

This is a postprint version of the article published as:

Thorsen, K & Johannessen, A. (2022). How gender matters in demanding caring for a spouse with young-onset dementia: A narrative study. *Journal of Women & Aging*, 35(1), 81-97

Thorsen, Kirsten.^{1,2} Johannessen, Aud.^{1,3}

¹ Vestfold Hospital Trust, Norwegian National Advisory Unit on Ageing and Health
Tønsberg, Norway

² Oslo Metropolitan University, Oslo, Norway

³ Faculty of Health and Social Sciences, Department of Nursing and Health Sciences
University of South-Eastern Norway

Corresponding author: Aud Johannessen

Telephone: (+47) 97547979

E-mail: auj@usn.no

Postal address: Postal adress: Postbox 235; 3603 Kongsberg

Short running text: How gender matters in caring for a spouse with young-onset dementia

Authors' contributions

Both authors contributed to this project and read and approved the final manuscript.

Conflicts of interest declaration

The authors declare that there are no conflicts of interest or competing interests.

Funding

No.

Purpose: To examine narrated experiences of the meaning and content of spousal caring for partners with frontotemporal dementia analyzed from a gender perspective.

Method: A qualitative study using narrative interviews with ten wives and six husbands.

Findings: Both husbands and wives provide intensive care, and spousal caring preserves partners' shared existence and symbolizes the nature of love. Care is influenced by gendered caring norms, and experiences and mastery vary between husbands and wives. The study finds marked differences in the meaning, content, and sustainability of care, and needs for support vary. Wives endure more stress longer, with a greater emotional impact and negative health consequences, and their needs are more easily neglected. Husbands presented their needs more efficiently and obtained public relief earlier.

Conclusion: Public support for people with dementia and their spousal caregivers must be based on the gendered meaning and efforts of caring among husbands and wives, and the impact on stress and burden. Women may need more support earlier during different stages of caring. Spousal caregivers need person-centred support to live their own lives and preserve their selves.

Keywords: family caregiving; husbands; meaning; sex; spouse; wives; young-onset frontotemporal dementia

Introduction

Sustainable private care – with the most extensive efforts provided by spouses – is pivotal in enabling persons with dementia to remain at home for a long time. However, more knowledge is needed about what makes caring meaningful and manageable over time. What triggers transitions to higher levels of care and spouses to consider seeking support from public care? What is the tipping point when private care can no longer be sustained and the person with dementia is transferred to a nursing home? Are these points different for husbands and wives? More knowledge is needed to understand how caring processes between spouses develop over time and what sustainable and meaningful care looks like, taking into consideration gendered norms for caring.

The syndrome of dementia is an umbrella term for various progressive cognitive diseases, without treatment for the basic brain diseases. However, medication may modify symptoms (Engedal & Haugen, 2018), and care may improve well-being (WHO, 2020). Being diagnosed with a progressive lethal disease is life-changing for the person and the entire family, who foresee years with increasing caring challenges and transformed relationships (Johannessen, Engedal, Haugen, Dourado, & Thorsen, 2019; Johannessen, Helvik, Engedal, & Thorsen, 2017; Thorsen, Dourado, & Johannessen, 2018). A caregiver's experiences of stress caring for a person with dementia can have physical, mental, social and financial aspects (Balla et al., 2007) that may increase the risk of depression and anxiety (Etters, Goodall, & Harrison, 2008; Schulz & Beach, 1999).

Dementia is increasing in prevalence as people live longer and is typically considered an 'old age disease'. However, some people develop dementia at a rather young age (< 65 years old) when the disease is unexpected, little known and late recognized and diagnosed (Draper et al., 2016; Johannessen et al., 2017; van Vliet, 2012a; van Vliet et al., 2012b). The implications of dementia for individuals and their families are different when the disease occurs in the middle of life rather than late. In Norway, with a population of five million, the incidence of dementia in the age group 65 years and younger is estimated to be about 4500 (Kvvelo-Alme, Bråthen, White, & Sando, 2019; Zhu et al., 2015). The most common types of dementia among younger people are Alzheimer's disease (YO-AD) and frontotemporal dementia (YO-FTD). The syndrome implies symptoms such as lack of initiative and withdrawal, loss of inhibitions,

failing ability to solve problems, and disturbances in verbal ability (Engedal & Haugen, 2018; Pasquier & Petit, 1997). Frontotemporal dementia among younger people appears in nearly equal distribution in men and women (Onyike & Diehl-Schmid, 2013). In Norway and elsewhere, people with YO-FTD do not represent a large group, but they and their families encounter great challenges (Gentry et al., 2020; Haugen, 2012; Johannessen et al., 2017).

Compared to people with YO-AD, those with YO-FTD are more likely to experience personality changes and to have less insight into their situation. Moreover, those who care for persons with YO-FTD are often more distressed than other carers due to the patients' aberrant behaviours and denial of problems (Kimura et al., 2020; van Vliet, de Vugt, Bakker, Koopmans, & Verhey, 2010). The condition of younger persons suffering from dementia will have a significant impact on their entire family, characterized by increasing caregiving tasks, stigma, and greater risk for anxiety and depression, especially among the spouses of YO-FTD (Caceres et al., 2016; Hvidsten et al., 2019; Johannessen, Engedal, & Thorsen, 2016a; Kaiser & Panegyres, 2006; Nunnemann, Kurz, Leucht, & Diehl-Schmid, 2012). Problems and distress are found not only in the later stages of the dementia but also in earlier stages and during the process of establishing a diagnosis, especially a YO-FTD diagnosis (Draper et al., 2016; Johannessen et al., 2017; Massimo, Evans, & Benner, 2013; Rasmussen, Hellzen, Stordal, & Enmarker, 2019; Riedijk et al., 2008; van Vliet et al., 2010). The diagnosis of YOD is complex and challenging to establish (O'Malley et al., 2019), and this has been documented for frontotemporal dementia as well (Rosness, Haugen, Passant et al., 2008).

Review studies have established that there is a paucity of research focusing on carers' experiences and efforts to cope with life caring for a person suffering from frontotemporal dementia (Cabote, Bramble, & McCann, 2015; Caceres et al., 2016; Nunnemann et al., 2012). To the best of our knowledge, there is even less research on the experiences of spouses of persons with YO-FTD. A requirement is to increase knowledge about spouses' situations to be able to meet their needs for support. How their experiences, wishes and needs differ by gender present important questions.

Factors such as gender, relationship, personal qualities, culture, and norms for family care will impact on caregivers' experiences of the situation and their ability and capacity to provide care for persons with dementia (Etters et al., 2008). One study demonstrated that women, both

wives and daughters, experience a greater sense of burden in the role of caregiver to a person with dementia (all types) compared to husbands and sons (Chappell, Dujela, & Smith, 2015). The lowest self-esteem is felt by wives, followed by husbands, and then sons and finally daughters, who have the highest self-esteem. Female caregivers have a higher level of anxiety and more depressive symptoms than men and are sometimes less satisfied with the situation (Erol, Brooker, & Peel, 2016; WHO, 2012). A good two-way relationship with the person with dementia increases the likelihood of satisfaction for the caregiver and that the person needing care is valued as an individual and not seen mainly as a problem or burden (Andren & Elmstahl, 2005). However, the lack of insight and empathy characteristic of people with YO-FTD can hinder a good two-way relationship and communication.

A review of gender differences among spousal caregivers for persons with dementia found that most studies do not differentiate between caregivers of various patient groups/diseases and/or between different caregivers, their gender and kind of relationship (Arbel, Bingham, & Dawson, 2019). None of the studies focussed on spouses of people with YOD or on specific subgroups, for example, people with YO-FTD. The review pointed to a paucity of qualitative research since quantitative research findings related to gender differences cannot reveal underlying processes. This trend appears to be continuing despite studies that, for a long time, have underlined that caregiver subgroups differ significantly and, thus, should be studied as homogeneous groups (Carlson & Robertson, 1993; Harper & Lund, 1990). The norms for caring as well as the circumstances will differ. Findings of analyses of gender in specific groups are vital for specificity and equity in health policies and practice, and necessary for tailoring services to meet the needs of individuals with dementia and the caring spouses.

Dementia develops over a long time, with gradual changes in symptoms influencing caring relationships, which change radically when the person with dementia moves periodically or permanently to a nursing home. It is a process with both continuity and discontinuity (Atchley, 1989). What those involved in caring experience in these transitions, as well as their impact, may vary over time and be perceived differently by family carers, husbands and wives. experience,

Developments in stress theory have focused on the *meaning* of stresses (Burke, 1991; Lazarus, 1993; Lazarus & Folkman, 1986; Thoits, 2010; Yang & George, 2005), such as changes in health and disabilities. The extent to which stressors impact individuals depends on how they assess the *meaning* of difficulties – in our case, the burdens, challenges, and rewards of caring for a family member with progressive dementia. Various approaches emphasize the importance of appraising stresses and their salience for identity. The salience to the individual depends on the role domain in which they occur. Thoits (1995) suggested that the more important one's identity is, the more threatening the experience of stressors related to that identity will be. The gender-related meaning of caring for family members, the appraised stresses and the impact on identity will have consequences for resilience and care duration (Robinson, Bottorff, Pesut, Oliffe, & Tomlinson, 2014).

Gender is not a fixed category. As internalized and individually expressed, gender has biographical, biological, cultural and social sources, with constellations that change over time and with geographic and cultural variations. Theories about gender focus on intersectionality; gender intersects with factors including social class, region, religion, and culture (Bowleg, 2012).

As a modern and prosperous welfare state, Norway has equal rights and opportunities for men and women as a main ideological foundation. A 2020 ranking on equality between genders places Norway at the top of the list (Female Opportunity Index, 2021). Nevertheless, the country has a rather paradoxical situation, with a highly gender-divided labor market, especially in the healthcare sector, among nurses and subordinate healthcare workers (Norwegian Statistics, 2021), with physicians as exceptions. The study is based on a re-analysis of data material on experiences of caring for a spouse with YO-FTD. The purpose of the study is to re-analyze the data with a gender perspective. A general thematic analysis of the material has been presented in a previous article (Johannessen et al., 2017). We therefore wanted to examine narrated experiences of the meaning and content of spousal caring for partners with frontotemporal dementia analyzed from a gender perspective.

Method

The study represents a narrative qualitative approach (Wertz, 2011) to spouses of people with YO-FTD during the progression of the disorder. The method has the advantage of exploring the lifeworld of the participants and the ways they experience and narrate their feelings and cognitions as recalled in dialogues. A narrative method is designed to elicit stories and events (Mishler, 1986). People's narratives are 'a way of shaping an order of their experiences, a way of understanding one's own and others' actions, of organizing events and objects in a meaningful whole, of connecting and seeing the consequences of actions and events over time' (Chase, 2011, p. 421). Narrative interviews are open, based on listening intensively for both obvious and more-hidden meanings, invite reflections and reasons, and evolve during the dialogue. The stories may reveal many layers of meaning, referring to different relationships and situations. Narratives are, thus, "thick descriptions" of phenomena (Geertz, 1973).

Participants

With the intention of increasing heterogeneity, 17 spouses and one cohabitant of a person with YO-FTD were asked to participate. The persons with YO-FTD were at different stages of the disease. All participants were informed about their partner's diagnosis by a psychiatrist or geriatrician at a memory clinic. One spouse declined to participate in the study. The 16 participants comprised 10 wives, 5 husbands and 1 male cohabitant. In the following text, the spouses and the cohabitant are all denoted as spouses. They were recruited from one municipality dementia team, seven memory clinics and a nursing home designed to provide care for people with YOD. Characteristics of the participants and the persons with YO-FTD are described in Table 1. The included persons will be referred to as spouses, husbands/wives, men/women, and participants to vary the wording.

Insert Table 1 about here

The amount of time that had elapsed since diagnosis was confirmed varied from 0 to 11 years. The period during which the spouses at first recognized that 'something was wrong' *before* the diagnosis was confirmed varied from 1 to 11 years (Table 1). Thus, the total period of symptoms and illness varied from 2 years to 13–15 years. As indicated in Table 1, the exact time of onset was often difficult to establish. At the time of the interviews, five of the ill

spouses had moved permanently into nursing homes (three husbands and two wives), and one husband had died four months before the interview after a period in a nursing home.

The interviews

A convenient place for each participant was chosen for the interviews (Denzin & Lincoln, 2011; Kvale, 1983; Kvale, 1989). Twelve of the interviews were conducted in the participants' homes, two at the interviewer's workplace, and two in a municipality office. The interviews took place in 2014 and 2015. The second author conducted the interviews, which lasted from 31 to 79 minutes (mean = 57). A professional typist transcribed the tape-recorded interviews verbatim within two weeks. The interviewer conducted a quality control check by listening to the tapes while reading the transcriptions. The interviews were based on an interview guide with six open-ended thematic questions that focussed on the spouses' experiences of the development and progression of the dementia and on having a spouse with YO-FTD (Table 2). Depending on the replies, the aspects and ideas raised by the participants led to further questions to obtain additional information.

Insert Table 2 about here

The analysis

The analysis was conducted in line with the qualitative analysis outlined by Corbin and Strauss (2008). They emphasized that their modified methodological method is fruitful without the intention of developing a theory and suitable for exploring and developing new knowledge in fields little examined and researched. First, to get an overview, we condensed every interview into an 'abstract', showing the salient points in the narratives, especially the gender-relevant elements. The first analytical step in the method was 'open coding', that is searching with an open mind for themes or categories relevant to the research question – the gendered aspects of caring. We related the search to our preconceptions and knowledge about gender differences in caring, as recommended by Corbin and Strauss (2008). Then we examined the narratives of men and women separately and searched for subgroups among these narratives. Subcategories and variations were noted. We applied the constant comparative approach (Kolb, 2012). Agreement on categories were reached through

discussions and reflections among the authors. The researchers continuously related findings to the other empirical material, analyzing it ‘vertically’ by going back and forth in the narrative of each participant and ‘horizontally’ by comparing the narratives of men and women, and various subgroups. During the ongoing analytical process, our analysis returned to the total material to relate our findings in these groups to the experiences of all husbands and wives.

Initially, we present in detail two condensed cases (Graneheim & Lundman, 2004), one husband and one wife, who have experienced the transfer of a spouse to a nursing home. Studies of cases comprise more detail, richness, completeness, and variance – that is, depth – for the unit of study than do cross-sectional analyses (Flyvbjerg, 2006). In our study, ‘a case’ is an individual personal story about caring, in line with the medical and psychological conceptualization of ‘a case’. These cases demonstrate how caring is a process that evolves over time. Thereafter, the analytic presentation expands to the other persons in the ‘transition’ groups. Then, we analyze variations and commonalities in caring experiences among men and women in the total material.

Ethics

The study adhered to the ethical guidelines outlined in the revised Declaration of Helsinki (World Medical Association, 2013) and those of the Regional Committee for Ethics in Medical Research, Southern Norway (number X) and approved by the Norwegian Data Protection Authority (number X). The participants received both oral and written information prior to the study and gave written consent before they were interviewed.

Findings

The analysis revealed that the transition to a nursing home is one of the most stressful decisions of the caregiving experience and the one that most clearly exposed differences in reactions among men and women.

Husbands’ stories about relocation of their wives

Erik works as an administrative leader, a demanding job. He sensed that ‘something was wrong’ with his wife about two years ago. The signs included memory failure, lack of

concentration, an alcohol problem, difficulties understanding time and confusing day and night. She had no insight regarding her situation and did not admit to having any problems. She resisted being examined by her general practitioner (GP), whom she did not trust. Erik visited a private clinic and explained the situation: ‘The doctor took all the tests I asked him to take. I am rather clever to find professional assistance’. But all tests were negative. The main problem was getting an appointment with his wife’s GP, whose secretary refused since he was not a patient there. He then contacted the administrative leader of the clinic, who immediately understood the case and lectured the GP about YO-FTD. Thereafter, she underwent an MR-scan and was examined at a memory clinic. He stated, ‘I had to put pressure on the process all the time, I felt. All the time!’ The examination resulted in his wife’s diagnosis of YO-FTD, which had already progressed quite far; an outcome Erik had anticipated. He asked for a treatment plan and sought support. Whether she could remain at home seemed uncertain as her husband felt he had to be very strict and controlling. He experienced that it was ‘immensely demanding’ to coordinate care and support with the dementia team. He worked from home and paid female family members, a sister and cousin who were both healthcare workers, to care for her if he had to travel for his work.

The next problem Erik encountered was keeping his wife from driving the family’s car. Her GP informed him that she was unfit to drive ‘and *you* have to stop her!’ Erik commented, ‘All the time I tried to say that it is not *my* responsibility, *you* must take the responsibility, but nevertheless it became *my* responsibility. Really, all the time! My wife became terribly angry. It was all my fault.’

He learned to distract her and tell her ‘white lies’. He went through a difficult and challenging work period when he was working mostly at home. He noted, ‘Then I was a home nurse for her. All the time she was around me, clinging to me’. An offer of relief several days a week at a farm failed. He stated, ‘My wife is not a “group-person”’.

The coordination of public assistance became very demanding. He said, ‘At last I had to tell the administrative nurse “*you must* help me with the coordination. I can’t call four different instances every time something shall be arranged!”’ The nurse agreed and gave him more

information. We noted his emphasis in stating: ‘You *have* to help me’, indicating a man used to giving orders and being in command. As summer ended, he said to the health administrator, “This can’t go on. It is of no use! I have the best information (...). It is my wife, see?” In the interview, he stated:

So, I have been ahead of them and have put a lot of pressure on them. At the end, it was *I* who said: “Now we *must* do something else; we can’t just sit and observe if this and that function. I *must* have relief!” After much discussion, we should try to get my wife into a home for a relief stay. The best solution would be a place where she could later stay permanently.

Reminding her of her memory problems, she agreed to an observation period at the relief ward, but when the day arrived, she refused. Then he pushed her, stating, ‘You *have* to see the doctor!’ At their arrival, they met six or seven administrative personnel, and thereafter, a nurse led her to the relief ward. Erik said, ‘Then she agreed to go behind the locked door, and she has not been outside since then. I think that is rather special! I had no choice, really. I could not do it any other way’. Observing her advanced stage of dementia, the nursing home’s administrative team recommended a permanent stay. However, several of the healthcare personnel do not seem to realize how ill she really is. Her husband said, ‘It is annoying that they only see her in short glimpses and say, “This seems to go very well; she just went out walking.” She is physically fit. (...) They always give me some advice . . . “do this and that”... and I just know it is not so’.

He misses contact with people who show empathy and sympathy for his situation, someone who would listen to his experiences: ‘It is not to believe. Very few call me and just ask: “How are you?” It is incredibly few. Nearly no one’.

He is also planning and arranging for her well-being in the nursing home by organizing some of her daily activities and visits from family, seeing that she gets exercise and opportunities to walk outdoors, presented as a weekly schedule. He remarked, ‘I have struggled. (...). I have concluded that the most important [thing] for her is to feel safe and have competent care (...).

Her room is her safe place now... her world'. He summarizes his approach to mastering the problems he has:

I am very proactive. In my job I have had a lot of experience. (...) I am rather used to settling things and finding people who can answer my questions. So, I can navigate in the system. However, it is not easy. It is just not easy! (...) In critical situations, I just become calm and ask: "OK. What to do next?" Then we do this. It is no point that also I get stressed (...). I have struggled.

Saying farewell leaves, him in a depressive mood: 'I have to say to myself: "This is the way it is just now. (...) The emptiness!' Then he adds, 'Maybe she does not feel as miserable as I have felt'. At the end of the interview, he summarizes: I have done the work. I am used to speaking out. I have pushed for it (the public care), and then it has started rolling'. Erik's story represents a man who feels he has done his best. He had no choice. He has no regrets and no guilty conscience.

Tom stated that, after a period when he did not understand what was going on, he decided to keep a journal to inform the GP that 'something was wrong', and the GP became convinced. Tom stated, 'He was very clear and direct, so open that I nearly lost my balance'. The GP told him, 'This is a terrible disease. It is difficult to handle and a great strain for the family'. At the hospital where his wife's assessment took place, he was told: 'When you can't stand it any longer, just tell us. You have to apply for a nursing home early and get help where you can'. The diagnosis was confirmed in May, and in early autumn, the local administration quickly mobilized assistance, in-home help and daily visits by an in-home nurse. He emphasized: 'For me, it was no alternative not to work!' In the spring, he applied for a place in a nursing home. His wife was offered a respite leave in August, and this quickly turned into a permanent stay. He does not regret this or doubt that he has made the right decision, and he is confident that his wife is receiving the best available care from a competent staff. He appreciates his new life:

Then, suddenly getting the freedom to do things on your own and start a new life – it was wonderful. A relief! Really! Not having responsibility for an ill person any longer. I was so ready for it; I had no problems letting her go'.

Tom has sought and established contact with a new female friend.

Both Erik and Tom demonstrate highly purposeful, rational and efficient ways of mastering their challenging situations by applying for and receiving nursing home placement for their wives.

Female stories about relocation of their husbands

Anne's husband received his diagnosis when he was in his early fifties. During the two years before the diagnosis, there 'was much quarreling'. He was gradually demoted at his workplace and went from a leading position to being almost a warehouse worker. When she wondered what was happening, he just answered, 'I don't know'. At last, the wife of one of his colleagues told Anne that her husband was seriously ill, which resulted in a medical consultation and a sick leave. The first tentative diagnosis was 'burnout'. But after an assessment at a memory clinic, YO-FTD was confirmed, and the doctors estimated that he might have been ill for about five years before the diagnosis was established. She remarked:

Looking back, I recognize that something was wrong, but I did not understand it (...). He has always been excellent in economics. Whatever it was – loans, buying a house, I never had to think. He fixed it, but then the reminders came, reminders and reminders, with [late payment] charges. I had to take [control of] affairs and spent two years settling them. No one said to me, "Let us make a plan for repayment". (...) I thought it was very difficult to take over, really. I have learned a lot.

With great effort, Anne had to learn to handle loans, pensions like disability payments and sickness benefits, and insurances. Her husband wandered about at night, causing great anxiety for the family. His behavior when shopping 'was horrible'. The total workload for Anne was heavy, demanding and stressful:

I missed someone to share the work with, like repairing the garage and the house. All the "male jobs". Furthermore, I did absolutely everything at home, and I [also] had to

look after him, to see that he got washed, got food and went where he should go. It was like having a kid.

At the time of the interview, he had been in a nursing home for six years. She commented that if he had been offered a relief stay at an adult daycare center, she could have kept him at home longer: 'But I *had* to work, somebody had to earn money'. Staying at home alone, he did not eat or go to the toilet. When Anne came home after work, she had to change all his clothes. He would be strolling along, and when people offered him a lift home, he would just say, 'I am waiting for her'. Finally, she applied for a permanent place in a nursing home with competent staff and a special ward for people with YOD.

I was excited about this place. I worked hard for it, did not give in. I did not want for him any other place. I think I spent two years on the application process, but you know, when the message came, "Take it or leave it, we need your answer now", then I had problems. I got a guilty conscience and said, "I have to think about it". They gave me one more day. I conferred with my children, and they said: "You *have* to take it, this may be the only chance!" I wept bitterly but thought, "I *have* to do it!" However, I kept him at home for another month. I was not able to let him go. It was incredibly difficult for me.

Anne expressed that she regrets having received no professional help throughout the process from people who were experienced in such decisions and relocations. She commented, 'You know, I had [an] extremely guilty conscience. I thought I was mean, since he was not that ill; he was just not able to take care of himself'.

Guilt and a bad conscience afflicted her for a long time and were aggravated by their farewells: 'When I visited him, and I did that very often, every time I should go, he wanted to go with me! I cried and cried. (...) I felt very bad and wicked'. She repeated that, at this stage, she needed someone to support her. She became terribly tired, both physically and mentally.

When asked how she has experienced the last years, she stated, 'I am a widow, but not on paper. It has been hard. I feel it has been very, very hard'. She continued: 'I am very lucky. I have a lot of friends; they see to me and are marvelous. But I have been depressed, and I think it [life] looks dark'. She described her former married life as 'very good'; her husband had been kind and considerate. She misses 'having someone around', and said, 'Everything became empty and quiet, and I have many dark moments, anxiety (...), especially during the dark season. I often feel lonely, and I have a constant bad conscience'. She characterized herself as 'a type who tries to pull herself together and seek help'. After a period of visiting her husband very frequently, she collapsed mentally and received psychological therapy. She commented, 'At the hospital, they thought I had experienced too much stress, both working (full time) and visiting. However, I have learned'.

Now she visits her husband at the nursing home once a week. The trip is nearly an hour's drive, and then she stays four or five hours after her workday. His dementia has progressed, and he is no longer able to follow her to the door when leaving. Earlier, the nurses had to restrain him and drag him back. 'It was very painful', she said. He has now been transferred to another ward for patients with serious dementia. Anne stated:

I am sure that he is well there. This is the right place for him. Absolutely. His language and conversational ability are greatly reduced, he mimics sentences, only "yeah" and "no" remain. But he still remembers all the songs of his favorite band!

Anne's story testifies to the intensively experienced guilty conscience of a wife when transferring a spouse with dementia to a nursing home, feeling guilt both when she applied for a place and afterward when visiting him.

Lise expressed having strong guilt-ridden reactions when her husband was transferred to a nursing home. It happened rather dramatically after an episode where he incessantly drank water and then 'vomited all over the place'. This incident was the tipping point. She called the health administration, explained the acute situation, and claimed, 'This can't go on!' About 15 minutes later, *Lise* heard back from the health administration: 'I got the message that they would come for him and they said, "He will not return tonight"'. In fact, he did not return

home thereafter. She stated, 'It was incredibly hard for me. What had I done?' At that time, she went to a psychologist who comforted her and supported her. She advised her not to visit him every day, emphasizing, "You can't do that!"

(...) It helped. I should at the same time be working full time and be a mother for two children. But the bad conscience remained. I should have been there all the time. He should not have had to live there. You got drawn in all directions. It is incredibly exhausting, physically and mentally.

She observed other spouses who visited their husbands every day, and she praised her psychologist who advised her not to do so and to take care of herself. She said, 'It was a great help because I am not good to take care of myself'. Now, she visits him once a week. Like Anne, Lise emphasized the support she has received from the personnel at the nursing home and others: 'They have persisted saying, "Now you shall live your own life. *We* shall take care of him"'.

By contrast, *Carol*, who also experienced transferring her husband to an institution, described having no guilty feelings and explained her reaction. Her husband had exhibited aberrant behavior for 15 or 16 years and had intermittent periods when he stayed at psychiatric wards. After he moved into a nursing home two years ago, 'then everything went smoothly', she said. She further expressed:

I have been so fed up with everything that has happened. I am not like other young women who lose their husband to a nursing home, for I had lost him many years ago. So, the difference is great! Many have said, "You may get a crack!" I have answered, "If I did not crack up earlier, I will not do it now!"

She commented on the very long time she had lived with a husband with deviant behavior: 'You can't just move out (of the home). I know that men do. A great many is my impression. *They* can move out, while we women have a conscience, you know!'

Caring for the spouse at home

Differing caregiving periods and loads

Husbands and wives alike provide extensive, challenging and exhausting care for their spouses, and it becomes more intensive and absorbing in the later stages of the disease process. Their efforts aim at preserving quality of life to the extent possible. The aspects of the YO-FTD disease inflict great emotional strain. We have found that gender matters markedly regarding the period of providing caring at home and for the meaning and impact of the care.

The signs and symptoms of their spouses' dementia were estimated by the women to have appeared a mean of 5 years *before* the diagnosis and 4.25 years after diagnosis, for a total of 9 years. The corresponding numbers of years given by the men were 3 years before diagnosis and 2.5 years after, for a total of 5.5 years. Testifying the challenges of combining caring and working – both at the job and at home – are the spouse's working position. Of the 10 women, only two (20%) were working full time; the rest were on disability benefits (three; two full time and one part time) or sick leave (five). All of the women had been in the workforce before the dementia disease struck their husbands. The corresponding numbers for the six men were three (50%) working full time, one on disability benefits, one on sick leave and one retired. The mean age of the women was 58.3 years and 61.7 years for the men. Thus, the women were relatively more often outside the workforce, partially or totally, than the men, even if they were younger at the time the disease struck. The women explained that combining a job with their caregiving responsibilities became too exhausting and problematic or reduced their work commitment. One woman remarked, 'I had to say "No, thanks" to jobs, to some positions I could not [take]. So, my work was influenced. You see, I always had to be home after work to look after him'. Of the three women – the youngest – who experienced that their spouse moved into a nursing home, one continued to work full time, which she had done during the whole caregiving period; as said, 'I needed the money'. The other two are on sick leave.

Distancing: Experiencing a transformed spouse and relationship

For some, the memory problems were the initial signs. One spouse stated, 'I did not understand why he was unable to receive information and all the time said, "Call me!" when

he should do something'. Other signs involved orientation problems, especially while driving. Most remarkable was the transformation of personality. Both men and women experienced a great transformation of their relationship to their partner, interfering with all aspects of partnership: emotions, working roles, solidarity, and cooperation, among others. The overriding trend in the relationship was *distancing* moving from a partnership initially based on love to experiencing that the spouse with dementia gradually became quite another person and that the feelings for the spouse changed.

A frequently narrative is that the spouse changes from being reasonable and agreeable to becoming irritable and more aggressive, sometimes even violent. Small frustrations stir disproportionate outbursts. One wife reported:

[Over] The last years I have seen that something is wrong. He has always been a person avoiding conflicts. He has never wanted trouble, never been mean. Yes, a very calm person. Now he has started scolding me. "Why do you bark at me?" I replied, "You are not kind at all!"

An important transition in the spousal relationship came with the diagnosis. 'Suddenly everything fell into place', remarked a woman. The new information changed the spouse's response set:

Now, I try to laugh at things, now when I know what it is. Try not to nag all the time. (...) I can't bother about trifles. But for me it is not trifles. When I said something, I felt he did not bother to listen to me. It just passes through, in a way.

The spouse gradually seems more disinterested in the partner, often also in the children and grandchildren, and withdraws from communication: 'I asked myself, is he not interested in me any longer?', wondered a wife. The spouses with dementia seem to 'drift away'.

A wife described being, for a long time, the victim of verbal complaints and criticism: He is so grumbling and difficult, has no self-insight. (...) My husband says that nothing of what I say is true. All the time he means just the opposite of me. (...). He says he does not ail anything. He has never felt as well as he does now.

Another one stated:

Every little detail is wrong. He says, “When I am home, I am the boss.” (...) If you stay two minutes too long in the shower, *that* is wrong. If the dinner is ready 10 minutes after he wants it, *that* is wrong, or the other way around. Small everyday things that you never consider, but he *must* have something to criticize.

Some husbands became very aggressive. One wife reported, ‘We get death threats. He has no barrier on what he says when he gets angry’. After an aggressive incident, one wife stated, ‘Emotionally, I fell into the basement and took sick leave for a week. Then I went to his doctor’. The incident was the tipping point for her; her mental health was suffering, and she could not tolerate the situation any longer.

At later stages of the disease, the spouses must direct and supervise all everyday activities. Some women compare their spouse to a child, sometimes their own children or grandchildren. Anne remarked, ‘It is awful. Not fun at all, directing one in his fifties. It is easier to explain to my granddaughter – five years old; at least she remembers what I have told her. He forgets immediately. It is completely surreal’. Another wife made this comparison:

I have one son, and I sense I have the same feelings for my husband that I had for my son as a kid. I felt that I was the only one that was able to give him what he needed, and I am there now.

Only women used the metaphor ‘like a child’ to describe a husband with YO-FTD; no men used this term to describe a wife with the disease. Also, the wives were described as getting ‘angrier’, they became irritated more easily.

The changes in the personality of the spouse with YO-FTD are summarized by expressions such as ‘quite the opposite of his/her former self’, ‘completely changed’, and ‘quite another person’. One wife exclaimed, ‘This type of dementia he has, I would not wish for my worst

enemy'. Another remarked, 'I don't think they really understand that the spouses suffer much, living with such a disease. So, it seems'.

Gradually, the caring spouses no longer recognized the former identity of their partners. During the progression of the dementia, the spouse's reaction pattern also changed, and new aspects of his or her own personality emerged. A wife said, 'It has been frustrations. I have felt like a person who is always nagging, nagging, nagging. Then he strikes back, terribly irritated'.

One wife summarized her caring conscience and role in the family, where she also wanted to care for her younger family members:

So it is very often that I put others before myself. I suppose it is a classical female thing. Doing something for my children and grandchildren means very much to me. (...) An then I myself come as the very last [person] on the priority list.

As demonstrated, the women tolerate more and endure more for a longer time than the men.

The spouses also spoke about how they experience becoming foreign to each other and being lonely together. A loving relationship gives way to emotional distance when their partner's dementia progresses, and at the same time, they have to become closer because of the caregiving situation. At home, a stressful situation for many spouses is the constant surveillance of their spouse and the feeling of incessant contact – often described as clinging – when the spouse is unable to concentrate. The intimacy required for care develops at a cost to their loving intimacy. They feel that their own selfhood, their own personality, 'shrinks' without confirmation from their partner.

Distancing from others

The social isolation experienced by the caregiver is exacerbated, especially for the women, by being more outside the workforce than the men. The contact with colleagues is a welcome

distraction from caregiving responsibilities and confirms the healthy spouse's other capacities beyond care and concern. Working life is experienced as a 'time of relief' when they are occupied by thoughts and tasks other than caregiving. However, for all spouses, a great worry is what their spouses do when they are left alone. Furthermore, the working load may be felt to be too great.

The spouses' isolation and loneliness are often exacerbated by the lack of understanding from others. Gradually, outside contact is reduced. Caregiving is time-consuming and requires great effort, leaving little space and energy for contact with others. Arranging relief is complicated and sometimes not worth the effort when new worries arise. As shown, caregivers also miss *genuine* contact with others, someone asking them 'how they really get on'. Others' interest is felt as shrinking, lacking or being based on misunderstandings. Potentially increasing the distance from others, even close family members, is the lack of knowledge about YO-FTD. Sometimes, even healthcare personnel underrate the seriousness of the disease because they do not know enough about YO FTD and may be deceived by an image of a fit and healthy-looking younger person.

Needing assistance and relief

All participants have needed increasing levels of assistance and relief and have mentioned that these have come too late and have been too little. As demonstrated, the caring load also implies great administrative efforts to try to get the support needed. We have seen that some of the men explicitly underline their abilities to get more information and 'push' the system. Women seem less insistent; they wait longer before they apply for help and longer before they receive it. If it does arrive, they are more grateful for any support and relief they are given.

A woman appreciated the advice about being nice, calm and agreeable that she got from a psychiatrist:

Don't try to explain if there are things he does not like or disagree with. Whatever happens, you should be quiet and controlled, use a gentle voice, even smile if you don't feel like it. Just put on a smile and say nicely, "So it is. And there is nothing more to discuss."

She added, ‘Such advice you don’t find on the Internet’.

One aspect of the caregiving spouse’s problems is being too ‘bounded’, lacking time for their own interests and ‘time for being themselves’. ‘Free time is very difficult. Extremely difficult’, exclaimed a husband. They need relief. The caregiving men longed for holidays on their own; they mentioned fishing and hunting and other appreciated activities; the women just wanted ‘time out’ to relax and recover.

Several of the spouses described having received, at some time, advice and administrative support from *dementia coordinators*. Among the relief measures, assistance from a *support contact* is most appreciated since when it is individually tailored to the interests of the spouse with dementia and adapted to his or her schedule. At later stages, *respite relief* a few days a week at a daycare centre or intermittent weekly stays at a nursing home have been needed and were valued. However, a few discovered that their spouse, especially wives, was not ‘a group person’. Others found that the activities offered were not interesting for younger people, especially men who are still fit and interested in outdoor activities. Some, like Anne, mentioned that her spouse could have remained at home longer if they had been able to receive respite relief earlier.

At a later time, the difficult question arises of how to suggest that the spouse should spend some time at a nursing home. One husband greatly needed relief but would not dare to suggest to his wife a respite stay at a nursing home. He said, ‘I think she would conceive it as I would put her away or throw her out of the house. She would react very, very negatively if this came up, and our everyday life would be much worse’. Another husband did receive an intermittent relief stay weekly in a nursing home; his wife described the transition process:

The first week when he should leave for relief, he denied. Then it was *war!* It was very tough! From the first moment he opposed, did not understand why he should be together with those “half-dead”. He found no companion, no one to talk to. He sat in his room looking at TV programs.

As shown, moving a spouse into a nursing home can be experienced as the most critical transition, the end of the partnership and a shared life. Some of the women expressed needing support during the transition process. Even a short relief stay at a nursing home may cause intense feelings of guilt and sorrow. Saying farewell has great symbolic power, even at short visits in care facilities. The wives were grateful for getting the advice that they should take care of themselves and for the assurance that the nursing home would take good care of their spouse. All the caregiving spouses expressed a desire for more support and relief – more hours and more days.

Discussion

We conducted a study of one of the most vulnerable groups of caregivers, those who, at a younger age, are providing care to spouses with progressive YO-FTD, an incurable disease that transforms personality, memory, ability, insight and awareness and is difficult to detect and diagnose (Engedal & Haugen, 2018; Rasmussen et al., 2019; Rosness, Haugen, & Engedal, 2008). Spousal caregivers of people with YO-FTD have an increased risk of negative health consequences, such as depression, frailty, and cognitive and functional impairments (Caceres et al., 2016; Gentry et al., 2020; Nunnemann et al., 2012; van Vliet et al., 2010). Being a younger caregiver, many of whom are middle-aged, employed, and have children living at home and many role conflicts, adds particular challenges to the caregiving situation (Cabote et al., 2015).

A basic premise for the analysis has been that mastery of caregiving is grounded in *meaning*, related to gender-influenced norms for caring, engraved within the personality, developed over one's life history and applied to the challenges of the caring situation. Caregiving takes place within contexts – the wider family relations as well as the public support available.

The study has demonstrated that the care provided by both the husbands and wives of people with YO-FTD is absorbing and all-encompassing, in line with studies of spouses' caregiving of people with YOD (Bruinsma et al., 2020; Kaiser & Panegyres, 2006) and FTD (Massimo et al., 2013). Also, *men* are committed, capable and competent caregivers (Cahill, 2000; McDonnell & Ryan, 2013; Sanders & Power, 2009). Both male and female spouses have offered emotional, cognitive and practical support to maintain their partner's quality of life,

functioning and daily structure, and to attempt to compensate for their increasing helplessness. The narratives reveal how comprehensive and intense spousal care can be and how far this *labor of love* (Finch & Groves, 1983) can go. It is, at the same time, a *duty of love* upheld by intense feelings of guilt – especially among the wives – if they feel that their care is insufficient or not good enough. Caring for one’s spouse is seen as an inseparable part of the marriage bond, maybe even the essence of it in later years. It is supported by strong norms concerning moral duties and regulated by legal rules about moral responsibilities (Daatland & Herlofson, 2003; Daatland, Veenstra, & Lima, 2010). We have seen that this kind of care takes its toll. The caregivers in our study – especially the wives – saw their own health deteriorate, due in part to experiencing great burdens imposed by caregiving, feelings of responsibility and role conflicts, and the transformation of personality that their spouse underwent in the progression of the disease. This transformation is related to the traditional gender roles; the men are more often described as self-assertive, the ‘boss’, aggressive, even brutal and without empathy, while the women are more likely to be seen as angry, irritable, and not complying.

Studies have shown health problems to be overrepresented among female caregivers of people with dementia (Erol et al., 2016; Thyrian et al., 2016; WHO, 2012). Yet, even when care is extremely exhausting, the norms and meaning of care in marriages and partnerships further the continuation of care.

There is a great deal of literature on gender and caring regarding gender and family caregiving for persons with chronic and longstanding diseases and, more specifically, for people with dementia. Theories of intersectionality posit that multiple social categories like gender, class, race and family relations intersect; individual experiences reflect and intersect with systems on the macro level, such as norms, legal regulations, culture and religion (Bowleg, 2012).

Intersections between caregivers and recipients are often blurred and imprecise because the groups are mixed, and the causal processes and caregiving trajectories are not outlined. Several reviews have pointed to the limitations of studies of gendered differences because they lack differentiations within the male and female groups (Arbel et al., 2019; Baker & Robertson, 2008; McDonnell & Ryan, 2013). It is necessary to evaluate subgroups within the genders. Chappell et al. (2015) found that wives were the most vulnerable among family

caregivers when burden and self-esteem were considered, and the longitudinal effects of caregiving differed between husbands and wives (Arbel et al., 2019).

Psychological stress and coping theories connect appraised outer circumstances to psychological experiences. A classical definition of stress is: ‘Psychological stress refers to a relationship with the environment that the person appraises as significant for his or her well-being, and in which the demands tax or exceed available coping resources’ (Lazarus & Folkman, 1986, p. 63). *Primary appraisal* includes ego involvement and personal commitment such as self-esteem, moral values, ego ideal and ego commitment (Krohne, 2002; Lazarus, 1993). Stress and coping theories are highly relevant for studying longstanding family caregiving for people with chronic and progressive illnesses like dementia.

Care work is both *body work* (Twigg, 2000) and *emotion work* (Herron, Funk, & Spencer, 2019). The word *work* accentuates that caring is more than good intentions and attitudes. Providing care is often difficult and challenging work. Body work is work with bodies and can include washing, bathing and toileting functions. Professional healthcare workers as well as family care workers must ‘negotiate nakedness and touch, manage dirt and disgust, balance intimacy and distance’ (Twigg, 2000, p. 389). As noted in our study, at later stages of dementia, spousal caregivers must supervise, assist and later take over personal hygiene tasks such as washing, showering, and managing incontinence – what Twigg (2000) called ‘dirty work’. This intimate body work transgresses the roles and emotional and sexual relationships between spouses. As shown, some wives compared their new tasks with caring for a child, a metaphor for the transformed relationship (Thorsen & Johannessen, 2021). This connects their caregiving for the husband with the other main female role, caring for children. Aligned with other studies (Tretteteig, Thorsen, & Rokstad, 2019), we found that body work involving incontinence seemed to be a tipping point. Others were their spouse’s aggression and their own exhaustion. These tipping points (Gladwell, 2006), in the case of caregiving, cause caregivers to reframe their experiences and decide that it is ‘too much’. This moment may arrive earlier for husbands than for wives.

Emotion work (Herron et al., 2019) is described as family caregivers displaying a positive attitude, ‘a mask’, even when they feel frustration, anger and resentment. They place the person with dementia first and suppress their own genuine feelings to avoid conflict and arousing anger. They must argue ‘the right way’. These contradictory reactions – being patient and positive while suppressing their own negative feelings – generated great emotional stress for the interviewees in our study. Long-term caregiving for persons with YO-FTD will, due to the extraordinary transformation of their personality, inflict intense emotional demands on their spouses at the cost of their selfhood and identity.

We have demonstrated that the narratives of spouses who have experienced the transition of their partners into nursing homes accentuate the gendered reaction patterns. They reveal the accumulated effects of appraisals of stress on experiences and coping (Krohne, 2002; Lazarus, 1993). Becoming separated from a spouse who requires institutionalization is among the most critical transitions in later life (Robertson, 2014). The men in ‘the transfer group’ exhibited task-oriented coping and problem-solving strategies to get needed support when caring for their wife at home. They demonstrated efficient use of available resources, including other female family members. The men systematically searched for information and applied for public assistance – finally, a place in a nursing home – when they felt that their resources, patience and resilience were overstretched. The transition process was eased by feeling that there was no choice, and that the nursing home would provide better care than they were able to provide. The women in our study had been caring for their spouse longer than the men before they reached a point where they had to ‘give in’. Also, women have searched for information about relief, but the narratives indicate that the men have been offered a place in a nursing home quicker than the women. Women may seem more insecure in their wishes and push for a place less than men, while more men may signal and get accepted that their primary preference is to continue working. As noted, only two of the ten caregiving women were employed, while half of the men were. Continuing their working role has been the primary option for the men, not for the caring women. Giving up employment for caring may not be such a serious blow to women’s identity as it is for the men.

A significant gender difference in our material is that the men do not have bad conscience, feel regret or guilt for the transference of the spouse to a nursing home. In contrast, two of the

three women in the “transfer-group” had intense guilt, felt “burn out” (Truzzi et al., 2012) and experienced breakdown, and had to get psychological assistance. The third woman had no bad conscience, she felt entitled to relief and a place in a nursing home for her husband after great caring hardships for a long time. The women need and value the assurance from health personnel that they have the right to live their own life and “find themselves” (Johannessen et al., 2017). Most of the other women still caring for their spouses at home, express great stress, some narrate despair and exhaustion. Thus, a cluster of factors intersect and support the participants` gendered appraisals, reasons and choices to terminate their care at home and transfer it to an institution or continue caring at home.

Our results are in accordance with studies and reviews highlighting that caring is more dominant in female identities (Pinquart & Sörensen, 2006). A meta-analysis of sex differences in interests (Su, Rounds, & Armstrong, 2009) summarized their findings by the title: ‘Men and Things, Women and People’. Women prefer ‘people-oriented’ careers over ‘thing-oriented’ careers, and a ‘people orientation” is dominant within the caring professions.

However, overriding the gendered differences, we have shown that both husbands and wives have exhibited extensive caring for a long time and shown commitment in their caring efforts. A study of older male spouses (Cahill, 2000) caring for wives with dementia, underline that they showed a strong commitment to their role, and cared out of “love, marriage, duty and a combination of each. Tretteteig et al. (2019) ask if care by *love* is better than care given by *duty*? A case study demonstrates that even a son`s caring for a despised father with Alzheimer can be adequate and good. The son took the responsibility and arranged with his sisters a coordinated private caring plan, with different roles, depending on the sibling`s competence and resources. Duty was his motivation. In contrast, a daughter showed “loving care” by empathic caregiving, identifying with her mother, feeling limitless duty and responsibility and having problems with limits for care. Also, our study has shown that female caring with experiences of being totally absorbed – engulfed by caring and thereby losing their selves and their identity – takes a large toll in their psychic health. The appraised stress and feeling of bad conscience may overtax their resilience and cause burnout (Truzzi et al., 2012) and breakdown – they may need psychological and social assistance.

Mature care is contrasted to *altruistic care* (Pettersen, 2012). *Mature care* takes account of the interests of both parties in the caring relationship. Compassion is an aspect of *mature care*, but unlike the altruistic concept, the other-regarding feeling is not granted superior status in *mature care*. In *altruistic care* – unselfish and unconditional care – primacy is given to the interests of the other/others. Self-damage and exhaustion may be the result of altruistic and unselfish care, which will also affect other relationships and roles. Important is that the carer is not exploited. In caring for a spouse with YO-FTD, the symptoms make it less improbable that the care receiver will feel empathy and concern for the experiences of the spousal caregiver – rather the contrary.

Couples can feel that dementia affects their lives both as individuals and as a couple – dementia is a *shared* experience. A study, primarily of elderly couples facing dementia finds that they often speak in terms of ‘us’ and ‘we’ and fight to hold on to their shared existence, their ‘togetherness’ and shared identity as ‘we’ (Riekkola et al., 2019; Wadham, Simpson, Rust, & Murray, 2016). In contrast to the study of elderly couples by Wadham et al. (2016), our study of spouses of people with YO- FTD finds that less weight is given to the word *we* and more to the words *I* and *he/she*. The vocabulary is stressing individuality and increased distance between the spouses. We have shown how caregiving spouses describe the increasing emotional distance they experience from their wife or husband as frontotemporal dementia progresses. In its later stages, the disease has completely transformed their relationship as a couple – the emotional as well as the physical aspects – and their roles. The experience of *we* – with equality, communication, and respect – has gradually disappeared. The situation becomes more and more imbalanced. ‘*We*’ seems to be mostly a memory of the former marriage.

All the participants described that public support, especially adult daycare centres and the support contact, has been essential for their ability to continue to provide care. The daycare centre gave them free time to pursue their own normal life activities and interests, and breaks from their worries (Tretteteig, Vatne, & Rokstad, 2015; Tretteteig, Vatne, & Rokstad, 2017) – a better life to both caregivers and receivers. The support contact offers the spouse with dementia individualized support, flexible, adapted to their interests and resources (Johannessen, Engedal, & Thorsen, 2016b) and is felt to be “like a friend”. When the

dementia progresses, the caregiving spouses wish more days of relief and more hours a day. Additional stress has been seeking information and strivings with bureaucracy, sometimes denoted as “fights”. Even if men seem more efficient to handle it, both genders find the bureaucratic system very complicated and the coordination of the services difficult. Support and advices by *dementia teams* and *dementia coordinators* are valuable. These services have escalated in Norway in accordance with advices in three Dementia plans (2015; 2020; 2025) and have provided needed and appreciated assistance.

A trend in research on dementia caring is to focus on the *positive aspects* (Pendergrass et al 2019), in contrast to most of the research concentrating on the negative aspects (De Labra et al., 2015). Improving understanding of positive aspects could improve innovative support programs based on the positive caring experiences (Carbonneau, Caron, & Desrosiers, 2009). These experiences may be more prominent at the early stages of dementia and more relevant for some syndromes, such as Alzheimer`s, than for others, like YO-FTD. The caring impact of different dementia syndromes will differ. Foreseeing the great demographic increase in older age groups and people with dementia (WHO, 2020) the challenge will result in more private care – and more caring burden. A large Norwegian study (Gjora et al., 2021) has found that the number of people with dementia was much higher than earlier estimated. Too much emphasis on the positive aspects may overshadow that caring is also great emotional stress and hard work. Private caregivers of people with YO-FTD will be among the victims if not adequate support is provided. More private caring will be more negative for the female spouses, due to the age differences and sex-roles. Public support will also have impact on gender equality.

Public care – whether at home, in daycare centres, as part of a relief stay or permanent nursing home care – has to be person-centred (McCormack, 2003; 2004; McCormack & McCance, 2010; Sharma et al., 2015) to support the identities of both the person with dementia and their spouse, taking gender into consideration.

Strengths and limitations

Strengths of this study include the rich and candid stories of spouses of persons with YOD and their experiences of the dementia over time, their needs and support, expressed through the narrative method. Although the small sample does not allow generalization in a statistically representative way, we believe the results have wider validity relating to the fields of caring and dementia research.

A limitation is the binary gender conceptualization. There were no limitations on gender and partnership/spousal relations in the recruitment process. The binary-gender differentiated spousal sample was a result of the recruitment procedure. A larger sample might have included same-gendered couples. Our sample included one unmarried couple. However, they did not deviate from the rest in any way and, therefore, were not analyzed separately. In a larger sample, people in partnerships and with several marriages might exhibit particular characteristics.

Further research should aim to look more closely at subgroups and to be more precise in researching subgroups of family caregiving for various dementias, taking into consideration gender, age and family relationships. Longitudinal studies will contribute more knowledge about changing experiences and needs in different relationships.

Conclusion

Both husbands and wives provide committed caregiving for their spouses with YOD. The care and concern provided by spouses are influenced by gendered norms for family caregiving. We have demonstrated that women care for their spouse at home longer than men do and with greater impacts on their working career and health. Providing care is more central to women's identities as nurturers than it is to men's, and in situations where caregiving becomes a problematic and exhausting responsibility, women's identities will be threatened to a greater extent than men. However, variations exist within and between the situations of wives and husbands, as does overlapping. Public support for spouses and family caregivers will have to be person-centred and gender-related in order to support resilience and avoid burnout, especially among women.

References

- Akkerman, R. L., & Ostwald, S. K. (2004). Reducing anxiety in Alzheimer's disease family caregivers: The effectiveness of a nine-week cognitive-behavioral intervention. *American Journal of Alzheimer's Disease and Other Dementias*, *19*(2), 117–123. doi:10.1177/153331750401900202
- Andren, S., & Elmstahl, S. (2005). Family caregivers' subjective experiences of satisfaction in dementia care: Aspects of burden, subjective health and sense of coherence. *Scandinavian Journal of Caring Sciences*, *19*(2), 157–168. doi:10.1111/j.1471-6712.2005.00328.x
- Arbel, I., Bingham, K. S., & Dawson, D. R. (2019). A scoping review of literature on sex and gender differences among dementia spousal caregivers. *Gerontologist*, *59*(6), e802–e815. doi:10.1093/geront/gny177
- Atchley, R. (1989). A continuity theory of normal aging. *Gerontologist*, *29*(2), 183–190.
- Baker, K. L., & Robertson, N. (2008). Coping with caring for someone with dementia: Reviewing the literature about men. *Aging and Mental Health*, *12*(4), 413–422. doi:10.1080/13607860802224250
- Balla, S., Simoncini, M., Giacometti, I., Magnano, A., Leotta, D., & Pernigotti, L. M. (2007). The daily center care on impact of family burden. *Archives of Gerontology and Geriatrics*, *44 Suppl 1*, 55–59. doi:10.1016/j.archger.2007.01.008
- Bowleg, L. (2012). The problem with the phrase *women and minorities*: Intersectionality – An important theoretical framework for public health. *American Journal of Public Health*, *102*(7), 1267–1273. doi:10.2105/ajph.2012.300750
- Bruinsma, J., Peetoom, K., Millenaar, J., Köhler, S., Bakker, C., Koopmans, R., . . . De Vugt, M. (2020). The quality of the relationship perceived by spouses of people with young-onset dementia. *International Psychogeriatrics*, 1–10. doi:10.1017/S1041610220000332
- Burke, P. J. (1991). Identity processes and social stress. *American sociological review*, 836–849.
- Cabote, C. J., Bramble, M., & McCann, D. (2015). Family caregivers' experiences of caring for a relative with younger onset dementia: A qualitative systematic review. *Journal of Family Nursing*, *21*(3), 443–468. doi:10.1177/1074840715573870
- Caceres, B. A., Frank, M. O., Jun, J., Martelly, M. T., Sadarangani, T., & de Sales, P. C. (2016). Family caregivers of patients with frontotemporal dementia: An integrative

- review. *International Journal of Nursing Studies*, 55, 71–84.
doi:10.1016/j.ijnurstu.2015.10.016
- Cahill, S. (2000). Elderly husbands caring at home for wives diagnosed with Alzheimer's disease: Are male caregivers really different? *Australian Journal of Social Issues*, 35(1), 53–72. doi:10.1002/j.1839-4655.2000.tb01303.x
- Carbonneau, H., Caron, C. D., & Desrosiers, J. (2009). [Development of an adapted leisure education program for persons with dementia.] *Canadian Journal on Aging*, 28(2), 121–134. doi:10.1017/s0714980809090114
- Carlson, K. W., & Robertson, S. E. (1993). Husbands and wives of dementia patients: Burden and social support. *Canadian Journal of Rehabilitation*, 6(3), 163–173.
- Chappell, N. L., Dujela, C., & Smith, A. (2015). Caregiver well-being: Intersections of relationship and gender. *Research on Aging*, 37(6), 623–645.
doi:10.1177/0164027514549258
- Chase, S. E. (2011). Narrative inquiry: Still in the field of making. In Y. S. Denzin (Ed.), *SAGE handbook of qualitative research* (pp. 421–434). London: Sage.
- Corbin, J., & Strauss, A. (2008). *Basics of qualitative research*. Los Angeles, CA: Sage Publications Ltd.
- Cuijpers, P. (2005). Depressive disorders in caregivers of dementia patients: A systematic review. *Aging and Mental Health*, 9(4), 325–330. doi:10.1080/13607860500090078
- Daatland, S. O., & Herlofson, K. (2003). 'Lost solidarity' or 'changed solidarity': A comparative European view of normative family solidarity. *Ageing and Society*, 23(5), 537–560. doi:10.1017/S0144686X03001272
- Daatland, S. O., Veenstra, M., & Lima, I. (2010). Norwegian sandwiches: On the prevalence and consequences of family and work role squeezes over the life course. *European Journal of Ageing*, 7(4), 271–281. doi:10.1007/s10433-010-0163-3
- De Labra, C., Millán-Calenti, J. C., Buján, A., Núñez-Naveira, L., Jensen, A. M., Peersen, M. C., . . . Maseda, A. (2015). Predictors of caregiving satisfaction in informal caregivers of people with dementia. *Archives of Gerontology and Geriatrics*, 60(3), 380–388.
doi:10.1016/j.archger.2015..03.002
- Denzin, N., & Lincoln, Y. (2011). *SAGE handbook of qualitative research*. London: Sage Publications Ltd.
- Draper, B., Cations, M., White, F., Trollor, J., Loy, C., Brodaty, H., . . . Cumming, R. G. (2016). Time to diagnosis in young-onset dementia and its determinants: the INSPIRED study. *Int J Geriatr Psychiatry*, 31(11), 1217–1224. doi:10.1002/gps.4430

- Engedal, K., & Haugen, P. K. (2018). *Demens: sykdommer, diagnostikk og behandling (Disease, diagnostics and treatment)*: Aldring og helse akademisk.
- Erol, R., Brooker, D., & Peel, E. (2016). The impact of dementia on women internationally: An integrative review. *Health Care Women Int*, 37(12), 1320–1341. doi:10.1080/07399332.2016.1219357
- Etters, L., Goodall, D., & Harrison, B. E. (2008). Caregiver burden among dementia patient caregivers: a review of the literature. *J Am Acad Nurse Pract*, 20(8), 423–428. doi:10.1111/j.1745-7599.2008.00342.x
- Finch, J., & Groves, D. (1983). *A labour of love: Women, work, and caring*: Routledge.
- Flyvbjerg, B. (2006). Five misunderstandings about case-study research. *Qualitative inquiry*, 12(2), 219–245.
- Geertz, C. (1973). *The Interpretation of Cultures*. New York: Basic Books. Inc.
- Gentry, M. T., Lapid, M. I., Syrjanen, J., Calvert, K., Hughes, S., Brushaber, D., . . . Rosen, H. (2020). Quality of life and caregiver burden in familial frontotemporal lobar degeneration: Analyses of symptomatic and asymptomatic individuals within the LEFFTDS cohort. *Alzheimers Dement*, 16(8), 1115–1124. doi:10.1002/alz.12095
- Gjora, L., Strand, B. H., Bergh, S., Borza, T., Brækhus, A., Engedal, K., . . . Selbæk, G. (2021). Current and Future Prevalence Estimates of Mild Cognitive Impairment, Dementia, and Its Subtypes in a Population-Based Sample of People 70 Years and Older in Norway: The HUNT Study. *J Alzheimers Dis*, 79(3), 1213–1226. doi:10.3233/jad-201275
- Gladwell, M. (2006). *The tipping point: How little things can make a big difference*: Little, Brown.
- Graneheim, U. H., & Lundman, B. (2004). Qualitative content analysis in nursing research: concepts, procedures and measures to achieve trustworthiness. *Nurse Educ Today*, 24(2), 105–112. doi:10.1016/j.nedt.2003.10.001
- Harper, S., & Lund, D. A. (1990). Wives, husbands, and daughters caring for institutionalized and noninstitutionalized dementia patients: Toward a model of caregiver burden. *International Journal of Aging and Human Development*, 30(4), 241–262. doi:10.2190/9BLE-FED4-BRAC-RPX3
- Haugen, P. K. (2012). *Demens før 65 år. Fakta, utfordringer og anbefalinger: Utviklingsprogram om yngre personer med demens, Demensplan 2015 [Dementia before the age of 65. Facts, challenges and recommendations: Development program*

- for younger people with dementia, Dementia plan 2015].* Tønsberg: Forlaget Aldring og helse [Publisher Ageing and Health].
- Herron, R. V., Funk, L. M., & Spencer, D. (2019). Responding the “wrong way”: The emotion work of caring for a family member with dementia. *Gerontologist, 59*(5), e470–e478. doi:10.1093/geront/gnz047
- Hvidsten, L., Engedal, K., Selbæk, G., Wyller, T. B., Benth, J. Š., & Kersten, H. (2019). Quality of life in people with young-onset dementia: A Nordic two-year observational multicenter study. *Journal of Alzheimer's Disease, 67*(1), 197–210. doi:10.3233/JAD-180479
- Johannessen, A., Engedal, K., Haugen, P. K., Dourado, M. C., & Thorsen, K. (2019). Coping with transitions in life: a four-year longitudinal narrative study of single younger people with dementia. *J Multidiscip Healthc, 12*, 479–492. doi:10.2147/jmdh.S208424
- Johannessen, A., Engedal, K., & Thorsen, K. (2016a). Coping efforts and resilience among adult children who grew up with a parent with young-onset dementia: a qualitative follow-up study. *Int Journal of Qualitative Studies on Health Well-being, 11*, 30535. doi:10.3402/qhw.v11.30535
- Johannessen, A., Engedal, K., & Thorsen, K. (2016b). Family carers of people with young-onset dementia: Their experiences with the supporter service. *Geriatrics, 1*(4), 28. doi:10.3390/geriatrics1040028
- Johannessen, A., Helvik, A. S., Engedal, K., & Thorsen, K. (2017). Experiences and needs of spouses of persons with young-onset frontotemporal lobe dementia during the progression of the disease. *Scand J Caring Sci, 31*(4), 779–788. doi:10.1111/scs.12397
- Kaiser, S., & Panegyres, P. K. (2006). The psychosocial impact of young onset dementia on spouses. *Am J Alzheimers Dis Other Demen, 21*(6), 398–402. doi:10.1177/1533317506293259
- Kimura, N. R., Simões, J. P., Santos, R. L., Baptista, M. A. T., Portugal, M. d. G., Johannessen, A., . . . Rodrigues, V. M. (2020). Young-and late-onset dementia: a comparative study of quality of life, burden, and depressive symptoms in caregivers. *J Geriatr Psychiatry Neurol, 0891988720933355*. doi:10.1177/0891988720933355
- Kolb, S. M. (2012). Grounded theory and the constant comparative method: Valid research strategies for educators. *Journal of emerging trends in educational research and policy studies, 3*(1), 83.
- Krohne, H. (2002). Stress and Coping Theories. Johannes Gutenberg-Universität Mainz, Germany. In.

- Kvale, S. (1983). The qualitative research interview. *Journal of Phenomenological Psychology, 14*(1–2), 171–196. doi:10.1163/156916283X00090
- Kvale, S. (1989). To validate is to question. In S. Kvale (Ed.), *Issues of validity in qualitative research*. (pp. 73–92). Lund: Studentlitteratur [Student literature].
- Kvvello-Alme, M., Bråthen, G., White, L. R., & Sando, S. B. (2019). The prevalence and subtypes of young onset dementia in Central Norway: A population-based study. *Journal of Alzheimer's Disease, 69*(2), 479–487. doi:10.3233/JAD-181223
- Lazarus, R. S. (1993). Coping theory and research: past, present, and future. *Psychosom Med, 55*(3), 234–247. doi:10.1097/00006842-199305000-00002
- Lazarus, R. S., & Folkman, S. (1986). Cognitive theories of stress and the issue of circularity. In M. H. Appley and R. Trumbull (Eds.) *Dynamics of stress* (pp. 63–80). Boston, MA: Springer.
- Massimo, L., Evans, L. K., & Benner, P. (2013). Caring for loved ones with frontotemporal degeneration: The lived experiences of spouses. *Geriatric Nursing, 34*(4), 302–306. doi:10.1016/j.gerinurse.2013.05.001
- Mc Donnell, E., & Ryan, A. (2013). Male caregiving in dementia: A review and commentary. *Dementia, 12*(2), 238–250. doi:10.1177/1471301211421235
- McCormack, B. (2003). A conceptual framework for person-centred practice with older people. *International Journal of Nursing Practice (9)*, 202–209.
- McCormack, B. (2004). Person-centredness in gerontological nursing. *Journal of Clinical Nursing, 13*(3A), 31–38.
- McCormack, B., & McCance, T. (2010). *Person-centred nursing: Theory and practice*. Chichester, West Sussex: Blackwell Pub.
- Mishler, E. G. (1986). *The analysis of interview-narratives*. In T. R. Sarbin (Ed.), *Narrative psychology: The storied nature of human conduct* (pp. 233–255). Praeger Publishers/Greenwood Publishing Group.
- Norwegian Ministry of Health and Care Services. (2011). *Demensplan 2015, Den gode dagen [Dementia plan 2015, The good day]*. Retrieved from: <https://www.regjeringen.no/no/dokumenter/demensplan-2015-den-gode-dagen/id663429/>
- Norwegian Ministry of Health and Care Services. (2015). *Demensplan 2020, Et mer demensvennlig samfunn [Dementiaplan 2020, A more dementia friendly society]*.

- Retrieved from: <https://www.regjeringen.no/no/dokumenter/demensplan-2020/id2465117/>
- Norwegian Ministry of Health and Care Services. (2020). *Demesplan 2025, Handlingsplan [Dementia plan 2025, Action plan]*. Retrieved from: <https://www.regjeringen.no/no/dokumenter/demensplan-2025/id2788070/>
- Nunnemann, S., Kurz, A., Leucht, S., & Diehl-Schmid, J. (2012). Caregivers of patients with frontotemporal lobar degeneration: A review of burden, problems, needs, and interventions. *Int Psychogeriatr*, 24(9), 1368–1386. doi:10.1017/s104161021200035x
- O'Malley, M., Parkes, J., Stamou, V., LaFontaine, J., Oyebode, J., & Carter, J. (2019). Young-onset dementia: scoping review of key pointers to diagnostic accuracy. *British Journal of Psychiatry Open*, 5(3), e48. doi:10.1192/bjo.2019.36
- Onyike, C. U., & Diehl-Schmid, J. (2013). The epidemiology of frontotemporal dementia. *International Review of Psychiatry*, 25(2), 130–137. doi:10.3109/09540261.2013.776523
- Pasquier, F., & Petit, H. (1997). Frontotemporal dementia: Its rediscovery. *European Neurology*, 38(1), 1–6.
- Pettersen, T. (2012). Conceptions of care: Altruism, feminism, and mature care. *Hypatia: A Journal of Feminist Philosophy*, 27(2), 366–389. doi:10.1111/j.1527-2001.2011.01197.x
- Pinquart, M., & Sörensen, S. (2006). Gender differences in caregiver stressors, social resources, and health: An updated meta-analysis. *Journals of Gerontology, Series B: Psychological Sciences & Social Sciences*, 61(1), P33–45. doi:10.1093/geronb/61.1.p33
- Rasmussen, H., Hellzen, O., Stordal, E., & Enmarker, I. (2019). Family caregivers' experiences of the pre-diagnostic stage in frontotemporal dementia. *Geriatric Nursing*, 40(3), 246–251. doi:10.1016/j.gerinurse.2018.10.006
- Riedijk, S., Duivenvoorden, H., Rosso, S., Van Swieten, J., Niermeijer, M., & Tibben, A. (2008). Frontotemporal dementia: Change of familial caregiver burden and partner relation in a Dutch cohort of 63 patients. *Dementia and Geriatric Cognitive Disorders*, 26(5), 398–406. doi:10.1159/000164276
- Riekkola, J., Rutberg, S., Lilja, M., & Isaksson, G. (2019). Strategies of older couples to sustain togetherness. *Journal of Aging Studies*, 48, 60–66. doi:10.1016/j.jaging.2019.01.005

- Robertson, G. (2014). Transitions in later life: A review of the challenges and opportunities for policy development. *Working with Older People*, 18(4), 186–196.
- Robinson, C. A., Bottorff, J. L., Pesut, B., Oliffe, J. L., & Tomlinson, J. (2014). The male face of caregiving: A scoping review of men caring for a person with dementia. *American Journal of Men's Health*, 8(5), 409–426. doi:10.1177/1557988313519671
- Rosness, T. A., Haugen, P. K., & Engedal, K. (2008). Support to family carers of patients with frontotemporal dementia. *Aging and Mental Health*, 12(4), 462–466. doi:10.1080/13607860802224334
- Rosness, T. A., Haugen, P. K., Passant, U., & Engedal, K. (2008). Frontotemporal dementia: A clinically complex diagnosis. *International Journal of Geriatric Psychiatry*, 23(8), 837–842. doi:10.1002/gps.1992
- Sanders, S., & Power, J. (2009). Roles, responsibilities, and relationships among older husbands caring for wives with progressive dementia and other chronic conditions. *Health and Social Work*, 34(1), 41–51. doi:10.1093/hsw/34.1.41
- Schulz, R., & Beach, S. R. (1999). Caregiving as a risk factor for mortality: The Caregiver Health Effects Study. *Journal of the American Medical Association*, 282(23), 2215–2219. doi:10.1001/jama.282.23.2215.
- Sharma, T., Bamford, M., & Dodman, D. (2015). Person-centred care: An overview of reviews. *Contemporary Nurse*, 51(2–3), 107–120. doi:10.1080/10376178.2016.1150192
- Statistisk Sentralbyrå [Norwegian Statistics]. (2021). *På stedet hvil: Kvinner dominerer offentlig sektor, menn privat sektor [On the spot: Women dominate the public sector, men the private sector]*. Retrieved from: <https://www.ssb.no/arbeid-og-lonn/artikler-og-publikasjoner/pa-stedet-hvil-kvinner-dominerer-offentlig-sektor-og-menn-privat>
- Su, R., Rounds, J., & Armstrong, P. I. (2009). Men and things, women and people: A meta-analysis of sex differences in interests. *Psychological Bulletin*, 135(6), 859–884. doi:10.1037/a0017364
- The Female Opportunity Index (2021). *An analysis of gender equality in 100 countries looking at advancements in female leadership within government, corporations, STEM and entrepreneurship, as well as success enablers like access to education and parental leave*. Retrieved from: <https://n26.com/en-eu/female-opportunity-index>
- Thoits, P. A. (1995). Stress, coping, and social support processes: Where are we? What next? *Journal of Health and Social Behavior*, (Spec. no.), 53–79. doi:10.2307/2626957

- Thoits, P. A. (2010). Stress and health: Major findings and policy implications. *Journal of Health and Social Behavior*, 51(Suppl.), S41–S53. doi:10.1177/0022146510383499
- Thorsen, K., Dourado, M. C. N., & Johannessen, A. (2018). Developing dementia: The existential experience of the quality of life with young-onset dementia – A longitudinal case study. *Dementia (London)*, 1471301218789990. doi:10.1177/1471301218789990
- Thorsen, K., & Johannessen, A. (2021). Metaphors for the meaning of caring for a spouse with dementia. *Journal of Multidisciplinary Healthcare*, 14, 181–195. doi:10.2147/jmdh.S289104
- Thyrian, J. R., Winter, P., Eichler, T., Reimann, M., Wucherer, D., Dreier, A., . . . Hoffmann, W. (2016). Relatives' burden of caring for people screened positive for dementia in primary care: Results of the DelpHi Study. *Zeitschrift für Gerontologie + Geriatrie (Z Gerontol Geriatr)*. doi:10.1007/s00391-016-1119-9
- Tretteteig, S., Thorsen, K., & Rokstad, A. M. M. (2019). Pårørendes erfaringer av kvalitet i helse- og omsorgstjenester til personer med demens og deres pårørende. En longitudinell narrativ studie. (Experiences of quality in health and caring services to persons with dementia and their family caregivers.) *Tidsskriftet aldring og helse (Ageing and Health)*, 4, 56–65.
- Tretteteig, S., Vatne, S., & Rokstad, A. M. M. (2017). The influence of day care centres designed for people with dementia on family caregivers – A qualitative study. *BMC Geriatrics*, 17(1), 5. doi:10.1186/s12877-016-0403-2
- Tretteteig, S., Vatne, S., & Rokstad, A. M. M. (2015). The influence of day care centres for people with dementia on family caregivers: An integrative review of the literature. *Aging and Mental Health*. doi:10.1080/13607863.2015.1023765
- Tretteteig, S., Vatne, S., & Rokstad, A. M. M. (2017). Meaning in family caregiving for people with dementia: A narrative study about relationships, values, and motivation, and how day care influences these factors. *Journal of Multidisciplinary Healthcare*, 10, 445–455. doi:10.2147/jmdh.S151507
- Truzzi, A., Valente, L., Ulstein, I., Engelhardt, E., Laks, J., & Engedal, K. (2012). Burnout in familial caregivers of patients with dementia. *Brazilian Journal of Psychiatry*, 34(4), 405–412. doi:10.1016/j.rbp.2012.02.006
- Twigg, J. (2000). Carework as a form of bodywork. *Ageing and Society*, 20(4), 389–411.

- van Vliet, D. (2012a). Young onset dementia: Characteristics and impact [doctoral thesis]. *Maastricht: Department of Psychiatry and Neuropsychology, School for Mental Health and Neuroscience, Maastricht University.*
- van Vliet, D., de Vugt, M. E., Bakker, C., Koopmans, R. T., & Verhey, F. R. (2010). Impact of early onset dementia on caregivers: A review. *International Journal of Geriatric Psychiatry, 25*(11), 1091–1100. doi:10.1002/gps.2439
- van Vliet, D., de Vugt, M. E., Bakker, C., Pijnenburg, Y. A., Vernooij-Dassen, M. J., Koopmans, R. T., & Verhey, F. R. (2012b). Time to diagnosis in young-onset dementia as compared with late-onset dementia. *Psychological Medicine, 43*(2), 423–432. doi:10.1017/s0033291712001122
- Wadham, O., Simpson, J., Rust, J., & Murray, C. (2016). Couples' shared experiences of dementia: A meta-synthesis of the impact upon relationships and couplehood. *Aging and Mental Health, 20*(5), 463–473. doi:10.1080/13607863.2015.1023769
- Wertz, F. J. (2011). *Five ways of doing qualitative analysis: Phenomenological psychology, grounded theory, discourse analysis, narrative research, and intuitive inquiry*: Guilford Press.
- WHO. (2012). *Dementia. A public health priority*. Retrieved from: United Kingdom: <http://www.who>
- WHO. (2020). *Dementia*. Retrieved from: www.who.int/news-room/factsheets/detail/dementia
- World Medical Association. (2013). Declaration of Helsinki. Retrieved from: <http://www.wma.net/en/30publications/10policies/b3/index.html>
- Yang, Y., & George, L. K. (2005). Functional disability, disability transitions, and depressive symptoms in late life. *Journal of Aging and Health, 17*(3), 263–292. doi:10.1177/0898264305276295
- Zhu, X. C., Tan, L., Wang, H. F., Jiang, T., Cao, L., Wang, C., . . . Yu, J. T. (2015). Rate of early onset Alzheimer's disease: A systematic review and meta-analysis. [Erratum appears in *Ann Transl Med*. 2016 May; 4(9):E4; PMID: 27275503]. *Annals of Translational Medicine, 3*(3), 38. doi:<https://dx.doi.org/10.3978/j.issn.2305-5839.2015.01.19>