (i.e. the outcome is uncertain in a specific case). In economic analyses, this uncertainty is usually omitted, since QALY, as a von Neumann-Morgenstern based utility, incorporates the risk attitude of decision-makers, while the cost of treatment is spread out across society, resulting in risk-neutrality due to the Arrow-Lind's theorem (1970). In economic analyses, therefore, it is important to estimate the expected value of benefit (in QALY) and cost. Public decisions often use the ratio of benefits (additional expected number of QALY when using the considered alternative instead of the current one) to the cost—the so-called *incremental cost-effectiveness ratio* (ICER). The literature defines when such an approach is applicable, see Weinstein and Zeckhauser (1973) and Laska et al. (1999). The use of the ICER coefficient was indicated as one of the criteria in Polish legislation (the Reimbursement Act, Article 12, paragraph 13., and the Regulation of the Minister of Health on minimum requirements, paragraph 5, paragraph 2, point 2.).

The estimates of the expected values are derived from random samples (e.g. randomized controlled trials); hence, the obtained estimators are subject to stochastic error (second order uncertainty, see Briggs et al., 2012). In addition, an important factor is that the *willingness to pay* (WTP) for clinical effects, i.e. the monetary valuation of QALY, is typically not given precisely (attempts were made to determine the threshold based on the observed decisions, see

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<sup>10</sup> Such an approach is going to be most likely used in the ongoing EQ-5D-5L elicitation study, as presented during a conference: M. Jakubczyk (2017), „Modelowanie użyteczności stanu zdrowia — aspekty ekonometryczne (czyli piki, triki i wyniki)”, XV Conference of the Polish Pharmacoeconomic Society, Warszawa, Poland.

Devlin and Parkin, 2004, or Niewada et al., 2013)<sup>11</sup>. One of the methods of assessing the uncertainty associated with the available decision options (medical technologies) most frequently used in public decisions are the cost-effectiveness acceptability curves (CEAC), see van Hout et al. (1994), Löthgren and Zethraeus (2000), and Fenwick et al. (2004). The CEACs show (in the Bayesian interpretation) the probability that a given alternative is optimal (with respect to the cost and effect criteria) depending on the WTP values, see Briggs (1999). The CEAC curves are recommended for use in Poland (AOTMiT guidelines, 2016).

Several counter-intuitive properties of CEACs were indicated in the literature. Fenwick et al. (2001) showed that when choosing between two alternatives for a given WTP value, the choice based on the probability of maximizing the net benefit (NB) and the choice based on the expected value of NB are equivalent only if the uncertainty characterizing both alternatives (effects and costs) is given a symmetrical distribution. Fenwick et al. (2004) provided an example of non-monotonic CEACs when comparing two alternatives (violating the intuition that the increases in WTP should consistently favour one of the alternatives). Barton et al. (2008) noticed that in case of choosing from more than two alternatives, also for symmetric probability distributions defining the uncertainty of estimation, maximizing the expected NB value for a certain WTP level may not coincide with maximizing the probability of maximizing the NB for any WTP level.

The aim of **Jakubczyk and Kamiński (2010)** study, located in the stream of literature discussed above, was to characterize the undesirable properties of the CEAC method in different classes of decision problems (in the study the choices between two technologies and among  $n > 2$  technologies were considered; with uncertainty characterized by normal distribution or an undefined classes of distributions, with possible correlation between the uncertainty of estimates for costs and effects). The study was a theoretical one. The local (i.e. for a given WTP level) and global (i.e. the shape of CEAC for different WTP) properties of the CEACs were considered.

When choosing from  $n = 2$  alternatives, we demonstrated the bounds on the possible discrepancy between the options' attractiveness calculated as standardized (divided by standard deviation) expected NB difference and probability of NB maximization by the alternative under consideration (page 957 in the original publication). The upper bound for the expected value of perfect information is also indicated when the decision-maker makes a selection based on the maximization of the probability of maximizing NB.

The study pointed out the consequences and limitations of CEAC when comparing the alternatives in the case of first-order stochastic dominance (FOSD). In the situation of choosing between two options, FOSD guarantees compliance of approaches based on maximization of the expected value of NB and probability of maximizing NB. In the case of many alternatives (when the estimation errors of their attributes are independent) the CEAC curve for the FOSD-dominated alternative must lie below (i.e. indicate lower attractiveness for the decision maker) the CEAC for the dominant option (no proof of this claim was provided in the study, I later completed this gap in Jakubczyk, 2015b, study described below).

It was indicated that for  $n > 2$  alternatives, pairwise comparisons with CEACs (i.e., comparing options in pairs with respect to the probability that one offers a larger NB) may generate a cyclical relationship of strict preferences; additionally, we presented the upper bound on the lowest probability in such a cycle (75% in general, i.e. for any number of alternatives).

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<sup>11</sup> This observation constituted a major motivation for the studies Jakubczyk and Kamiński (2017) and Jakubczyk (2015a) included in the monothematic series and presented below.

Another innovative result obtained in the study was to show that the use of CEAC as a decision rule (i.e. for a given WTP level, the selection of the option that maximizes the probability of offering the largest NB) violates the  $\alpha$  property of the choice functions: adding a new element to menu of alternatives may result in a previously-available-but-not-chosen element be selected (the property of independence of irrelevant alternatives is not satisfied). A similar result was previously shown by Sadatsafavi et al. (2008), but they discussed this property only in the context of a static set of alternatives (i.e. not in the context of choice function properties) and only in the case of correlated characteristics of the alternatives, while in Jakubczyk and Kamiński (2010) it was shown that the variance of NB estimates is a crucial element (p. 958 according to the numbering of the original publication).

Finally, in the discussed study we generalized the results presented by Fenwick et al. (2004), showing the influence of the parameters describing the alternatives (in the case of  $n = 2$  alternatives) on the monotonicity of CEAC (see Chapter 3 in the publication, in particular Table II). In particular, it was shown that for symmetric distributions describing the uncertainty of estimations, the non-monotonicity of CEAC is constrained: the CEAC can cross the 50% line (the natural threshold when making decisions for  $n = 2$  alternatives) at maximum once. At the same time, it was shown that in the case of skewed distributions, this property does not hold, i.e. it is possible for CEAC to cross the 50%-line multiple times. A summary of the most important results of the Jakubczyk and Kamiński study (2010) is presented in Table III in the original publication.

I continued the research described above in **Jakubczyk (2015b)**. The aim was to propose CEAC modification as a decision rule, in view of the undesirable properties of the standard CEACs as discussed above. The study considered the case of symmetrical probability distributions describing the uncertainty of estimates of the compared characteristics of more than two alternatives. In Jakubczyk (2015b), I presented the proof of a proposition phrased but not proved in Jakubczyk and Kamiński (2010) on the relationship between CEACs in case of FOSD (Statement 1 in Jakubczyk, 2015b). A new, significant result of the study was the description of the consequences of correlation between the uncertainty of estimates of alternatives' characteristics (Table 2 in the original publication).

In the study, I proposed two new choice functions to choose from among any number of alternatives (but finitely many) based on CEAC: a recursive and conservative decision making rule (Definitions 1 and 2 in the original publication, respectively). The idea is to compare each alternative using CEAC only with one other alternatives: respectively, selected recursively from the remaining alternatives or providing the least favourable result. I carried out the analysis of the properties of the proposed methods for a fixed value of WTP (i.e., I studied the local properties of CEAC, according to the terminology introduced above). I showed (Theorem 2 in the original publication) that both the proposed methods have desirable and intuitive properties in the case of uncertainty of the description of the alternatives with a multidimensional (also correlated) normal distribution: they are consistent with the criterion based on maximizing the expected value of NB, they satisfy the  $\alpha$  property of choice functions (in particular they are robust to adding a clone of a decision alternative to the menu), they are robust to the changing of the correlation structure of uncertainty, and they comply with the FOSD criterion.

I showed the compliance of the conservative rule with FOSD in the case of independent random variables characterizing the alternatives being compared (Theorem 3 in the original publication). I proved that in case of pair-wise CEAC comparisons forming a transitive relation, both the proposed choice functions indicate the maximal element of this relation (Theorem 4 in the original publication), i.e. the multitude of alternatives does not in itself lead to the internal incoherence of the choice function. In the study, I also conducted an empirical case study,

illustrating both methods introduced based on the data set used by Barton et al. (2008), following Goeree et al. (2002).

I continued research on how to use CEACs in the context of the difficulty with determining the exact WTP value. The starting point was the observation that the estimates of the *value of statistical life* presented in the literature are very diverse, see Bellavance et al., (2009). The difficulty in determining WTP in the context of health results from the lack of market experience, which results, for example, in an extremely large discrepancy between WTP and *willingness-to-accept* (WTA), see Horowitz and McConnell (2002). The difficulty of determining the WTP value (WTA) is not only statistical in nature (i.e. due to the estimation based on a random sample)—a single respondent would usually find it difficult to accurately determine own value of life (i.e. how much they think the public regulator should value one QALY when making decisions on behalf of society).

To account for imprecision in describing various phenomena, fuzzy sets were introduced in the literature (Zadeh, 1965): an extension of standard sets that enable partial membership of elements, as defined by a membership function with values possibly spanning the entire  $[0,1]$  range. This approach was used in **Jakubczyk and Kamiński (2017)**. The aim of the study was: (1) to propose an axiomatic foundations to define the fuzzy relation of preferences in the context of two-criterion comparisons of medical technologies (based on effect and cost); (2) to link the such defined preference relation to the WTP and WTA values redefined as fuzzy sets and demonstrate the possibility of empirically estimating these parameters; (3) to demonstrate the possibility of accounting for the stochastic uncertainty of estimates (of cost and effect) in supporting the decision maker.

The study was both an empirical and a theoretical one. The empirical part (chapter 3.1 in the original publication) presented the results of the authors' survey among HTA experts in Poland about their subjective values of WTP and WTA, accounting for the lack of precision in how these values are perceived. The results of the survey confirmed, on one hand, the significance of both criteria (i.e. also of the financial criterion) when comparing health technologies, but also, on the other hand, that the values of WTP/WTA threshold are only assessed with imprecision. The obtained results also suggest a new possible explanation of the discrepancy between the WTP and WTA values observed in the literature (the WTP-WTA disparity). Whereas the membership functions for WTP and WTA when treated as fuzzy sets reach the membership of 0.5 for similar monetary values, the respondents in experimental research report (and in actual decisions use as a threshold) WTP values for which their conviction is much stronger (the membership clearly exceeding 0.5), see Fig. 4 in the original publication. Thus, the observed discrepancy may be due to the respondents being conservative when interpreting own imprecision regarding the introspectively determined preferences.

In the theoretical part of the study, we proposed the axiomatic foundations of the fuzzy preferences between health technologies in pair-wise comparisons (chapter 3.2). As part of this axiomatization, the fuzzy WTP and WTA (Definitions 1 and 2) were defined, also indicating how to use surveys (for example, as the one used in the paper) to approximate the membership function. An innovative element of the study was the formal use of fuzzy sets to model the imprecision in preferences between health technologies.

Combining the stochastic uncertainty with non-stochastic imprecision (regarding the WTP and WTA values), we defined the *fuzzy expected net benefit* (fENB) and *fuzzy expected acceptability* (fEA) for a given decision alternative as alternative methods for assessing the attractiveness of the available alternatives. Depending on the assumed level of conviction (in the sense of the membership function values), these notions can be interpreted as, respectively,

the expected value of NB offered by a given alternative with a given conviction, and the probability that a given alternative offers the larger NB. It was shown that the properties of fENB and fEA are intuitive: membership functions are monotonic (Proposition 2 in the original publication). At the same time both notions extend in a natural way the classic concepts: expected NB and standard CEAC for a given WTP, i.e. are equal to those equivalents if we remove the fuzziness (i.e. we adopt crisp values of WTP and WTA, see Proposition 3 in the original publication). That the fENB and fEA have intuitive limit properties was also shown in the case when the amount of stochastic uncertainty in the problem is being reduced (Proposition 4 in the original publication).

I continued the above-introduced research in **Jakubczyk (2015a)**.<sup>12</sup> I extended the methodology of Jakubczyk and Kamiński (2017) by accounting for the possibility of more than two decision alternatives (assuming all alternatives are being compared to a single *status quo*). I showed that the fuzzy net benefit (fNB) respects the dominance and extended-dominance criteria (Proposition 1 and 2 in the original publication), often used in the standard CEA analysis. As part of the model under consideration, I proposed a choice function that can be used in a menu of (finitely) many alternatives based on comparing fNB (Definition 3 in the original publication) and proved the correctness of its definition (Proposition 3).<sup>13</sup> The obtained results allow for the practical application of a fuzzy approach to real decision problems, in which more than two decision alternatives are often compared.

#### **4.4. Using the fuzzy sets in the modelling of lack of precision in the preferences for health states**

As presented in section 4.3, Jakubczyk and Kamiński (2017) indicated how to take into account the lack of precision in preferences for health (QALYs) and cost. The follow-up to this stream of research was to study how to account for the lack of precision in preferences for health itself, i.e. when making trade-offs between quality and longevity of life and between various aspects of the health-related quality of life.

**Jakubczyk and Golicki (2018)** introduced a model of fuzzy preferences in the context of the EQ-5D-3L descriptive system. Simplifying, it was assumed that the utility associated with the deterioration of a given dimension to a given level is described not by crisp number but by an interval (in the discussed study normal and rectangular fuzzy numbers were considered, i.e. the membership function takes only values 0 and 1). An innovative element of the study was accounting for the lack of precision, i.e. the inability of the decision-maker to determine which of the many numbers (with the value of the membership function equal to 1) is a more appropriate description of the actual utility. In this sense, the sets describing utility should be interpreted as the *epistemic fuzzy sets* (see Couso and Dubois, 2014).

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<sup>12</sup> The difference in the publication date was due to the publishing process of Jakubczyk and Kamiński (2017), which was available online in 2015.

<sup>13</sup> I continue the stream of research. In 2016, I presented in the 6th World Conference on Soft Computing, Berkeley, USA, 2016, the study “Estimating the membership function of the fuzzy willingness-to-pay/accept for health via Bayesian modelling” — the papers presented are supposed to be published by Springer. During the 4th Meeting of the International Academy of Health Preference Research, Hamburg, Germany, 2016, I presented the poster “Estimating fuzzy willingness-to-pay/accept in healthcare: survey results from England” (co-author: Ł. Tanajewski), in which we used the method from Jakubczyk and Kamiński (2017). In the The IV AMMCS (Applied Mathematics, Modeling and Computational Science) International Conference, Waterloo, Canada, 2017, I presented the paper “Estimating the Crossover Point of a Fuzzy Willingness-to-Pay/Accept for Health to Support Decision Making”, and the paper is currently submitted for publication.

The study showed how to estimate the parameters of the proposed model of preferences based on datasets collected in a standard TTO experiment (i.e. an experiment not directly targeted at interval-type results): a model of the TTO data generating process was constructed based on a given preference structure and then the method of estimating its parameters in the Bayesian framework was proposed. An advantage of the proposed approach is the possibility of using previously collected standard TTO data for estimation under the new preference model (standard TTO data have so far been collected in several dozen countries).

The feasibility of using the proposed method has been empirically demonstrated using the data collected by Golicki et al. (2010), discussed above. The results obtained demonstrate (see Table 1 in the original publication) that (intuitively) greater imprecision is related to the third level for all dimensions and that the lack of precision translates into approximately the length of sections describing the utility of individual dimensions (on the third level) amounting to ca. 0.06 (on the standard QALY scale). The PD dimension was associated with more imprecision—the length of the interval amounts to 0.13.

Jakubczyk and Golicki (2018) is located in the currently emerging stream of research that discusses the specificities of the TTO method. Firstly, the data arise as the result of a series of pairwise comparisons; hence, also the observations obtained before the indifference was established are available. Secondly, due to the TTO study protocol, the number of possible final answers is limited; hence, that the exact indifference was identified should only be considered as an approximation. Currently, research is being conducted on how to take this approximation into account when estimating preferences, when the preferences are defined in the standard, crisp way, see Ramos-Goñi et al. (2017). An innovative element in the Jakubczyk and Golicki (2018) is to consider that preferences themselves are fuzzy (hence the estimand of the study is redefined).<sup>14</sup>

In parallel to the above, I participated in the study **Jakubczyk, Kamiński, and Lewandowski (2018)**. The aim of the work was to construct a model of preferences accounting for imprecision, which can be estimated based on data collected in the discrete choice experiment (DCE). DCE is an alternative to TTO, the other most commonly used method of eliciting the utility values of health states, see Bansback et al. (2012).

In the study, we used data coming from a representative United States of America sample, originating from the DCE data modelling competition (predicting the structure of choices between health states based on the observed choices in the training sample).<sup>15</sup> In the data, EQ-5D-5L descriptive system was used: each dimension (identical as in EQ-5D-3L) is described using five levels, see Herdman et al. (2011). In Jakubczyk, Kamiński, and Lewandowski (2018) an alternative model of preference was proposed, in which the disutilities of individual dimension/level combinations are defined as fuzzy sets. In contrast to Jakubczyk and Golicki (2018), in Jakubczyk, Kamiński, and Lewandowski (2018) the general membership function were assumed (taking values also in (0,1), although it was assumed that the membership function is linear) and the fuzzy sets are interpreted as ontic fuzzy sets, instead (see Couso and Dubois, 2014).

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<sup>14</sup> I continue working in the discussed stream of research within a research grant „*A fuzzy approach to time trade-off experiment in EQ-5D-3L valuation*” awarded by the EuroQol Reserach Foundation. I propose a modification of TTO, to directly elicit fuzzy utility assessment (defined in a more general way, as trapezoid and normal fuzzy numbers), as well as within a OPUS10 NCN grant „*Metody analizy decyzji w problemach wielokryterialnych i szacowania skłonności do zapłaty/akceptacji przy wykorzystaniu modelowania rozmytego*”.

<sup>15</sup> More information on the competition can be found at <http://iahpr.org/eq-dce-competition/> [last accessed 1.2.2018] and in the paper Jakubczyk et al. (2017).



In the study, a model of how respondents choose between pairs of health states (with duration) was proposed, along the method of estimating the parameters such a model based on the likelihood function maximization. The proposed elicitation method also made it possible to assess the estimation error (with the bootstrap approach). The results of the study (Table 1 in the original publication) indicate a large imprecision in societal preferences, especially for PD and AD dimensions.

An important feature of the proposed model is how it respects the Pareto dominance between the compared alternatives, as compared to the standard approach to modelling choices in DCE experiments (e.g. when using the exponential Bradley-Terry model): in the study discussed here, if durations are equal and one health state Pareto-dominates the other (i.e. all dimensions are set at a lower level) and the ranges of disutilities for differing levels in individual dimensions for which the membership function is in  $(0,1)$  interval are disjoint, the decision-maker will select the dominant state with probability 1.

#### **4.5. Summary of the results obtained in the series of publications**

The research conducted by me that was included in the monothematic series of publications is interdisciplinary: it relates the economics considerations (involving the elements of decision theory, operational research, and econometrics) to the perceived importance of health (both the relative importance of various attributes of health and the importance of health relative to the financial aspect), also taking into account demographic heterogeneity and psychological phenomena (inherent imprecision of preferences and the impact of *Weltanschauung*, e.g. religiosity, on preferences).

The series discussed above contains both theoretical and empirical studies. Among the theoretical ones there are Jakubczyk and Kamiński (2010), Jakubczyk (2015b, 2015a), and Jakubczyk and Kamiński (2017). The first two studies examined the properties of the CEAC as decision-making method and proposed modifications to remove the counter-intuitive (and undesirable) properties of standard CEAC; the last two studies demonstrate (by providing axiomatic foundations) how to formally account for the lack of precision in the health preferences (by using the fuzzy set theory).

Among the empirical studies there are Jakubczyk (2009), Golicki et al. (2010), Jakubczyk, Golicki, and Niewada (2016), Jakubczyk and Golicki (2018), and Jakubczyk, Kamiński, and Lewandowski (2018). In all of them, I examined the societal preferences for health states; the last two studies additionally accounted for the inherent imprecision of preferences in individual respondents. The empirical studies also contained some innovative methodological elements indicated above (the use of fuzzy set theory). Similarly, there were some empirical elements present in the theoretically-oriented papers: Jakubczyk and Kamiński (2017)—an empirical questionnaire on WTP/WTA values, and Jakubczyk (2015b)—a case study demonstrating the possibility of using the CEAC modifications in actual decision problems.

The papers described above cover the streams of both the positive and normative economy. The first stream is represented by studies of actual preferences, as reported by respondents in surveys: Jakubczyk (2009), Golicki et al. (2010), Jakubczyk, Golicki, and Niewada (2016), Jakubczyk and Golicki (2018), and Jakubczyk, Kamiński, and Lewandowski (2018). The studies from the second stream include works on the new methods of comparing the decision alternatives: Jakubczyk (2015a, 2015b) and Jakubczyk and Kamiński (2017).

Fuzzy sets constitute an important element of my research; they are a tool that allows formally accounting for the imprecision of preferences. This method is used and studied in Jakubczyk (2015a), Jakubczyk and Kamiński (2017), Jakubczyk and Golicki (2018), and Jakubczyk, Kamiński, and Lewandowski (2018). The studies used the fuzzy sets in various ways; in particular in Jakubczyk and Golicki (2018), the fuzzy sets should be interpreted in the epistemic sense, while they should be interpreted in the ontic sense in the remaining three studies. I consider it important that in my research I tried to indicate how the theoretical concepts can be put to practical use, e.g., by showing the methods how the parameters of the preference models can be estimated based on empirical data.

In my research, I used empirical data acquired using various methods: time trade-off experiments (e.g. Jakubczyk, 2009), discrete choice experiments (Jakubczyk, Kamiński, and Lewandowski, 2018), or Likert scales (Jakubczyk and Kamiński, 2017). The research was carried out using data collected in Poland (e.g. Jakubczyk, Golicki, and Niewada, 2016) or abroad, in the United States (Jakubczyk, Kamiński, and Lewandowski, 2018).

I consider the following elements as the most important contribution of the papers collected in the series into the development of the economics discipline:

1. determining the societal preferences in Poland for the health states, i.e. the importance of criteria describing various aspects of the health-related quality of life and longevity of life (in Golicki et al., 2010; Jakubczyk, 2009), proving by statistical analysis the robustness of the results to the study protocol, i.e. the number of thought experiments performed by one respondent (Golicki et al., 2010), and demonstrating the interdependencies between health state attributes in the impact on utility, i.e. demonstrating the complementarity of various quality of life attributes (Jakubczyk, 2009);
2. describing the heterogeneity of Polish society in terms of demographic characteristics impacting the health preferences (Jakubczyk, 2009), as well as demonstrating how the *Weltanschauung* of respondents limits the possibility of using TTO (time trade-off) to elicit preferences and proposing new methods to de-bias the results (through econometric modelling or modification of the TTO method, Jakubczyk, Golicki, and Niewada, 2016);
3. identifying the limitations of standard methods for comparing alternatives in the decision problems related to comparing health technologies under risk and with two criteria (cost-effectiveness acceptability curves; Jakubczyk and Kamiński, 2010); and subsequently constructing new methods to use in such comparisons (the recursive and conservative cost-effectiveness acceptability curves; Jakubczyk, 2015b) and demonstrating their more favourable properties in decision problems of various types (with two or more than two alternatives, with estimation errors defined by symmetric or asymmetric probability distributions, in the presence or absence of correlation between the estimation errors);
4. axiomatization of fuzzy preferences between decision alternatives in the context of comparing health technologies (described using two criteria: cost and effect) under risk, in case of pairwise comparisons (Jakubczyk and Kamiński, 2017), and then proposing methods of comparing multiple (possibly more than two) decision alternatives when preferences are imprecise and demonstrating the intuitive properties of the proposed methods (Jakubczyk, 2015a); additionally, proposing a definition of the willingness to pay/accept (WTP and WTA) as fuzzy sets, which may partially explain the discrepancy between WTP and WTA values observed in the literature (Jakubczyk and Kamiński, 2017);
5. demonstrating how to use the fuzzy set theory to construct formal models of health preferences allowing for imprecise perception of the importance of individual criteria (attributes describing health-related quality of life in the EQ-5D system) and developing methods for

estimating such preferences based on empirical data obtained in standard elicitation methods (hence, also the ones collected in the past): TTO (time trade-off) and DCE (discrete choice experiments) (Jakubczyk and Golicki, 2018, Jakubczyk, Kamiński, and Lewandowski, 2018).

What I find important, the obtained results also have practical implications. The results of Golicki et al. (2010) are officially indicated as value set to be used in Poland in CEA analyses when assessing health technologies in decisions by the public regulator. Jakubczyk (2015b), Jakubczyk and Kamiński (2017), and Jakubczyk (2015a) introduced methods of comparing decision alternatives that can be directly applied to the real-life problems when comparing health technologies in the context of stochastic uncertainty due to estimation error.

Finally, I would like to emphasize the personal satisfaction I continue to draw from the direction of scientific research I chose years ago.<sup>16</sup> What is important to me as an economist, my research allows me to actually see utilities as actual, tangible numbers; while the notion of utility remains only an elusive, theoretical concept for many. I am also convinced that the results of my studies allow for making better decisions, i.e. for managing the limited resources in health care market in a more rational way. As the renowned economist Peter Wakker (2010) stated: „*the medical domain is one of the most important fields of application for decision theory*”.<sup>17</sup>

## 5. Presenting the major remaining publications

Apart from the works described above, my research also resulted in other publications, the most important of which I discuss in a synthetic way in the present section (the full list of publications is included in the bibliometric analysis attached to the application).

During my scientific career I participated also in other research related to the issue of describing health preferences or describing the decisions of a public regulator regarding health technologies. The paper by **Jakubczyk et al. (2017)** discusses the course and results of the competition for predicting choices in DCE by individual respondents (in the US population) that was held by the International Academy of Health Preference Research.<sup>18</sup> The team of Discreetly Charming Econometricians which I managed won the competition. The publication discusses the prognostic models submitted to the competition, and the winning model in particular. The most important conclusions from this model are as follows: firstly, the significance of dimensions varies depending on the duration of the state of health (e.g. the significance of the UA increases when long-term health states are considered); secondly, time is perceived non-linearly (similar conclusion follows from the models used by other teams in the competition), i.e. contrary to the standard QALY model; thirdly, when transforming the difference in utility between compared alternatives into the probability of one option being selected the Cauchy distribution should be used (and not a normal or logistic distribution, as in a probit or logit approach typically used); fourthly, comparing a non-degenerate health state to immediate death requires a different model than comparing two non-degenerate health states.

In the study **Niewada et al. (2013)** we attempted to identify factors related to the positive or negative decisions regarding the financing of health technologies as issued by the Agency for Health Technology Assessment (Consultative Council / Transparency Council) in Poland. The study covered over 300 recommendations issued up to 2011. As a result of the

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<sup>16</sup> Even if it is not an easy direction, because it is interdisciplinary and requires dealing with discomforting issues of illness, pain, death, etc.

<sup>17</sup> Chapter 3.1 in the book.

<sup>18</sup> See <http://iahpr.org/eq-dce-competition/> [last access 1.2.2018].

analysis, no statistically significant effect of ICER on decisions was found (the study covered the time before the current Reimbursement Act defining the WTP threshold directly). The aim of **Jakubiak-Lasocka and Jakubczyk (2014)** was to compare the use of various types of economic analyses (analyses in which QALY is used as a measure of the effect, and analyses that use years of life, not adjusted for quality: LYG, *life-years gained*). The analysis covered all applications submitted to the Agency for Health Technology Assessment in Poland until 2011. It was shown that the analyses using the quality of life (i.e. the effect was expressed as utility) constituted the majority. Counter-intuitively, although including the utility of health requires the introducing greater number of parameters in the analysis, the estimates of ICER was associated with smaller error on average. An attempt to detect bias in selecting the form of analysis (CEA or CUE) did not indicate the presence of this (undesirable) phenomenon.

I also conducted theoretical research in the field of game theory and decision support, not related to any specific market area. In the **Amir, Jakubczyk, and Knauff (2008)**<sup>19</sup> study, we considered the properties of symmetric supermodular games (with complementary player strategies). We indicated the conditions (various sets of conditions, e.g. the case of full joint interests between players—Proposition 2 in the original publication—or the superjoin property with additional technical requirements—Proposition 4 in the original publication), when asymmetric Nash equilibria (we considered only pure-strategy equilibria) pay-offs that are Pareto dominated by pay-offs of some symmetric Nash equilibrium. In turn, the objective of the **Kamiński, Jakubczyk, and Szufel (2018)** study was to propose new methods (and to examine their properties) to conduct sensitivity analyses in sequential problems under risk as described by decision trees. A conceptual framework was proposed, within which the stability of solutions can be studied (with respect to the disturbances of probabilities describing the decision problem) and the solutions robust to pessimistic/optimistic/mode-favouring perturbations can be sought. Importantly, the publication is accompanied by free software, enabling the use of the presented methods.<sup>20</sup>

An important stream of my research relates to the theoretical and empirical aspects of estimating cost of illness and other health-related phenomena, in particular the cost incurred from the societal perspective (i.e. *indirect costs*, cf. Jakubczyk et al., 2010, and Koopmanschap and van Ineveld, 1992). For example, **Jakubczyk i Koń (2017)** present a theoretical economic model of a company which takes into account the employer's expectations and adjustments. Within this model it is shown how the adjustments may impact the true value of indirect cost, and in particular that the adjustments can result in lowering of the cost, contrary to the earlier results by Pauly et al. (2002). **Lasocka, Jakubczyk, and Siekmeier (2013)** studied the lost productivity cost in Poland resulting from tobacco-related diseases using the human capital method (see Liljas, 1998). The estimated annual cost amounted to approx. 15 billion PLN. I also studied the impact of diseases on the costs incurred from the public payer perspective. In the series of publications **Czech et al. (2015)**, **Jakubczyk et al. (2015)**, and **Jakubczyk et al. (2016)**, we estimated the risk of hypoglycaemic episode depending on the antidiabetic therapy used (based on systematic review and meta-analysis using Bayesian modelling), the structure of resource consumption in case of such an episode (again, on the basis of literature review and meta-analysis), and the total costs of hypoglycaemia in nine Central and Eastern European countries. I participated in the cost-effectiveness studies of the application of medical procedures. In the work of **Pelczarska et al. (2018)** various screening strat-

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<sup>19</sup> The study was started before my obtaining of PhD degree. The results presented in the study (or the very topic of the study) are unrelated to the PhD dissertation.

<sup>20</sup> <https://github.com/pszufe/chondro/>, [last access 1.2.2018].

egies aimed at detection of familial hypercholesterolemia were examined, using the own-developed economic model describing the course of diagnostic and treatment process. We demonstrated from the point of view of economic and medical criteria (taking into account the influence of treatment on the utility of health states), the rationale behind implementing the studied screening strategies.

## 6. A summary of scientific achievements

At the time of submitting the application, my scientific achievements awarded MNiSW points (including only publications prepared entirely after obtaining the doctoral degree<sup>21</sup>) include:

- 14 papers published in the academic journals having *impact factor* (IF),
- 14 papers published in the academic journals contained in the list B of the Polish Ministry of Science and Higher Education (MNiSW),
- 8 chapters published in scientific monographs,
- serving as an editor in two scientific monographs.

The total IF for the publications mentioned above amounts to 44.984, and the total sum of the MNiSW points amounts to 577. The total number of citations of my works (according to the bibliometric by the Library of SGH, irrespectively of the date of publication) amounts to

- 121 (according to the Web of Science Core Collection, autocitations excluded),
- 162 (according to Scopus),
- 366 (according to Google Scholar).

The citations result in the Hirsch index amounting to

- 5 (according to the Web of Science Core Collection, autocitations excluded),
- 7 (according to Scopus),
- 9 (according to Google Scholar).

I presented the results of my research after obtaining the doctoral degree at many scientific conferences in Poland and abroad:

- 7 oral presentations at the international scientific conferences abroad,
- 4 oral presentations at the international scientific conferences held in Poland (in English),
- a presentation at the scientific seminar (*University of Illinois at Chicago*),
- 2 presentations of my results by invited discussants with authors' comments (additionally, I served as a discussant and presented somebody else's results),
- 20 poster presentations at the international scientific conferences abroad,
- 11 oral presentations at national conferences.

I presented my results, among others, at the following conferences: *EuroQol Group Plenary Meeting* (twice an oral presentation by a discussant, once a co-author of a poster), *International Academy of Health Preference Research* (three oral presentation, once a poster), *International Workshop on Multiple criteria decision making* (twice), *INFORMS Annual Meeting*,

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<sup>21</sup> The bibliometric analysis prepared by the Library of SGH and also attached to the application contains all the published studies, irrespectively of the date. All my scientific achievements awarded MNiSW points is also presented in the Appendix 4, where my contribution to each publication is described in details (along assessed contribution in percentages). In this appendix I also presented my didactic and organizational achievements.

28<sup>th</sup> European Conference on Operational Research, 6<sup>th</sup> World Conference on Soft Computing, The 10<sup>th</sup> Conference on the European Society for Fuzzy Logic and Technology, The IV AMMCS (Applied Mathematics, Modeling and Computational Science) International Conference, and the EuroScience Open Forum.

I participated in the work of organizing and scientific committees at international conferences (e.g. 32<sup>nd</sup> EuroQol Plenary Meeting and International Conference on Group Decision & Negotiation), I served as a moderator at the national and international conferences (during INFORMS Annual Meeting and International Society for Pharmacoeconomics and Outcomes Research Annual European Congress).

I reviewed papers submitted for publication in multiple journals included in the Journal Citation Reports database:

- *Annals of Operations Research* (2015, IF = 1.217, MNiSW = 25)<sup>22</sup>,
- *The European Journal of Health Economics* (2013 and 2017, IF = 1.774, MNiSW = 35),
- *European Journal of Operational Research* (2012, IF = 2.358, MNiSW = 40),
- *Expert Review of Pharmacoeconomics & Outcomes Research* (2015, IF = 1.669, MNiSW = 20),
- *Health Economics* (2010 and 2012, IF = 2.227, MNiSW = 35),
- *Health and Quality of Life Outcomes* (2016, IF = 2.12, MNiSW = 35),
- *Kardiologia Polska* (2014, IF = 0.539, MNiSW = 15),
- *Pharmacoeconomics* (2010 and 2011, IF = 2.45, MNiSW = 30),
- *Quality of Life Research* (four times in 2016 and 2017, IF = 2.486, MNiSW = 35).

I reviewed two applications for the Fulbright Junior Research Award per invitation by the programme committee. I regularly review the applications and final reports submitted to the scientific association the EuroQol Group.

I am a member of the following scientific organisations:

- *EuroQol Group* (<https://euroqol.org/>, fewer than 90 members worldwide; within the group I am a member of the *Valuation Working Group* handling, e.g., the reviewing the grant applications and final reports),
- *International Academy of Health Preference Research* (<http://iahpr.org/>, fewer than 40 tenured members and 40 members worldwide),
- *International Society for Pharmacoeconomics and Outcomes Research*,
- Polish Pharmacoeconomic Society (Polskiego Towarzystwa Farmakoekonomicznego),
- Polish National Group of the International Society for Clinical Biostatistics (Polskiej Grupy Narodowej Międzynarodowego Towarzystwa Biostatystyki Klinicznej).

I am the manager of three ongoing research grants:

- since 2016, in the grant “**Methods of decision analysis in multi-criteria decisions problems and of estimating the willingness-to-pay/to-accept using fuzzy modelling**” („**Metody analizy decyzji w problemach wielokryterialnych i szacowania skłonności do zapłaty/akceptacji przy wykorzystaniu modelowania rozmytego**”), OPUS 10 grant awarded by the National Science Center, agreement no. UMO-2015/19/B/HS4/01729;

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<sup>22</sup> Here, the newest available MNiSW and IF points are presented.

- since 2016, in the grant “**A fuzzy approach to time trade-off experiment in EQ-5D-3L valuation**”, awarded by the EuroQol Research Foundation (no.: EQ Project 2015320);
- since 2016, in the grant “**Building values sets based on TTO results by averaging model predictions and actually observed means**”, awarded by the EuroQol Research Foundation (no.: EQ Project 2016640).

I am a scientific partner in the POLONEZ grant („**Evidence use in the allocation of health care budgets: A comparative study of drug reimbursement in Poland, Canada, and Germany**”, manager: W. Wranik) and an investigator in the Horizon 2020 grant (“**Raising Open and User-friendly Transparency-Enabling Technologies for Public Administrations (ROUTE-TO-PA)**”, manager: B. Kamiński) and two grants awarded by *The EuroQol Foundation* (“**EQ-5D-5L valuation in Poland**” and “**EQ-5D-5L valuation in Poland: a methodological extension**”, supervisor: D. Golicki). I manager a grant awarded by MNiSW within the program „Juventus Plus” („**Recursive cost-effectiveness acceptability curves in comparing the health technologies**” / „**Rekursywne krzywe akceptowalności w porównaniach technologii medycznych**”).

I received a scholarship from the Ministry of Science and Higher Education for outstanding young scientists (2011); I was awarded the Fulbright Senior Award scholarship (2015-2016). I received prizes and congratulatory letters from the Rector of the SGH Warsaw School of Economics for scientific and organizational activities.

My scientific achievements along with didactic and organizational achievements are described in more detail in Appendix 4 to the application.

## 7. Abbreviations and acronyms used in the self-presentation

AD	<i>anxiety/depression</i> , a dimension in the EQ-5D-3L/5L descriptive system
AOTMiT	Agencja Oceny Technologii Medycznych i Taryfikacji (Agency for Health Technology Assessment and Tariff System)
CEA	<i>cost-effectiveness analysis</i>
CEAC	<i>cost-effectiveness acceptability curve</i>
CUA	<i>cost-utility analysis</i>
DCE	<i>discrete choice experiment</i>
fEA	<i>fuzzy expected acceptability</i>
fENB	<i>fuzzy expected net benefit</i>
fNB	<i>fuzzy net benefit</i>
FOSD	<i>first-order stochastic dominance</i>
GDP	<i>Gross Domestic Product</i>
HDI	<i>Human Development Index</i>
HTA	<i>health technology assessment</i> )
HRQoL	<i>health-related quality of life</i>
ICER	<i>incremental cost-effectiveness ratio</i>
IF	<i>impact factor</i>
LYG	<i>life years gained</i>
MNiSW	Ministerstwo Nauki i Szkolnictwa Wyższego (Ministry of Science and Higher Education)
MO	<i>mobility</i> , a dimension in the EQ-5D-3L/5L descriptive system
NB	<i>net benefit</i>
PD	<i>pain/discomfort</i> , a dimension in the EQ-5D-3L/5L descriptive system
QALY	<i>quality-adjusted life years</i>
SC	<i>self-care</i> , a dimension in the EQ-5D-3L/5L descriptive system
TTO	<i>time trade-off</i>
UA	<i>usual activities</i> , a dimension in the EQ-5D-3L/5L descriptive system
VAS	<i>visual analogue scale</i>
WTA	<i>willingness to accept</i>
WTP	<i>willingness to pay</i>



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