Objective: To investigate the efficacy of 3-D printed bone models as a tool to facilitate initiation of bisphosphonate treatment among individuals who were newly diagnosed with osteoporosis.

Design: Fifty eight participants with estimated fracture risk above that at which guidelines recommend pharmacological intervention were randomised to receive either a standard physician interview or an interview augmented by the presentation of 3-D bone models.

Main outcome measures: Participants’ beliefs about osteoporosis and bisphosphonate treatment, initiation of bisphosphonate therapy assessed at two months using self-report and pharmacy dispensing data.

Results: Individuals in the 3-D bone model intervention condition were more emotionally affected by osteoporosis immediately after the interview ($p = .04$) and reported a greater understanding of osteoporosis at follow-up ($p = .04$), than the control group. While a greater proportion of the intervention group initiated an oral bisphosphonate regimen (alendronate) (52%) in comparison with the control group (21%), the overall initiation of medication for osteoporosis, including infusion (zoledronate), did not differ significantly (intervention group 62%, control group 45%, $p = .19$).

Conclusion: The presentation of 3-D bone models during a medical consultation can modify cognitive and emotional representations relevant to treatment initiation among people with osteoporosis and might facilitate commencement of bisphosphonate treatment.

Keywords: illness perceptions; risk perceptions; visualisation; treatment initiation; 3-D printing; bone models
Existing interventions to improve treatment initiation and adherence largely focus on the provision of educational information. Although improvements in patient understanding of fracture risk can be obtained, educational interventions are inconsistent in modifying patient behaviour (Cranney et al., 2008; Sale, Beaton, Posen, Elliot-Gibson, & Bogoch, 2011). The development of novel intervention strategies to improve treatment uptake, which can also be scaled to the large number of people living with osteoporosis, is necessary.

The adoption of a treatment regimen is influenced by a patient’s beliefs regarding their illness, and perceptions of the importance of the therapy (Bassett & Petrie, 1999; Horne et al., 2013; Petrie & Weinman, 2012). Changing illness perceptions and medication necessity beliefs offers the prospect of improved self-management of illness, and consequently, better health outcomes (Broadbent, Ellis, Thomas, Gamble, & Petrie, 2009a, 2009b; Petrie, Perry, Broadbent, & Weinman, 2012).

While symptoms often act as key motivators for the engagement of protective health behaviour in a number of illnesses (Leventhal, Brissette, & Leventhal, 2003), in osteoporosis, the symptoms of increasingly porous bones are hidden from patients. In fact, patients may not be aware of the illness unless they have a bone scan or suffer from a bone fracture. Making these symptoms visible to patients with osteoporosis may be a way to motivate health behaviour change (Petrie & Weinman, 2012). Recent work in other chronic illnesses suggests that presentation of health information through visual means, whereby an individual is exposed to visual representations of the disease and the consequences of not taking treatment, can improve the understanding of the implications of personal health behaviours, and consequently, motivate adherence behaviour (Hollands & Marteau, 2013; Karamanidou, Weinman, & Horne, 2008; Perera, Thomas, Moore, Faasse, & Petrie, 2014).

Evidence suggests that people with osteoporosis have difficulties in effectively translating verbal information from their physicians into an accurate understanding of what a high fracture risk means and how treatment will impact on their health (Douglas et al., 2012). On these grounds, we propose that the use of visual aids might help modify treatment-relevant perceptions among people with osteoporosis and consequently encourage the initiation of bisphosphonate treatment.

There are two different forms of effective treatment for osteoporosis available in New Zealand. The first is an oral medication (alendronate), which is taken daily. The second is an intravenous infusion (zoledronate), typically administered once annually. As these have quite different behavioural demands for patients, we were interested in how the intervention impacted on the initiation of both treatments. Here, we report on a study using 3-D bone models as a tool to change the perception of disease and improve initiation of bisphosphonate treatment (either alendronate or zoledronate) among people who were newly diagnosed with osteoporosis.

**Methods**

**Participants**

Adult patients referred to bone mineral density measurement at the Auckland City Hospital bone densitometry service over a five-month period were screened for eligibility. Potential participants were mailed information about the study two weeks prior to their bone mineral density measurement appointment. Individuals were eligible to
participate if their estimated fracture risk (https://www.garvan.org.au/bone-fracture-risk) met National Osteoporosis Foundation (2013) recommendations for the consideration of pharmacologic intervention. These criteria were: (a) have had a hip or vertebral fracture, (b) have low bone density at the hip, neck of femur or spine, (c) have a five-year risk of having a hip fracture of greater than 1.5% and a major osteoporotic fracture risk of greater than 10% as determined by the GARVAN risk calculator or (d) is currently on glucocorticoids and has low bone density at the total hip, neck of femur or spine. Patients were ineligible if they were already taking treatment for osteoporosis, younger than 60 years, unable to speak English, or cognitively impaired.

**Procedure**

Participants were randomised to receive either a standard medical interview or a medical interview augmented by the use of 3-D bone models. A research assistant, who was blind to the study aims, conducted the randomisation procedure using a computer-generated random number sequence. Numbered envelopes (1–58) were used which contained a sheet of paper indicating if the participant was to receive the bone model intervention consultation or the control consultation. Immediately prior to the consultation, the physician opened the envelope to see the participants’ allocated group. All interviews were conducted by the same physician, and a script was used to ensure consistency in the information given to each participant. The researcher assessing outcomes was blind to participant allocation, and only the physician delivering the intervention was aware of group allocation.

The standardised script is shown in the Appendix. No medication was prescribed during the study visit because standard clinical care involves the provision of a bone mineral density report to the referring physician (normally the patient’s general practitioner), with whom the final responsibility for osteoporosis management lies. Participants in the intervention group were presented with 3-D models of healthy and osteoporotic bones (Figure 1). The purpose of the 3-D models was to provide the participant with a visual representation of osteoporosis and to explicitly highlight the difference in microarchitecture between healthy and osteoporotic bone. The participant was encouraged to interact with the models by holding them and taking note of the structural differences between them, particularly differences with regard to density, weight and porosity. The models were based on human biopsy specimens from the iliac crest of a healthy individual and an individual with osteoporosis. The models were printed at the Auckland Bioengineering Institute using a Dimension Elite 3-D printer (Stratasys, Minnesota).

Participants completed questionnaires before and immediately after their interview. A final questionnaire was administered via telephone two months later by a research assistant blind to group assignment.

**Measures**

**Illness perceptions**

The 9-item Brief Illness Perception Questionnaire (Broadbent, Petrie, Main, & Weinman, 2006) was used to assess participants’ illness representations at baseline, immediately after the interview, and at two-month follow-up. The original scale was
modified so that the wording was specific to osteoporosis. Items on the scale measured participants’ perceptions about the severity, emotional effects, symptoms, understanding and causes of osteoporosis. The questionnaires administered post-interview and at two-month follow-up also included items assessing participants’ beliefs about the duration and controllability of osteoporosis. Each item was rated on an 11-point scale (0–10) with relevant anchors.

**Medication beliefs**

Three questions related to the necessity-concern framework (Horne et al., 2013) were included to assess perceptions about bisphosphonate treatment. The items quantified...
participants’ perceptions about the effectiveness of bisphosphonates (How much do you think medication can help osteoporosis?), the necessity of bisphosphonate treatment (How much do you feel that the medications prescribed for your osteoporosis will help your future bone health?) and their concerns about prescribed bisphosphonates (How concerned are you about medications that may be prescribed for your osteoporosis?). Each item was rated on an 11-point scale (0–10) with relevant anchors.

**Evaluation of bone models**

Questions to ascertain perceptions of the 3-D bone models were administered post-interview to participants who were randomised to view the models. Specifically, participants were asked: (a) ‘How helpful were the bone models in helping you understand osteoporosis?’ (b) ‘Did seeing the bone models change your motivation for taking treatment?’ and (c) ‘Did seeing the bone models make you more anxious about osteoporosis?’. Each item was rated on an 11-point scale from 0 (not at all) to 10 (extremely) with relevant anchors.

**Initiation of bisphosphonate treatment**

The primary outcome was initiation of bisphosphonate treatment within two months of the study visit. Initiation of treatment was assessed by self-report and pharmacy dispensing records from the Auckland District Health Board database. The type of bisphosphonate treatment initiated (oral – alendronate, or infusion – zoledronate) was also recorded.

**Statistical analyses**

The data were analysed using the SPSS version 20.0 software (Chicago, Illinois). Data were assessed for normality, and non-parametric testing was employed for analysis of variables which did not have a normal distribution. Independent sample t-tests, Mann Whitney U tests and χ² tests were used to assess differences between the control and intervention group in demographic variables. χ² tests were conducted to assess between-group differences in the proportion of patients who had initiated bisphosphonate treatment, including differences between the groups in starting an oral or infusion regimen, at the two-month follow-up. One-way analyses of covariance, controlling for baseline measures of the outcome variable, were used to examine the effects of the 3-D bone models on perceptions of osteoporosis and beliefs about bisphosphonates. For beliefs which were not measured at baseline, independent samples t-tests and Mann Whitney U tests were used to compare group data.

The protocol was approved by the University of Auckland Human Participants Ethics Committee (ID: 8903) and the Auckland District Health Board Research Review Committee (ID: A+5761). This study was registered with the Australia New Zealand Clinical Trials Registry (Trial ID: ACTRN1261300263796). All participants gave written consent.

**Results**

The flow of participants through the study is shown in Figure 2. The final sample comprised 58 participants (29 control, 29 intervention) with a mean age of 73 years. The
Participants were predominantly female (91%), not employed (76%) and of New Zealand European ethnicity (85%). Almost half of the sample reported their highest level of education as greater than secondary level (48%). There were no significant differences between the control and intervention groups at baseline with regard to demographic characteristics, illness beliefs or medication beliefs. The largest proportion of participants (28%) stated they did not know what caused their osteoporosis, 24% attributed the illness to ageing, 12% to genetics and 10% to diet, 7% to an accident, 6% to hormonal factors, with the remainder giving a variety of other causes.

Immediately following the consultation, participants in the 3-D bone group were significantly more emotionally affected by osteoporosis, \(M = 4.08, \text{SE} = .41, 95\% \text{CI } [3.25, 4.90]\) compared to participants in the control group \(M = 2.89, \text{SE} = .40, 95\% \text{CI } [2.08, 3.70]\), \(F(1,52) = 4.25, p = .044, \eta^2_p = .08\). At two-month follow-up, the two groups differed significantly in their perceived understanding of osteoporosis, \(F(1,51) = 4.38, p = .041, \eta^2_p = .08\), with participants in the 3-D bone consultation group reporting a greater understanding of osteoporosis \(M = 7.19, \text{SE} = .51, 95\% \text{CI } [6.17, 8.21]\) relative to participants who received the control consultation \(M = 5.72, \text{SE} = .49, 95\% \text{CI } [4.74, 6.70]\). There were no significant differences between the intervention group and control group in beliefs about osteoporosis \(ps > .11\) or perceptions of bisphosphonates \(ps > .48\), either immediately following the consultation or at two-month follow-up. Within the intervention group, the 3-D bone models were perceived to be helpful to the participants’ understanding of osteoporosis \(M = 9.18, \text{SD} = 1.4\). Participants in the intervention group also reported that viewing the models changed their motivation for taking treatment \(M = 7.75, \text{SD} = 3.0\) and moderately increased their anxiety about osteoporosis \(M = 6.0, \text{SD} = 3.3\).

We found a greater proportion of participants in the intervention group initiated an oral bisphosphonate regimen (alendronate) (52%) in comparison with the control group (21%), \(\chi^2 (df = 1, N = 54) = 6.05, p = .01\). The proportion of participants who initiated an infusion bisphosphonate regimen (zoledronate) did not differ significantly between the intervention (10%) and control group (24%), \(\chi^2 (df = 1, N = 58) = 1.93, p = .16\). Overall, there was no statistically significant difference at two-month follow-up between
the proportion of participants in the intervention group who had initiated any bisphosphonate treatment (62%) and the proportion of participants in the control group who had initiated any bisphosphonate treatment (45%), $\chi^2 (df = 1, N = 58) = 1.73, p = .19$.

**Discussion**

Effective strategies to improve rates of treatment uptake among patients with osteoporosis are needed. In this study, we investigated the effect on the initiation of treatment of presenting 3-D models of healthy and osteoporotic bone. We found that the incorporation of bone models into a medical consultation about fracture risk and treatment benefit did increase the proportion of participants who initiated oral but not infusion bisphosphonate treatment. This suggests that the addition of 3-D bone models to a standard consultation may improve uptake of recommended therapy, but replication with a larger sample size is needed. An effect of similar magnitude, if confirmed in a larger trial, would be clinically meaningful. The observation that the 3-D model intervention differentially influenced initiation of oral vs. parenteral therapy may be attributable to chance but could reflect that the intervention may have differential effects on patients’ acceptance of oral or infusion therapy and this could be explored in future research. Patients who viewed the bone models were more emotionally affected by osteoporosis immediately after the interview and reported a greater understanding of osteoporosis at two-month follow-up, in comparison with the control group. Participants who viewed the 3-D bone models reported elevated anxiety about osteoporosis but also considered the models to be helpful in increasing their understanding of osteoporosis and altering their motivation to initiate treatment. Although it is not clear from the way this question was framed whether this was necessarily in a positive direction, but given the heightened emotional response and enhanced understanding elicited by the 3-D bone models, it seems most likely that the 3-D bone models helped in motivating patients to initiate an oral treatment regimen. We could speculate that this increased in anxiety may have prompted patients to choose the more familiar treatment modality of medication, rather than the novel infusion treatment, which may have increased anxiety further.

A strength of the study is that it was a randomised controlled trial. Outcomes were also assessed by staff blinded to treatment allocation. Participants were also blinded to the study aims; participants were told that a study was being conducted to investigate different ways of presenting information but they were not aware of the exact nature of the different consultations. However, it should be noted that the study was limited by the small sample size, and recruitment was restricted to those attending for bone mineral density measurement. The sample size also restricted the identification of potential mediators that may be important in influencing the effect of the intervention, such as specific illness or medication beliefs. It is important that this is addressed in future research. Another limiting factor was that while patients received information about fracture risk and bisphosphonate treatment during their study interview, they were required to see their primary care physician at a later time point to initiate treatment. This may have diluted the effect of the intervention on treatment initiation. Medication is frequently under-prescribed by physicians even when a patient meets fracture risk thresholds for pharmacological treatment (Elliot-Gibson, Bogoch, Jamal, & Beaton, 2004). Thus, although the 3-D bone models may have motivated an individual to begin treatment, the effect of the models in driving treatment uptake may have been weakened.
by the patient’s subsequent consultations with other physicians and the influence of their preferences and beliefs. It should also be noted that due to the fact the design specifically examined whether 3-D bone models influenced treatment initiation, we cannot determine whether other forms of visualisation such as pictures may also be effective.

To our knowledge, this is the first trial to assess the effects of a visual intervention on initiation of treatment for osteoporosis. Previous studies have evaluated intensification of patient education and presentation of risk information, with mixed results. Two studies reported increases of 15–18% in initiation of bisphosphonate therapy in patients with fractures randomised to education-based interventions (Cranney et al., 2008; Majumdar et al., 2008). However, in a lower risk population, randomisation to an educative decision aid did not alter the initiation of bisphosphonate therapy (Montori et al., 2011).

A growing body of research supports the visual presentation of diagnostic and health risk information as a means to facilitate positive health behaviours, through the alteration of patients’ illness perceptions and medication beliefs (Devcich, Ellis, Broadbent, Gamble, & Petrie, 2012; Hollands & Marteau, 2013; Karamanidou et al., 2008). In an example of this approach, researchers used patients’ most recent blood test results to provide a moving pictorial representation of the patient’s CD4 count and HIV viral load in a mobile phone app (Perera et al., 2014). Protective barriers around the CD4 lymphocytes were made up of each of the antiretroviral medications the patient is taking. When any medication dose was missed, the protective barrier corresponding to that medication breaks down and virus begins attacking the CD4 lymphocytes. Thus, the model provides the patient with a real-time picture of their level of immune protection against the HIV virus and the consequences of non-adherence. The findings also fit with previous research showing visual representations of illness are important predictors of recovery from illness (Broadbent, Ellis, Gamble, & Petrie, 2006).

3-D printed bone models or other visualisation media hold promise as cost-effective clinical tools which could be readily integrated into consultations to help increase initiation of treatment among people with osteoporosis. The findings of this research support further evaluation of the 3-D bone model intervention. It is simple and easy to deliver, inexpensive to implement and requires little resource involvement from the patient or the physician. Future research might also investigate the impact of 3-D bone models on medium and long-term adherence to bisphosphonates following treatment initiation, and their combination with other educational interventions.

Disclosure statement

No potential conflict of interest was reported by the authors.

References


Appendix 1. 3-D bone intervention script

I am going to discuss with you some information about osteoporosis (thin bones), your risk of breaking a bone and the treatment you might consider to reduce that risk. The final decision as to whether you take treatment rests with you, after discussion with your doctor, who will receive a report about today’s results in the mail within the next two weeks.

The results from today’s assessment indicate that you have a high enough risk of breaking a bone in the next five years to consider taking a medication to reduce that risk. This advice is based on recommendations made by the National Osteoporosis Foundation in the USA, which are widely followed by international experts in the field of bone health.

Osteoporosis is a disease which thins the bones and increases the risk of breaking them. In osteoporosis, your bone density is reduced, accompanied by weakening of the architecture of the bone. Here are two plastic models that have been generated by reconstructing high-resolution x-rays of pieces of bone taken from the hip of people in their 60s. This is the model of the healthy bone from someone who did not have osteoporosis, and this is the model of bone from someone who has osteoporosis (Provide the bone models and invite participant to look and touch them). As you can appreciate, the bone from the patients with osteoporosis is thinner, lighter and more porous (space-filled) than the bone from the patient who does not have osteoporosis. You can see that the osteoporotic bone is weaker and more likely to break than the healthy bone.

The most effective treatment to improve your bone health is a medication called a bisphosphonate. Bisphosphonate medicines can be taken either as a tablet once a week or once a month, or as an infusion (by a ‘drip’) once every year. The two different bisphosphonate treatments are equally effective. During a course of treatment, bisphosphonates reduce fracture risk by improving bone density and structure, that is move your bone from being thin, porous and fragile (indicate osteoporotic model) towards being denser and stronger (indicate normal model). These drugs are safe but each has a side effect. The bisphosphonate tablet can cause indigestion in about one in five people. The bisphosphonate given by infusion (drip) can cause about one in five people to...
experience ‘flu-like’ symptoms (fever, muscle ache, headache) for one to three days after the treatment – the symptoms always get better completely.

The final decision as to whether you decide to take treatment to reduce your fracture risk rests with you, after consultation with your doctor. Your doctor will receive a report of the results from today’s scans, and we suggest you speak to him/her at that time. He or she will be able to answer any questions you have about aspects of your bone health.

**Control consultation script**

I am going to discuss with you some information about osteoporosis (thin bones), your risk of breaking a bone and the treatment you might consider to reduce that risk. The final decision as to whether you take treatment rests with you, after discussion with your doctor, who will receive a report about today’s results in the mail within the next two weeks.

The results from today’s assessment indicate that you have a high enough risk of breaking a bone in the next five years to consider taking a medication to reduce that risk. This advice is based on recommendations made by the National Osteoporosis Foundation in the USA, which are widely followed by international experts in the field of bone health.

Osteoporosis is a disease which thins the bones and increases the risk of breaking them. In osteoporosis, your bone density is reduced, accompanied by weakening of the architecture of the bone. The most effective treatment to improve your bone health is a medication called a bisphosphonate. Bisphosphonate medicines can be taken either as a tablet once a week or once a month, or as an infusion (by a ‘drip’) once every year. The two bisphosphonate treatments are equally effective. These drugs are safe but each has a side effect. The bisphosphonate tablet can cause indigestion in about one in five people. The bisphosphonate given by infusion (‘drip’) can cause about one in five people to experience ‘flu-like’ symptoms (fever, muscle ache, headache) for one to three days after the treatment – the symptoms always get better completely.

The final decision as to whether you decide to take treatment to reduce your fracture risk rests with you, after consultation with your doctor. Your doctor will receive a report of the results from today’s scans, and we suggest you speak to him/her at that time. He or she will be able to answer any questions you have about aspects of your bone health.