Fetal monitoring at home in high-risk pregnancy. An integrated clinical and economic evaluation

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Citation for published version (APA):
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In this thesis, we have evaluated an obstetric program on de-institutionalized care, viz. the introduction of domiciliary antenatal fetal monitoring as compared to conventional in-hospital antenatal fetal monitoring in a selected group of high-risk pregnant women. De-institutionalization is the transfer of the location where health care is provided from highly specialized medical settings to medically less structured settings. We have applied de-institutionalization to obstetric care, an area in the Dutch health care system in which preferably non-clinical care is provided whereas other countries would consider routine clinical care.

This chapter consists of two parts. In the first part, section 11.1, the study results are summarized and put in perspective with the incentives to de-institutionalized care (technical feasibility, effectiveness, efficiency, quality of care, individualization of care, and de-medicalization). The remainder of this chapter, section 11.2, is devoted to the general discussion. Several methodological aspects from this thesis are discussed in section 11.2.1, whereas aspects of the implementation of domiciliary monitoring and its place in obstetric care are addressed in section 11.2.2.

11.1 Summary

Chapter 1 provides an introduction to de-institutionalization and an overview of experiments on cardiotocography-assisted domiciliary antenatal fetal monitoring. Domiciliary monitoring has been tried in pregnancies at risk for preterm labor and birth, pregnancies complicated by prelabor preterm rupture of membranes (PPROM), and in pregnancies at risk for fetal distress. The first application has shown that domiciliary monitoring is not effective in preventing preterm delivery. The second application is lacking firm evidence on the equivalence of neonatal outcome (low statistical power) and only few women are eligible for this mode of monitoring. Studies from the third category, antenatal monitoring for fetal distress, show that domiciliary monitoring is feasible and acceptable to the pregnant women. Safety, i.e. the equivalence of neonatal outcome, is established convincingly in only one study. No firm evidence exists on cost-effectiveness, women's preferences, maternal health-related quality of life, quality of care or effects on the social environment.

This thesis addresses the evaluation of domiciliary monitoring of pregnancies at risk for fetal distress. Our aim was to answer the following five questions.

1. Is domiciliary monitoring effective? Is domiciliary monitoring feasible?

The feasibility of domiciliary antenatal fetal monitoring and its effectiveness as compared to in-hospital monitoring have been evaluated in Chapter 2, 3 and 4. High-risk pregnant women, all with an indication for clinical surveillance (antenatal fetal CTG-
monitoring and hospitalization), were invited to participate in a randomized controlled trial. Women allocated to in-hospital monitoring were admitted to the hospital and received care under standard arrangements: daily fetal heart rate monitoring, daily measurement of blood pressure and additional care if necessary. Women to whom domiciliary monitoring had been assigned were visited daily at home by a research midwife. Fetal heart rates were recorded for at least 30 min. (Oxford Sonicaid System 8000, a computerized system for antenatal fetal heart rate registration and analysis) and blood pressure was measured. Fetal heart rates were stored digitally and sent by modem to the hospital where the traces were computer-analyzed, printed and reviewed by an obstetrician. Women monitored at home had to visit the outpatient department once a week for routine surveillance. If necessary, additional care such as lab tests or ultrasound examination was given. All women should deliver in the hospital. Fetal surveillance did not include intrapartum monitoring.

In Chapter 2, safety is evaluated in terms of perinatal morbidity. The primary outcome was Prechtel's neonatal neurological optimality score, a proxy for long-term outcome. At least 150 women were needed to test a clinically relevant difference of two points in mean (SD 4.3) Prechtel scores (two-tailed $\alpha = 0.05$, $1-\beta = 0.80$, equivalence design). The assignment of monitoring strategy occurred by computerized block-randomization stratified for gestational age (< 37, 37-42, $\geq 42$ weeks). Of the 174 eligible women, 24 women declined study participation and 150 were randomized to in-hospital monitoring ($n=74$) or domiciliary monitoring ($n=76$). There was one case of perinatal mortality in each group. Both cases were not considered attributable to the study. Prechtel's optimality score (median 59), the proportion of optimals (Prechtel score $\geq 58$; 49 (67%) in the in-hospital group versus 53 (71%) in the domiciliary group) and the secondary outcomes (incidence of complications, obstetric interventions at labor, birth weight, gestational age at delivery, and maternal and neonatal admission rates) did not differ significantly. We conclude that domiciliary fetal monitoring is as safe as in-hospital monitoring. Obstetric and fetal outcome are not affected if in-hospital monitoring is replaced with domiciliary monitoring.

Chapter 2 also allows the evaluation of the feasibility of domiciliary monitoring. Domiciliary monitoring is highly accepted: only 24 (14%) of the eligible women declined trial participation. Technical problems with data transmission occurred twice. In one case, the monitoring equipment had a technical defect that was solved easily. In the other case, a woman's telephone did not function appropriately. Professional care was available 24 hours per day and 7 days per week (the monitoring team during office hours, the obstetrician on call at out-of-office hours). Information exchange between caregivers occurred without problems. The GP and community midwife were informed at inclusion. During domiciliary monitoring, all information was accessible to the caregivers in a daily log at the women's home. As far as we know, women largely complied to bed rest if advised. Of the 76 women allocated to domiciliary monitoring, 23 (31%)
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were hospitalized antenatally, without delay, for intensive surveillance, mostly because of ominous fetal heart rates (n=16). All women delivered in the hospital except one woman randomized to in-hospital monitoring who delivered at home in the night before hospitalization. There were no problems with transportation from the home to the hospital. We conclude that domiciliary monitoring in our group of high-risk women is feasible.

In Chapter 3, the inter-observer variability of Prechtl scores is evaluated to validate the observed equivalence of Prechtl scores. Two well-trained observers independently performed Prechtl's neurological examination in 21 fullterm healthy neurological normal newborns. The interclass correlation coefficient was 0.31. Kappa for optimality was only 0.19. A systematic difference of 1.3 points existed between the two observers. We conclude that Prechtl's optimality concept is a valid tool for measuring neonatal health outcome but due to interobserver variability, only differences of at least two points are considered clinically relevant.

In Chapter 4, we evaluated maternal health outcomes in high-risk women antenatally (at study entry) and at 8 weeks after the delivery. Health status was measured as generic health-related quality of life (RAND SF36) and social support (Social Experiences Questionnaire). We used two reference groups to compare health outcomes: low-risk pregnant women and a reference group of United States women aged 18-44. Antenatal health, particularly physical health, of low-risk and high-risk women was considerably lower compared to the US reference group; being pregnant has a negative effect on maternal health status. Antenatal mental health and physical health were less favorable in high-risk women than in low-risk women (p < 0.05). After the delivery, physical health had improved considerably in all groups but mental health was still lower in high-risk women than in low-risk women (p < 0.01). Within the high-risk group, domiciliary and in-hospital monitored women had equivalent physical health but mental health was consistently superior among in-hospital monitored women. This may be due to a response shift. Alternatively, in-hospital women may have felt more reassured about their condition as compared to home-monitored women. A complete mental recovery presumably takes more time than a physical recovery. We conclude that maternal health outcome is not adversely affected if in-hospital monitoring is replaced with domiciliary monitoring.

2. Is domiciliary fetal monitoring efficient? Does it contribute to cost containment?

The economic efficiency (cost-effectiveness) of domiciliary fetal monitoring has been evaluated in Chapter 5. As neonatal and maternal outcome did not differ between domiciliary and in-hospital monitoring, economic outcome was equal to the cost difference between the strategies. The median length of the antenatal period (8.0 days) was equal between the strategies. In the in-hospital-monitoring group, 14 women were discharged
antenatally and re-hospitalized at the onset of the delivery. In the domiciliary monitoring group, 24 women were hospitalized antenatally for increased surveillance. Seven of them were also discharged antenatally; they continued domiciliary monitoring. The in-hospital-monitoring group spent on average 9.3 days in the hospital and 2.9 days at home. Women in the domiciliary monitoring group spent on average 1.9 days in the hospital and 9.3 days at home. The mean (SD) antenatal costs per woman were $3558 ($2841) in the in-hospital monitoring group and $1521 ($1459) in the domiciliary monitored group, producing a $2037 antenatal cost difference (p < 0.001). Cost-shifting (antenatal cost decrease at the expense of increased costs of delivery and post partum costs) did not occur. We conclude that domiciliary monitoring is not only safe, it is also more efficient as it reduces costs by one half, and de-medicalizing. Domiciliary monitoring is therefore preferred to in-hospital monitoring.

3. Does domiciliary monitoring influence continuity of care?

In Chapter 6, the effects of domiciliary and in-hospital monitoring on the primary family caregiver have been evaluated. Family burden was measured with an ad hoc questionnaire in terms of changed household activities and, attendance, worries, professional caregivers' attitudes, caregiver's health status, the impact on caregiver's own life, and effects on children. Family burden in a group of low-risk pregnant women was used as reference. Psychometric properties of the questionnaire were acceptable (Cronbach's α = 0.87). The family caregivers of low-risk women reported better health, fewer worries, and less burden of professional caregivers' attitudes than the caregivers of the high-risk women. Within the high-risk group, differences in family burden between the domiciliary and in-hospital group were absent. Adjusted for co-variables, the informal caregivers of in-hospital monitored women reported slightly less favorable health (p=0.12) and a higher burden on children (p=0.02). The role of burden and rewards of informal family care in individual and societal decision making is discussed. We conclude that the family burden of domiciliary fetal monitoring does not exceed the family burden of in-hospital monitoring.

4. Does domiciliary monitoring allow for individualization of care; what is the balance of individualization and efficiency as viewed from the woman's perspective?

Whereas Chapter 5 supports domiciliary monitoring as the societally preferred option, the room for individualization of care is evaluated in Chapter 7. At 6-8 weeks after the delivery, women from both monitoring groups were asked to assign one valuation to the allocated (i.e. actually experienced) strategy and one valuation to the strategy not allocated (0-10 visual analogue scale). Both groups valued domiciliary monitoring on average higher than in-hospital monitoring (7.9 vs. 4.7 in the domiciliary group, and 7.1 vs. 6.2 in the in-hospital group). The valuations of domiciliary monitoring did not differ by allocated strategy (7.9 vs. 7.1). In-hospital monitoring, however, was valued lower by the
women allocated to domiciliary monitoring (4.7 vs. 6.2, p=0.02). 67% of the women preferred domiciliary monitoring, 28% preferred in-hospital monitoring and 5% was indifferent. Although the majority prefers domiciliary monitoring and domiciliary monitored women have stronger preferences for that modality, these results are probably affected by selective participation (an over-representation of women preferring domiciliary monitoring), selective response (an over-representation of women in the domiciliary group) and asymmetric treatment experience. The substantial variability in individual valuations supports individualized decision making. The clinical study design is responsible in part for biased valuations as selective participation and asymmetric treatment experience are typical of randomized clinical trials in which patients hold prior preferences and the preferred strategy can only be obtained through trial participation.

Chapter 8 displays a theoretical framework on the meaning and measurement of ‘non-health’ outcomes. Chapter 7 has shown that women have preferences for either strategy although health outcomes are equal. Preferences must therefore be based on ‘non-health’ outcomes or so-called ‘process utilities’. ‘Non-health’ outcomes are conventionally not incorporated in cost-effectiveness analysis or clinical evaluations, nevertheless their role in decision making may be important and we believe there is no theoretical objection to exclude them. Besides measurement, we discuss several options as to how ‘non-health’ outcomes may be incorporated in cost-effectiveness analysis. Either they may be incorporated in the costs-side (in which case they should be measured in terms of money) or they may be included in health effects-side (in which case they should be measured in terms of the respective health effects). Both options have been tried in Chapters 9 and 10.

In Chapter 9, we have quantified the value of the ‘non-health’ outcomes associated with the individually preferred monitoring modality using the willingness to pay (WTP) method. Firstly, high-risk women and low-risk women were asked which modality they would prefer. Next, they were asked to evaluate 27 trade-offs: the preferred modality including a pre-defined payment (raised stepwise from £10 to £2125) versus the initially non-preferred modality without payment. Women were asked to indicate the preferred option (domiciliary, in-hospital, indifferent) and the strength of preference (definitely, probably, possibly). Of the participants, 67% preferred domiciliary monitoring but women preferring in-hospital monitoring reported stronger preferences (p < 0.01). After the payment had been raised, 50% of the women initially preferring in-hospital monitoring shifted preference as compared to 82% of the women preferring domiciliary monitoring (p < 0.01). As a result, median WTP was much higher in the women who preferred in-hospital monitoring (£1472 vs. £283). We conclude, firstly, that the majority of women prefers domiciliary monitoring but the minority preferring in-hospital monitoring has stronger preferences and considerably higher WTP. This finding supports a ‘flexible’ monitoring policy in which individual women may select the preferred strategy. Secondly, ‘non-health’ outcomes exist, they can be measured, and women consider them in decision-making.
Compared to Chapter 9, Chapter 10 shows an alternative approach to the valuation of 'non-health' outcomes. Firstly, we asked high-risk and low-risk women to indicate which monitoring modality they initially would prefer, assuming a 16% temporary adverse neonatal risk, equal for both monitoring modalities. Next, they were asked to evaluate a sequence of trade-offs: the preferred modality at an increased (> 16%) adverse neonatal risk (increased stepwise from 17% to 24%) versus the initially non-preferred option at a constant 16% risk. Again women were asked to indicate the preferred option and its strength of preference. Of the participants, 59% preferred domiciliary monitoring. Preferences were stronger if in-hospital monitoring was preferred. After the adverse neonatal risk had been increased, only 28% of the women preferring in-hospital monitoring were willing to accept domiciliary monitoring; of the women preferring domiciliary monitoring, 65% was prepared to accept in-hospital monitoring. Consequently, the median acceptable risk was higher in the in-hospital preference group (24% vs. 20%). The response asymmetry may be explained by asymmetric risk perceptions or by psychological differences between groups. Although the women in both groups are risk averse, most of them are prepared to accept a temporary health disadvantage, but the extent differs considerably between groups. We conclude that different perceptions of the balance between safety and home benefits could support individualized care.

In conclusion, domiciliary antenatal fetal monitoring for a selected group of high-risk pregnancies is feasible, safe in terms of neonatal outcomes and very likely for maternal health outcome as well, efficient in economic terms, and preferred by the majority of pregnant women. Nevertheless, a substantial minority of women (30% to 40%) still prefers in-hospital care, probably because of the perceived safety. Therefore the implementation of domiciliary fetal monitoring in the obstetric system may be recommended. Individual choice with or without co-payment may be considered.

11.2 General discussion

11.2.1 Methodological aspects

In this section we present some general findings with respect to the methodologies applied in this thesis. The first section below discusses the outcome definition and the problems associated with description and statistical testing in case different outcomes exist that are difficult to rank or to relate to each other. The second section is about one particular class of outcome which appeared to be relevant in decision making but which is conventionally not included in health outcome evaluations: 'non-health' outcomes or 'process utilities' (as opposed to conventional 'health outcome' utilities). The topic of this section is how to adapt the economic evaluation framework as an aid to societal decision making in order to include these non-health outcomes. The final section is devoted to individual rather than societal decision making and the way how preference
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measurement can aid of individualized decision making. The conceptual and practical approach in this thesis appeared complex; in particular the 'correct', 'representative' patient appeared difficult to identify.

1. Outcome in obstetrics

In our study, the primary outcome measure was the neonate's health status, viz. the Prechtl score as a short term proxy of neurological and, ultimately, developmental outcome. The primary outcome was part of a more complex design in which multiple outcome measures existed. Initially, we tested the Prechtl scores and conditional on that result we tested maternal health status while an (underpowered) check was carried out for neonatal mortality. Hence, the multiple outcome framework showed at least two difficulties:

1. Morbidity and other effects play a role, apart from mortality;
2. Both the mother as well as the child are separate stakeholders of outcome, a situation characteristic for obstetrics.

In fact, the definition of outcome we applied was even broader as we included the effects on the most significant family caregiver (family burden). From a general point of view, two options are available to combine heterogeneous outcome aspects (see 1) and multiple patients or stakeholders (see 2) if a comprehensive evaluation is the purpose. One option is to weight multiple aspects or stakeholders in terms of one composite measure. A conditional analysis is the other option.

Weighting implies the application of some aggregation rule. Regarding multiple outcomes, the trade-off between morbidity and mortality (i.e. the QALY-approach and its variants\(^2\)) is a well-known example. The sum of QALYs can be interpreted as a composite measure of outcome. Regarding multiple stakeholders, we are not aware of an explicit weighting procedure other than group decision making.

The second option, conditional analysis, implies the definition of a hierarchical set of comparisons with one outcome being compared at a time. If the first outcome comparison gives no clear difference, the second outcome measure is evaluated, and so on. In the context of clinical evaluations, such a hierarchy is available and indeed we have applied a conditional analysis to overcome the multiple outcome/multiple stakeholder problem: mortality overrules morbidity unless mortality risks can safely be ignored or unless morbidity is very severe. A hierarchy for multiple stakeholders is also available: the mother's interests prevail over the child's unless infringement of her interests is unlikely or negligible, and the interests of the family are subordinate to the interests of the child.

The conditional analysis we applied closely resembles the conditions imposed by clinical practice. In other cases, however, one might prefer a QALY-like approach to combine outcome aspects within one stakeholder or an alternative hierarchy of outcomes in the conditional analysis. For example, in a recent study on severe pre-
eclampsia/HELLP treatment, the equivalence of maternal outcome is determined first, before the child's outcome and family burden are assessed. In this case the high prevalence of severe maternal morbidity justifies its primacy.

2. Non-health outcomes

Apart from the design to manage the health outcomes of multiple stakeholders, we also defined and measured the non-health outcomes for the pregnant women. These non-health outcomes were thought of as an additional outcome alongside conventional mortality and morbidity measures. Table 1 presents the general evaluation format for evaluations in obstetric care. It may easily be adapted to accommodate the needs of other clinical settings or special contexts.

<table>
<thead>
<tr>
<th>Patient/Stakeholder</th>
<th>Health outcome</th>
<th>Non-health outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mortality</td>
<td></td>
</tr>
<tr>
<td>Pregnant woman/mother</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Fetus/infant</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Family ('significant others')</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Table 1. The general format for evaluations in obstetric care

Our primary aim was to investigate as to how to account for these non-health outcomes in the context of economic evaluation (cost-effectiveness analysis, cost-utility analysis). As this issue emerges in many health care evaluations as well, we here discuss some of the theoretical and practical findings (see Chapter 8 for details) on the concept and measurement of non-health outcomes.

The discussion on non-health outcomes requires a view on the context of economic evaluation and cost-utility analysis (CUA) as its most prevalent framework. The aim of CUA is to support (not: replace) societal decision making on alternative health care options. More precisely, the decision to be supported in its most simple form is the choice between two mutually exclusive options A and B for a circumscribed patient group. A secondary aim is to achieve goal-oriented efficiency across many such medical choices. The ideal is to compose an optimal health care insurance package which does not exclude health care below a defined efficiency threshold (a quantified cost-utility ratio) and roughly excludes all options above that threshold.

The dominant tool to arrive in QALYtopia is CUA, an adaptation of the cost benefit framework, an accepted model to evaluate allocative efficiency. The adaptation is embodied in the definition and valuation of health outcome. In CUA, health outcome is defined and valued in terms of QALYs (based on mortality and health-related quality of life) and not in monetary terms. Consequently, the cost-utility ratio (unit: incremental
costs per QALY gained) is the universal measure of efficiency. Obviously, not all factors that in reality influence societal decision making are accounted for in CUA (e.g. ethical considerations, equal access, convenience, patient preferences). Although their importance for decision making has been acknowledged, so far these additional factors have have been ignored in CUA.

At least three options seem to exist to incorporate non-health outcomes in decision making:

- to treat non-health outcomes as an investment, a (negative) cost to be valued; in that case we should consequently try to 'project' non-health outcomes on the costs. The inclusion of productivity losses as indirect costs in CUA may support this view;
- to treat non-health outcome as something like a health benefit; consequently we should aim to 'project' non-health outcome on the health-related effects (usually QALYs);
- to leave them outside the CUA framework and incorporate them in a conditional analysis which declares the CU-ratio as irrelevant if the non-health outcome exceeds some threshold. This approach mirrors the conditional analysis mentioned above.

In this thesis we have explored the first two approaches. We presented pregnant women hypothetical choices between two treatment options with relevant non-health outcomes for the majority of women. Some favored monitoring at home, others preferred monitoring in the hospital, even if we emphasized that the choice would not affect health-related outcome. We experimentally added a disutility which could be calibrated to the initially favored option: a financial co-payment to arrive at a monetary value for the non-health outcomes or, alternatively, a temporary morbidity risk for the child to obtain a health equivalent. By raising the quantity of this disutility which women had to accept in order to obtain or adhere to the favored option, we tried to measure the co-payment and health risk, respectively, where the valuation of non-health outcomes equalled the hypothetical disutility. To avoid cognitive overload, we did not ask the reverse, viz. offer a compensatory payment or reduced health risk to accept the initially unfavored option.

Our experiments showed that these two approaches, methods derived from applied QALY-measurement techniques, offered fairly good estimates on the importance of non-health outcomes. Particularly the willingness-to-pay method performed well. However, from a theoretical point of view, the projection of non-health outcomes on the health-related effects may be preferable.

As mentioned before, non-health outcomes deserve a position in the CUA-framework in specific cases. Whether they should be included in the cost-side (valued with the willingness to pay method) or in the health outcome side (i.e. valued using a trade-off between health and non-health effects) cannot be concluded at this stage. Regardless the valuation method, a societal definition of admissible non-health outcomes is required. More empirical research is required to solve the issue on the appropriate valuation technique.
3. Individual versus societal preference

In the preceding sections, we elaborated the application and potential modification of CUA to account for some generally accepted non-health outcomes. In this section, we return to decision making in the clinical setting to advice a specific treatment to a specific patient. We reduce this question as to how to find out the patient's preference to support his/her personal decision, assuming that sufficient information is available and can be communicated effectively. Note that at least two elements differ from societal decision support. Firstly, the options in the clinical setting are always restricted to options relevant for that patient and that patient's condition. Considerations on across-patients and across-diseases efficiency are irrelevant here. Secondly, within this individualized context, it may safely be assumed that patients do not consider the costs involved, given the broad health insurance coverage in most Western countries.

Why is individual preference measurement relevant?
Despite, perhaps, low generalizability of individual patient preferences, the measurement of patient preferences may be relevant for several reasons. Particularly in the case of small differences between health outcomes and in the case of free choice between alternatives with unequal health outcomes, this information may be helpful (1) to support individualized decision making in the clinical setting, (2) to check patients' acceptability of new health care modalities to be implemented, and (3) for capacity planning purposes. The scientific measurement of patients' preferences for these three purposes also matches the general policy of acknowledging, more explicitly than before, patients' wishes ('tailored care'). Such a participation of patients in decision-making can best be supported by validly retrieved information on what option a well-informed patient ultimately selects, and what factors are decisive for that choice.

In our study, the clinical choice was between antenatal fetal monitoring in the hospital or at home, two options with negligible differences in clinical outcome of mother and child. Moreover, it could be expected that after the study period free choice would be allowed between these monitoring options, despite the cost differences. (Ironically, until now only the most expensive option is available). An extra argument for us to measure patients' preferences was mere interest in the actual attitude of pregnant women regarding domiciliary versus in-hospital monitoring, given the dominant view that all women prefer the own personal home environment for reasons of convenience. Taken together, the measurement and analysis of patients' preferences seemed timely and justified.

How can patient preferences be measured?
Generally several approaches exist. In our medical context the measurement of 'revealed' preference, i.e. the observation of real world decisions and changes of decisions is not possible: medical choices, like the ones in our study, are irreversible. Hence, we used stated preferences (expressed opinions or hypothetical judgments)
instead, a conventional approach in this situation. Here, we face two conceptually different approaches, both of which have been covered in this thesis.

Firstly, we can define preference as the response on a scaling-task. Mutually exclusive options are separately presented and a response technique is used to elicit a valuation that represents the ‘absolute’ strength of preference. The ordering of the resulting valuations is used to establish the ordering in terms of preference: the one with the highest valuation is the preferred option. This approach is applied in Chapter 7. The definition and consequent operationalization of this approach should be distinguished from an approach which simultaneously values options by a direct (pairwise) comparison.

Alternatively, preference is defined as the response on a choice or trade-off task consisting of mutually exclusive options. Some response technique is applied to elicit general preference and preferably its strength. Here, preference is used to obtain valuations. This approach has been applied in Chapters 9 and 10.

Who should be asked for his or her preference?
The last two decades have shown a shift towards the acknowledgement of the patient as the primary bearer of his own interest (except for some obvious situations). This has been translated into a dominant role of patients in empirical studies in search of preferences for treatment options. The general argument in favor of patients themselves as the judge of relevant choices, is that only he/she has full experience, including all difficult to document and represent aspects, and only he/she can account for the ultimate outcome (either positive or negative), resulting from the exposure to an option. In all our studies, preference information was retrieved from what could be regarded as ‘relevant’ pregnant women.

The choice between two monitoring options: the results
From a scientific point of view, our studies confirm the experiences by others who claim that under rigorous psychometrical and medical control, respondents (patients, laymen, physicians) can express their instantaneous preference in a reproducible, valid way. Supportive arguments are: feasibility (low numbers of missing values and withdrawals), high consistency across methods within and among respondents and, finally, consistent relations with known determinants. These findings – see also Chapters 7, 9 and 10 – support continued application of preference methods in this class of individualized decision support.

Our studies do not allow a firm choice between absolute measurement followed by comparison (Chapter 7) versus direct comparison providing a relative measurement (Chapters 9 and 10) as within one study both approaches were not compared. The first approach, absolute measurement, is more vulnerable for effects of implicit stimulus-differences. Also learning effects are a matter of concern. The advantage of this approach is its convenience when more than two relevant options exist and more generally the vast experience (compared to the other approach) with this type of measure-
ment. On balance, we prefer the second approach: a direct comparison of the two alternatives on relevant characteristics contributes to a deliberate decision as to whether accept or reject one alternative and, consequently, reject or accept the alternative option.

One operational choice deserves special consideration. It regards the framing of stimulus or response in terms of 'gain' or 'loss'. This issue emerges on many occasions. Regarding response, in Chapters 9 we measured preferences by willingness-to-pay for the preferred option. It is uncertain that results would have been the same if we had measured willingness-to-accept with financial compensation to the non-preferred option. Other studies show asymmetry; a forced co-payment does not equal the compensatory payment for the non-preferred option.

Although we based our stimulus (option) description on the expressed relevance of aspects by high-risk women, the paper and pencil presentation of a stimulus puts some limitation to the comprehensiveness of its content. Therefore, interpretation of results always has to consider the non-specified aspects, which experienced patients could have in mind. Future research should explore multi-media like presentations of stimuli.

A major finding of our studies was the difficulty in defining the 'right' patient. This appeared to cover two issues. The first issue is the limited own experience of a patient. Is a woman who already lost a child due to intra-uterine death a better respondent than a primiparous woman with uneventful pregnancy in judging the pros and cons of high-risk monitoring options?

Even more important is the contrast between the ex ante and ex post preference (Chapter 7). These preferences are not the same; particularly in case of low risk for high impact complications ex post views of the unlucky can deviate strongly from the average opinion before and from the 'lucky' opinions. There is no simple way out of this problem, and we still doubt which information should be used in the individualized treatment case.

One might consider to select a potential patient (somebody who may face the decision) rather than one actually facing the decision or one already experiencing the consequences of a choice. If we compare individual preferences with societal (CEA-based) preferences, then ex post preferences are the more obvious candidate as the CEA calculus is based on realizations of cost and effects after the treatment. However, it seems difficult to justify from the autonomy principle point of view, to reset the ex ante patient's preference with information that is available from adapted post treatment patients.

Our experiences point to another problem in using information from randomized clinical trials for future clinical decisions. Respondents and non-respondents most likely have different preference structures: those participating have different views on the 'subjective' loss involved in receiving the non-preferred option (a 50% probability in our case). If the options involved in a randomized clinical trial appear clinically equivalent and patients afterwards in the usual care setting may freely choose among options, the experiences and preferences of randomized clinical trials patients may not provide
a fair estimate on the average benefits for all patients. Particularly if trials suffer from low accrual rates, or if under current care arrangements only one of the options is available, this bias may be substantial.

**Conclusions on individual preferences**

We think that in current practices (Utrecht, Amsterdam) our study data were and are supportive for individual decisions; they also have shown helpful for the implementation of this new home monitoring option and for the planning of monitoring capacity.

In spite of their utility, we regard the acquisition and interpretation of these data not self-evident and straightforward. In particular, the ‘right’ patient and the ‘right’ moment are difficult to define. Due to this ambiguity, it will always be easy to cast doubt on results acquired. For this and other reasons, we think individual preference measurement to be useful but only in addition to conventional clinical outcome measures.

**11.2.2 Aspects of implementation**

1. **Model of de-institutionalized care**

Most studies on domiciliary monitoring have used hospital-based transferred care (‘outreaching’) as the model of de-institutionalization. Specialistic care is provided at home by the hospital but the secondary caregiver remains responsible for the patient management. ‘Outreaching’ is the obvious choice in the context of an experimental study in which the obstetrician is responsible for the management of patients who would otherwise have been hospitalized. In our study, only two hospital-based research-midwives were involved; primary caregivers (e.g. GPs or community midwives) were informed on indication but did not participate actively. The local home help organization was informed if home help was requested. Continuity of care could therefore be attained easily by thorough information exchange between the monitoring team and the specialist in the hospital.

This does not imply that ‘outreaching’ is the preferred model when domiciliary monitoring is actually implemented. Basically, this depends on who will ultimately be responsible for the management of high-risk women at home, and who will actually execute domiciliary surveillance. As domiciliary monitored woman are still at high-risk, it is likely that the obstetrician will also remain responsible in the future. ‘Combined care at home’ and ‘outreaching’ are therefore the preferred models.

The ‘combined care at home’-model in which professionally independent primary caregivers (community midwives) are involved, may need clear tasks and responsibilities and presumably generates more information exchange and coordination to maintain continuity. Furthermore, the caregiver who executes the surveillance should have expertise on high-risk pregnancies, specialized knowledge of pathologic pregnancies with their clinical expression, and be familiar with the interpretation of cardiotoco-
graphic traces and lab tests. It is uncertain whether community midwives want to be involved in the care of high-risk women and adopt the expert's role. This might change in the future when collaborative obstetric organizations between primary and secondary care also provide domiciliary monitoring. The 'outreaching'-model, on the other hand, is appealing: responsibilities are clear, the communication lines are short and coordination overhead is modest. Domiciliary fetal monitoring could then be fit in the *Index of Medical and Obstetric Risks*, a Dutch nation-wide accepted guide for referrals from primary to secondary obstetrical care.

The trend to hospital mergers, creating hospitals operating on a regional base offering a wide range of specialist care, will be supportive to the implementation of domiciliary fetal monitoring. However, the concentration of clinical care may increase the travel distance from patient's homes to the hospital, such that domiciliary home care based on clinical 'outreaching' may become unfeasible. This might raise opportunities for the 'combined care at home'-model.

### 2. Inclusion criteria

Domiciliary monitoring should only be accessible to pregnant women at (high-)risk for fetal distress for whom fetal heart rate monitoring is indicated. We included the following high-risk pregnancies in our study (1992): pregnancy-induced hypertension with a diastolic blood pressure < 110 mmHg with or without albuminuria; suspected impaired fetal growth after 32 weeks of gestation, confirmed by ultrasonographic biometry; pregnancies complicated with insulin dependent diabetes after 36 weeks of gestation; pregnancies complicated with non-insulin dependent diabetes after 40 weeks of gestation; post term pregnancies (gestational age of at least 42 weeks of gestation); and pregnancies complicated with prelabor preterm rupture of membranes after 33 weeks of gestation, without premature contractions and/or symptoms of infection.

Due to new scientific evidence and changing views on clinical surveillance and fetal monitoring, two of these criteria are no longer eligible for antenatal fetal monitoring and bed rest. Nowadays, post term pregnancies and pregnancies complicated with non-insulin dependent diabetes after 40 weeks of gestation are no longer hospitalized for fetal monitoring. Induction of labor is offered in these pregnancies unless induced labor is unfavorable (technically) or if the pregnant woman explicitly declines induction (patient's preference). As approximately 40% of the included women fell within these criteria, the cost reduction will be slightly less favorable when domiciliary monitoring is implemented.

Although professionals more or less agree on the criteria for clinical surveillance and fetal monitoring, these are predominantly determined on empirical grounds. The lack of evidence-based criteria leads to practice variation in the management of pregnancies at high risk for fetal distress. Some clinics prefer a more expectant role, others adopt a more intervention-oriented approach. Practice variation might hamper the dissemination of domiciliary monitoring on a local/regional scale.
The inclusion criteria will probably also change in the future following new scientific evidence or insights. We therefore recommend the evaluation of the management of high-risk pregnancies and the criteria for CTG-based fetal monitoring in order to develop national practice guidelines. Unjustified inclusion of pregnancies which otherwise would not be monitored in the hospital should be avoided: medicalization is increased and hospital resources are not saved.

3. **Domiciliary self-monitoring**

A variant of domiciliary monitoring, not tried in our study but occasionally applied in foreign studies, is domiciliary self-monitoring. Such a program could be advantageous: travelling time and costs and the time a monitoring midwife/nurse spends to home visits are reduced considerably. However, there are potential disadvantages. Firstly, women need to be instructed on the proper handling of the equipment, storage and telephonic transmission of the traces, and this may be problematic. (Dawson et al. found that 4% of the included women could not be instructed). Moreover, improper instruction or operation of the equipment might reduce the quality of the recordings, e.g. signal loss. Secondly, women may not accept the increased responsibility of unsupervised self-monitoring, or they may perceive feelings of unsafety when professional face-to-face surveillance is absent.

We regard unsupervised self-monitoring as too hazardous at this moment. Appropriate antenatal fetal surveillance of high-risk pregnancies requires more professional competence than only the recording and transmission of fetal heart rates. Home visits by professional caregivers are necessary for the evaluation of clinical signs and symptoms preceding or accompanying fetal or maternal distress. As such, supervised domiciliary monitoring is part of the obstetrician's responsibility to provide appropriate (quality of) care. Although a formal comparison of supervised domiciliary monitoring versus unsupervised self-monitoring is currently absent, we expect that supervised surveillance enhances support, confidence and feelings of safety.

4. **The implementation process**

The Dutch Working Group on Research of Quality (WOK) has developed a conceptual framework on the implementation of new care modalities, guidelines and organizational innovations. The framework origins from field work in general practice. Procedures for change are based on the scientific validity of the innovation, reliability of the results, clinical applicability, and acceptance of the innovation in practice. According to the model, implementation consists of four consecutive stages: orientation (caregivers and institutes are informed and confronted with the innovation), insight (understanding the consequences of the innovation and the differences compared to conventional care), acceptance (a positive attitude toward the innovation advances willingness to change), and change (the actual implementation).
Our study did not correspond with the framework. Within the context of an evidence-based study, the first three stages were taken as one with the selection of a well-defined innovation and the organization and conduct of the study. The fourth stage (actual change) did not correspond with the model either. The conduct of the trial in real practice – though protected by financial and legal arrangements of a clinical experiment –, also implied the exploration of barriers that have to be overcome once the trial has ended. The conduct of a clinical study therefore implies, in a sense, ‘pre-implementation’. An evidence-based approach may shortcut the four-stage model as implementation may already start with the collection of evidence.

We conclude that the WOK model, with its emphasis on attitudinal change, may be successful for innovation in general practice but it seems less appropriate as a general framework on professional change. Barriers outside the field of general practice deserve elaboration (e.g. legal, professional, insurance), particularly if the innovation involves changes across professions and/or health sectors. As ‘pre-implementation’ is essential to discover these barriers, we advocate the use of the research setting as the start of implementation. A potential disadvantage of this approach may be the difficulty to reverse changes once the innovation proves to be inferior compared to conventional care.

**Conditions for implementation**

Before domiciliary fetal monitoring can be implemented successfully, several conditions have to be fulfilled. Regarding the pregnant woman, housing conditions should be suitable with facilities for telecommunication, the woman should be able to have bed rest and support of informal caregivers should be available and if necessary professional family support should be arranged.

The caregiver’s main tasks are the selection and inclusion of women, visiting the women at home, evaluate maternal health, record and transmit the fetal heart rate traces and evaluate the maternal and fetal condition with the obstetrician in the hospital. During office hours the caregiver has to be available for calls and questions of the patients at home, out of office hours the obstetric or delivery department should be accessible to patients for questions. In hospital capacity should be available if the fetal or maternal condition at home deteriorates.

The number of eligible patients depends on the range of the region referring to the hospital and number of pregnant women referred to the hospital. The number of eligible women should be large enough to operate efficiently. However, the range of the region could pose logistic restrictions such as reduction of transport delay in case of emergency admission and the reach for daily home visits.

5. *Cost-effectiveness and financial coverage of domiciliary monitoring*

The introduction of domiciliary monitoring reduces costs from $3558 to $1521 per woman (−57%) (Chapter 5). Here, the cost estimates include all economic resources
(personnel, material and a share of fixed capital equipment and overhead) devoted to the production of health care. The interpretation of the cost reduction is that de-institutionalization releases more (value of) hospital-based resources and means than is absorbed by the implementation of domiciliary monitoring. Two remarks should be made on this result.

Firstly, de-institutionalization in our case is efficient because it produces a net gain. The resulting net release of resources (spare hospital capacity, empty beds) can be devoted to other patients who need hospital care more urgently than the high-risk pregnant women. The release of resources would amount to approximately 90–95 antenatal hospital admissions, and 830 hospital days each year. Neither the health gain associated with treatment of these ‘new’ patients, nor the increased nursing intensity due to the changed case-mix are included in the cost-effectiveness analysis. Nevertheless, these effects may be relevant when health care providers decide upon the implementation of domiciliary monitoring.

Secondly, regarding finance, the ‘value of resources released’ as measured by the cost reduction may differ from the ‘financial means released’. The greater part of the ‘true economic costs’ is embodied in departmental and hospital personnel, facilities, equipment and overheads. Much of the financial means embodied in these hospital resources are unaffected if patients – particularly a small group like ours – are de-institutionalized. Hence, the incremental financial benefit of substituting high-risk patients is probably low whereas the incremental expense needed to put domiciliary monitoring in operation – probably small-scale with high start-up costs – is likely to be high. This situation obviously complicates de-institutionalization.

This raises questions as to how to finance new hospital-at-home programs in a health care system in which financing is predominantly sector-oriented and capacity-based. Hospital admissions are an influential budget financing parameter but hospital admissions are not saved as domiciliary monitored women should give birth in the hospital. In-patient days, however, are saved but the budget financing parameter is negligible as a source to fund domiciliary monitoring. Currently, there is no official fee to reimburse (short-stay) domiciliary monitoring. Introducing such a fee would increase out-of-hospital budgets without decreasing hospital budgets and therefore violate budget-neutrality. This questions whether hospital capacity and budgets can be maintained at their current level in the longer run.

The implementation of domiciliary care is seriously jeopardized if care is transferred from hospital to home but the corresponding financial means are not. The current financing structure and the large share of capacity-based costs in health care pose considerable adverse incentives to the implementation of small-scale de-institutionalization programs, particularly if several health sectors or health care professions are involved.9 This contradicts previous recommendations that adverse financial incentives should be absent.
6. The place of domiciliary fetal monitoring in the obstetric system

Our study shows that there is no contraindication for the implementation of domiciliary fetal monitoring. Actual implementation requires the determination of inclusion criteria, the development of practice guidelines, and the definition of responsibilities between caregivers. Antenatal in-hospital surveillance will always co-exist for several reasons. Firstly, selected high-risk pregnancies with an indication for in-hospital surveillance for safety reasons will not be eligible for domiciliary monitoring. Secondly, domiciliary monitored women must be hospitalized if the maternal or fetal condition deteriorates. Thirdly, some women will definitely decline domiciliary surveillance.

An alternative to domiciliary monitoring, daily fetal monitoring at the outpatient ward, is currently evaluated at the Academic Medical Center, Amsterdam, The Netherlands. Clinical surveillance is limited to pregnancies at high risk for maternal or fetal distress for whom an intervention is expected in the short term. Surveillance of high-risk pregnancies at the outpatient ward is accessible when daily fetal and maternal monitoring are indicated and women are fully mobile. Domiciliary monitoring will be offered preferably to high-risk pregnancies with an indication for daily fetal and maternal monitoring who have restricted mobility. Pregnant women with an indication for fetal monitoring in either of these two schemes are followed prospectively to assess satisfaction and quality of care.

In the future, the high-risk status primarily determines whether or not a pregnant woman is hospitalized for clinical surveillance. The preferred setting for women eligible for de-institutionalized surveillance, either at home or at the outpatient ward, depends on women’s physical mobility. Free choice could be considered if domiciliary monitoring is recommended but the pregnant woman prefers clinical surveillance. A co-payment that (partially) compensates the increased costs of hospitalization might be asked, similar to low-risk women who prefer to deliver in the hospital (short-stay).

Co-existence of the above mentioned modalities will allow individualization of care and contribute to increased patient autonomy and decreased medicalization with room for individual decision making. With the opportunity to reserve expensive clinical surveillance for the most at risk pregnancies, effectiveness and efficiency of the obstetric care system will increase.
SUMMARY AND GENERAL DISCUSSION

References


