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Modelling the cost effectiveness of a potential new neck collar for patients with motor neurone disease

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Abstract

Background
Patients with motor neurone disease (MND) suffer from reduced motility and strength of the neck muscles over time. This commonly results in difficulties with correct positioning for communicating, eating and breathing. There are a number of neck braces which available for MND patients, however they are inadequate for the majority of MND patients, and a new collar is set to be designed. This paper conducts a brief analysis of the potential cost effectiveness of such a collar.

Methods
A cost-utility analysis was undertaken using a simple three state Markov model of disease progression. The key input to this model were utilities from a small health valuation survey based around scenarios describing MND and the associated symptoms and limitation relating to reduced neck motility and strength. A scenario sensitivity analysis was used to identify the price at which new neck braces could be cost-effective given different effets on uptake and effectiveness.

Results
Four scenarios were examined that examined different impacts on rates of use of a neck brace and effectiveness of the new neck brace. For moderate effectiveness and no impact on non-use the maximum annual incremental collar cost to the NHS, using a funding threshold of £20,000 per quality adjusted life year, was £720. Adjusting this scenario for a large effect increases the maximum allowable cost to £960. Altering these two scenarios to include a large impact on rates of non-use produced maximum allowable costs of £800 and £1,040, respectively,

Conclusions
Our analysis suggests that there is considerable scope for a new neck brace for MND patients with moderate and severe neck pain to represent a cost effective use of NHS resources. Even with a brace costing around £1,000, it is possible that it will be cost-effective. However, these results should be interpreted with great caution: the potential brace has not yet reached the final design stage and although we do not assume in this analysis that it will prove adequate for all MND patients with neck weakness, we do assume that it provides a very substantial increase in adequacy rates (18% to 50% for moderate patients, and 0% to 30-50% for severe patients).

Key words
Motor neurone disease, cost-effectiveness analysis, neck brace, quality of life, product development.

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Introduction

MND has a prevalence of 6-8 / 100,000 in the UK population. At any one time there are approximately 5,000 people living with the condition, and approximately 1,200 die due to MND per year. The majority of individuals can expect an average life expectancy of between two and five years from symptom onset, with only 10 – 15% of people surviving for longer than 5 years. Based on limited data we estimate that approximately 38% of MND patients have moderate or severe neck pain and require a neck brace, although this proportion could be higher.

MND is a degenerative disease affecting the motor neurones in which cellular injury and death of motor neurones causes increasing loss of the connection between the nervous system and the voluntary muscles. In the majority of cases this is a rapidly disabling condition resulting in progressive paralysis. The affected person develops progressive weakness and wasting of the muscles controlling the limbs, bulbar function and respiration.

Motility and strength of the neck muscles decreases over time and the patient is then unable to raise their head independently. This commonly results in difficulties with correct positioning for communicating, eating and breathing. An adult human head weighs between 4.5 and 5kg, and when the neck muscles become weaker the patient is no-longer able to provide adequate support for their head.

There are a number of neck braces which are used to varying extents by MND patients. These braces can enhance the ability of a patient to carry out activities such as eating and communicating, can prevent pain caused by poor neck positioning, and can avoid the development of restricted movement. However these braces are inadequate for the majority of MND patients, and a new collar is set to be designed. This paper conducts a brief analysis of the potential cost effectiveness of such a collar.

Methods

In undertaking our economic analysis we have primarily followed the methods developed and used by the Multidisciplinary Assessment of Technology Centre for Healthcare (MATCH) collaboration (Cosh et al 2007, McAteer et al 2007). These methods describe a process by which potential devices can be evaluated in order to ascertain the likelihood that they will prove to be cost effective and economically viable. The process involves five key stages: strategic considerations; clinical problem definition; headroom analysis; return on investment analysis; and further economic analysis.

The 'strategic considerations' stage primarily involves manufacturer considerations and market characteristics and as such we have not addressed this stage. The 'clinical problem definition' stage involves a statement of the proposed technology, a description of the relevant disease area, where the proposed technology fits in the disease process, estimates of disease prevalence and incidence, an analysis of current treatments and their cost effectiveness. Because the proposed neck brace that is the subject of this project has not been specifically defined it has not been possible to make a precise statement of the technology. However the disease context has been investigated and this has been particularly important because of the progressive nature of the disease in question. This has formed the basis of the structure of the economic model that we have developed. Also, we have discussed the current nature of neck braces for patients with MND – specifically that they are sub-optimal. We found no papers investigating the cost effectiveness of existing braces. Our data on the likely effectiveness of the proposed brace compared to current braces is uncertain – we are not sure of the proportion of patients for whom the proposed brace will be adequate, and we are not sure whether the proposed brace will lead to more patients who need a brace actually using one. Therefore we have conducted scenario analysis around assumptions for these parameters.

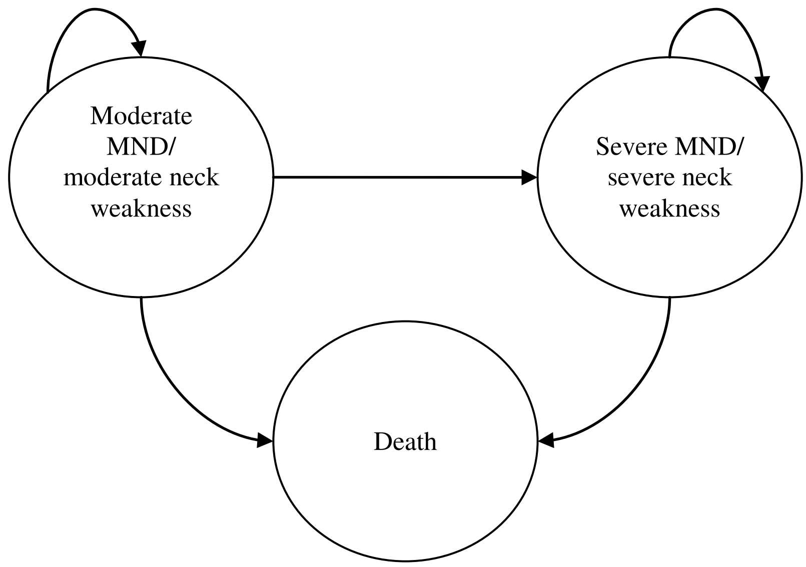
The next section of this report reports the 'headroom analysis' that we have undertaken for the proposed neck brace. Essentially, this stage of the MATCH framework aims to quantify the maximum incremental cost that the new technology could be associated with in order to be classed as cost effective, given assumptions around the clinical effectiveness of the technology. To conduct such an analysis it is necessary to estimate the quality of life impact in terms of quality adjusted life years (QALYs) associated with the new technology compared to current care. Given an estimated QALY gain and a cost effectiveness threshold (for example £20,000 as suggested by the National Institute for Health and Clinical Excellence (NICE)) the allowable incremental cost of the new technology can be estimated.

We have not addressed the 'return on investment' and 'further economic analysis' stages of the MATCH framework. These stages involve estimating the revenue and profit associated with the proposed technology, and conducting further sensitivity analysis to confirm the likely value of investment. These stages cannot be completed without finalised specific details about the technology because production costs need to be estimated.

Model Design

A simple 3-state Markov model was developed in order to assess the cost effectiveness of the potential new neck collar. The structure of the model follows a simplified structure of MND and neck weakness progression. The structure of the model is shown in the following diagram.

Figure 1: Markov model



We assume that a patient enters the model in a state of moderate MND with moderate neck weakness. The model does not include patients with mild MND because it is assumed that the new neck collar will only be a relevant treatment choice for those patients with moderate or worse neck weakness – we assume that for mild MND patients a beanie collar is adequate.

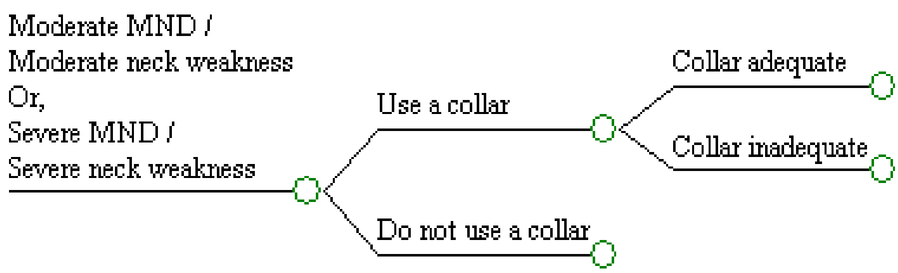
Figures from the Dementias & Neurodegenerative Diseases Research Network (DeNDRoN) suggest that 10-15% of MND patients survive for greater than 5 years from symptom onset. We took the mid-point of this estimate (12.5%) and applied a mortality rate in the model such that 87.5% of patients are dead 4 years after entering the model. We used a 4 year time-point rather than 5 years because we assume that patients spend on average 1 year in mild MND, and patients only enter the model when they have moderate disease. We assume that patients spend on average half of this 4-year time period in moderate MND with moderate neck weakness, and half of this time in severe MND with severe neck weakness. Given that we estimate that 12.5% of patients will not have died after 4 years, we also assume that 12.5% of patients will not have progressed to severe MND after 2 years. Thus, we apply a progression rate in the model such that 87.5% of patients will have progressed to severe MND with severe neck weakness after 2 years. We use a 1-year cycle length in the model.

We assume that the neck collar does not alter disease progression. However, within the moderate and severe health states different possible outcomes exist – a patient can either be wearing an adequate brace; wearing an inadequate brace; or wearing no brace despite the fact that one is needed. This is where a more effective brace will provide health benefits. Therefore, within the moderate and severe health states a decision tree defines the outcomes experienced by the modelled patient cohort. The decision tree is illustrated in the diagram below. The effectiveness estimates that inform this decision tree are discussed in the following section.

The decision tree dictates what proportion of patients are using an adequate, inadequate or no brace for moderate and severe health states. We assume that once allocated to one of these outcomes, the patient does not move into a different collar-specific state, i.e., if they are in the moderate disease health state and their collar is inadequate, there is no chance that their collar will become adequate in the following cycle. Hence, the only way in which a patient will move out of this state is if they die, or if they transition to severe disease, at which point they move through the severe state decision tree which determines whether their neck collar will be adequate, inadequate, or whether they wear no collar.

For each decision tree outcome within the moderate and severe health states a quality of life score and cost is applied. Costs and quality adjusted life years (QALYs) are discounted at annual rate of 3.5%. The model is run until everyone in the modelled cohort has entered the death state.

Figure 2: Within-state decision tree



<u>Effectiveness</u>

The planned neck collar has not yet reached the final design stage, and therefore the clinical benefits it may bring are assumptions in scenarios which provide an illustration of the potential cost effectiveness of the new neck collar. The effectiveness of the new collar is measured in two ways which are included in the economic model, described below:

- Increase in the proportion of MND patients who use a collar when one is needed. Because existing collars are not adequate for the majority of MND patients a significant proportion of patients who need a collar do not use one. We consider two possibilities with regard to the impact of the new collar on this assumption:

  - The new collar has no impact on the proportion of MND patients who use a collar when one is needed.

  - The new collar encourages all MND patients who need a collar to use one.

- Increase in the proportion of MND patients who use a collar for whom their collar is adequate. Because existing collars are sub-optimal for many patients, the key effectiveness benefit of the proposed new collar is that it will provide adequate neck support for more patients, which will be associated with an increased quality of life.

In the table below the effectiveness scenarios that we have considered in our cost effectiveness analysis are presented.

Table 1: Scenarios considered in the cost effectiveness analysis

Scenario 1	**Current situation**		**Forecast situation (with new collar)**	
	Moderate MND / Moderate neck weakness	**Severe MND / Severe neck weakness**	**Moderate MND / Moderate neck weakness**	**Severe MND / Severe neck weakness**
% that use collar given need	81%	100%	81%	100%
% for whom collar is adequate	18%	0%	50%	30%

Table 1 (cont...): Scenarios

Scenario 2	Current situation		Forecast situation (with new collar)	
	Moderate MND / Moderate neck weakness	**Severe MND / Severe neck weakness**	**Moderate MND / Moderate neck weakness**	**Severe MND / Severe neck weakness**
% that use collar given need	81%	100%	81%	100%
% for whom collar is adequate	18%	0%	50%	50%

Scenario 3	Current situation		Forecast situation (with new collar)	
	Moderate MND / Moderate neck weakness	**Severe MND / Severe neck weakness**	**Moderate MND / Moderate neck weakness**	**Severe MND / Severe neck weakness**
% that use collar given need	81%	100%	100%	100%
% for whom collar is adequate	18%	0%	50%	30%

Scenario 4	Current situation		Forecast situation (with new collar)	
	Moderate MND / Moderate neck weakness	**Severe MND / Severe neck weakness**	**Moderate MND / Moderate neck weakness**	**Severe MND / Severe neck weakness**
% that use collar given need	81%	100%	100%	100%
% for whom collar is adequate	18%	0%	50%	50%

These scenarios highlight the key effectiveness benefits that we assume the new collar will bring. The proportions presented in the 'current situation' cells reflect those seen in MND patients treated at the Royal Hallamshire Hospital, Sheffield. Thus, currently it is estimated that 81% of moderate patients who need a neck collar use one, and for 18% of these patients their collar is adequate. 100% of severe patients who need a collar use one, but in no cases are the collar adequate for the patient.

In scenario 1 it is assumed that the new collar does not alter the proportion of MND patients who use a collar, but the proportion for whom the collar is adequate increases from 18% and 0% for moderate and severe patients respectively, to 50% and 30%. Hence it is not assumed that the new collar will be adequate for all patients because this may be overly optimistic, but adequacy rates are increased appreciably. Scenario 2 is identical to scenario 1, except it is assumed that adequacy rates increase to 50% for both moderate and severe patients.

Scenario 3 is the same as scenario 1, except in this scenario it is assumed that the new collar will also have an impact on the proportion of MND patients who choose to use a collar, given that they need one. Thus it is assumed that 100% of moderate patients will use a collar in the forecast situation, compared to 81% in the current situation. Similarly, scenario 4 is identical to scenario 2, except that it is assumed that the use rate among moderate patients will increase to 100% in the forecast situation.

Quality of Life

In order to estimate the potential cost effectiveness of the proposed new neck collar, utility scores need to be estimated for each of the health states included within the economic model. MND utility scores are scarce in the existing literature, and certainly none exist which investigate the quality of life (QoL) scores associated with moderate and severe neck weakness with and without an adequate brace. Therefore, in order to populate our model we constructed a health state quality of life elicitation questionnaire, which we administered to a small (n=14) convenience sample of the general population. The questionnaire included health state vignettes for 6 health states which we constructed with considerable input from clinical experts.

The health states included were:

Health State A:	Moderate MND with moderate neck weakness, with an adequate brace
Health State B:	Moderate MND with moderate neck weakness, with an inadequate brace
Health State C:	Moderate MND with moderate neck weakness, with no brace
Health State D:	Severe MND with severe neck weakness, with an adequate brace
Health State E:	Severe MND with severe neck weakness, with an inadequate brace
Health State F:	Severe MND with severe neck weakness, with no brace

Participants were asked to read the health state descriptions and to imagine they were experiencing each state. They were then asked to complete the EQ-5D (Dolan 1997), the HUI3 (Feeney et al, 1995) and a Visual Analogue Scale (VAS) for each health state. The results of the study were as follows:

Table 2: Health state elicitations

Health State	EQ5D			HUI3			VAS		
	Mean	Lower CI	Upper CI	Mean	Lower CI	Upper CI	Mean	Lower CI	Upper CI
A	0.33	0.21	0.44	0.39	0.25	0.53	0.56	0.47	0.64
B	0.11	-0.02	0.24	0.14	0.02	0.25	0.46	0.37	0.54
C	0.07	-0.09	0.24	0.08	-0.05	0.20	0.42	0.33	0.50
D	-0.28	-0.40	-0.17	-0.18	-0.23	-0.13	0.31	0.22	0.40
E	-0.53	-0.61	-0.45	-0.26	-0.30	-0.21	0.18	0.11	0.26
F	-0.55	-0.62	-0.49	-0.26	-0.31	-0.22	0.14	0.06	0.22

As can be seen from Table 3, there were substantial point estimate differences between several of the health states for each valuation instrument. In particular, there is a large difference between an adequate brace and an inadequate or no brace when a patient is in either moderate or severe disease – these differences were often statistically significant. Also of note is that there were several negative scores elicited for severe disease when the EQ-5D and HUI3 were used. This implies that these states were valued as worse than death. However, when the VAS was used no participants ranked any of the health states worse than death.

In this paper, we present model results using the VAS quality of life scores. These represent a conservative estimate of QoL gain as the possible changes in utility values due to improvements in neck support are smallest using the VAS. These scores are allocated to the decision tree outcomes experienced by the modelled cohort.

<u>Costs and comparators</u>

The only costs included in the analysis are the cost of collars. Other resource use costs, such as physiotherapist appointments and GP consultations are not included due to a lack of data on neck-related MND appointments. Because it is likely that patients with better supported necks will require fewer appointments and consultations, our analysis will bias against the new neck collar.

In addition, we do not know the likely cost of the new collar. Hence the results presented here present a threshold analysis of the incremental cost allowable for the new collar, given its estimated clinical benefits. We have included the estimated costs of current collars, and so the estimated allowable incremental cost is over and above current collar costs (which are approximately £60 per annum, see Table 4).

A number of neck braces are currently available to MND patients. Initially patients use soft "beanie" style collars, which are generally popular as they are unobtrusive and comfortable to wear for extended periods of time. As neck weakness increases a "Headmaster" or "Oxford" collar or similar will be prescribed. These provide flexion control, but pressure sores can develop at the clavicles and under the chin if worn for long periods. The final orthotic intervention is an "Aspen" or "Philadelphia" cervical collar, designed for trauma victims. These collars hold the head rigidly, preventing all neck movement. These collars tend to be unpopular with patients, as there is insufficient padding for users who have no neck muscle control. See Box 1 for an overview of the suitability of existing collars.

Box 1: Existing neck collars for MND patients

The Beanie Collar
These are comfortable and popular with patients, but become inadequate as neck weakness progresses. These collars are sometimes used inappropriately.

The Oxford Collar
This is the only collar that has been designed specifically for MND patients. However it only restricts flexion of the head while supporting the head on the shoulders. This collar relies on the patient having control over the sideward tilting muscles of the neck. Patients with poor control of lateral movement due to their neck muscle weakness are not helped by this device.

The Headmaster Collar
This can be used for a greater degree of neck weakness however many patients get sore skin areas over the clavicle bones, with the weight of their head pressing onto their chest with no appropriate padding. This can result in patients not wearing the collar because it is uncomfortable. It also does not provide any lateral support.

The Philadelphia Collar
This collar is hot and claustrophobic to wear, and with more severe neck muscle weakness pressures are created by the areas surrounding the chin and mandible. There is inadequate padding for the clavicle area. When muscle weakness is severe the head can fall forwards onto the collar resulting in excessive pressures on the chin and jaw leading to skin necrosis, or pressure onto the clavicle area which can result in severe discomfort.

Table 3: Assumed collar pathways

MND Stage	Collar requirements	Costs (£)
Moderate MND, moderate neck weakness	Year 1: 4 beanie collars (£10 each) Year 2: 1 Oxford collar (£89)	Year 1: £40 Year 2: £89 Ave annual cost: £64.50
Severe MND, severe neck weakness	Year 1: 1 Oxford collar (£89) Year 2: 1 Philadelphia collar (£22)	Year 1: £89 Year 2: £22 Ave annual cost: £55.50

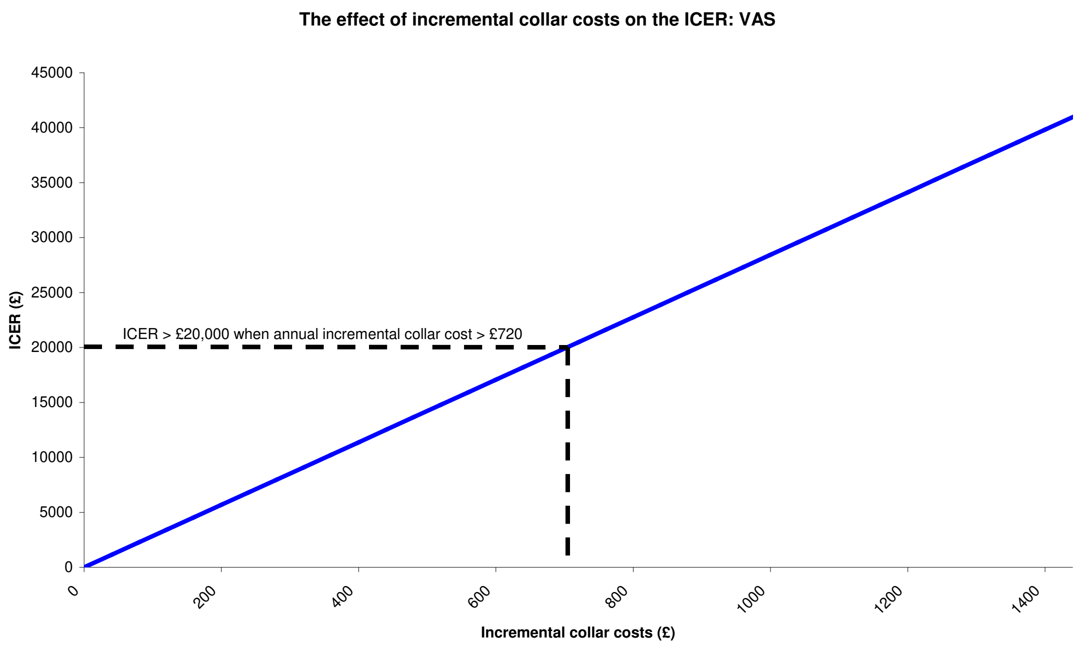
Note: Cost of collars from the MND Association

Results

The model results presented below illustrate threshold analysis of the allowable incremental costs of the new collar if it is to be classed as cost effective, for each scenario modelled. We used a cost effectiveness threshold of £20,000 as commonly used by the National Institute for Health and Clinical Excellence (NICE) in the UK. This means that for each additional QALY gained, the incremental cost must be no more than £20,000. If this criteria is satisfied a new collar would be very likely to be classed as cost effective by NICE and would therefore be reimbursed by the NHS.

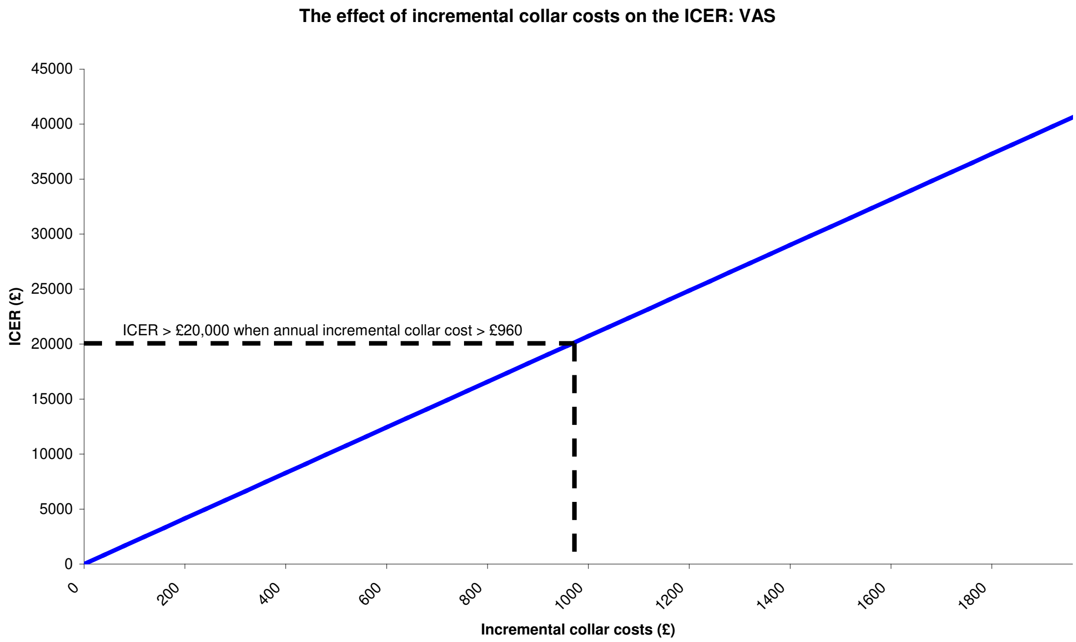
Scenario 1

In this scenario we estimate that if the new collar had an annual incremental cost of £720 or less it would generate an incremental cost effectiveness ratio (ICER) of less than £20,000.



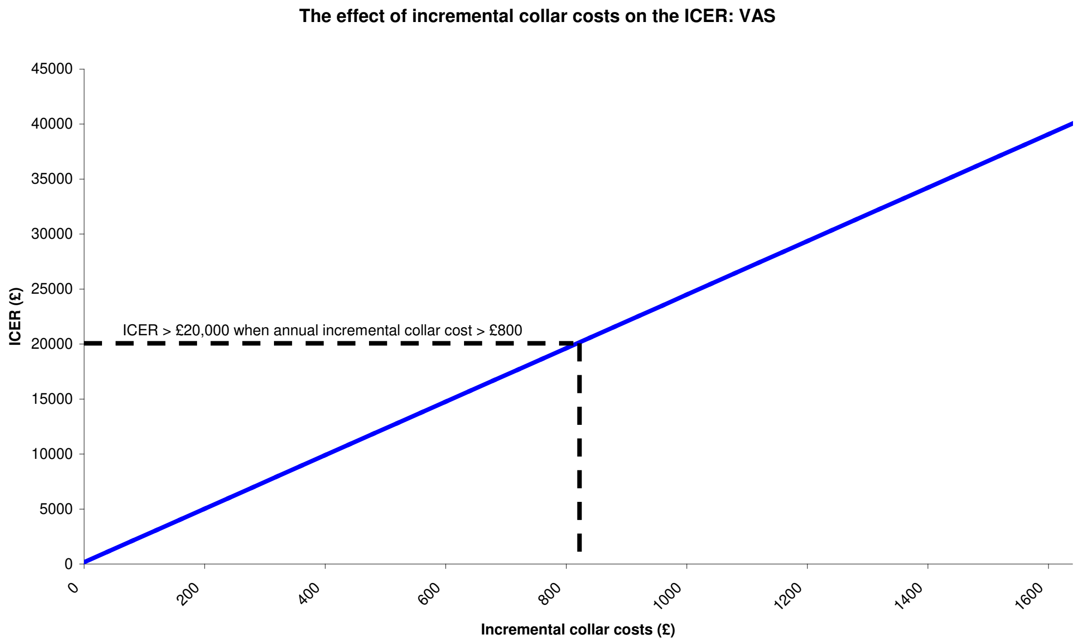
Scenario 2

In this scenario we estimate that if the new collar had an annual incremental cost of £960 or less it would generate an incremental cost effectiveness ratio (ICER) of less than £20,000.

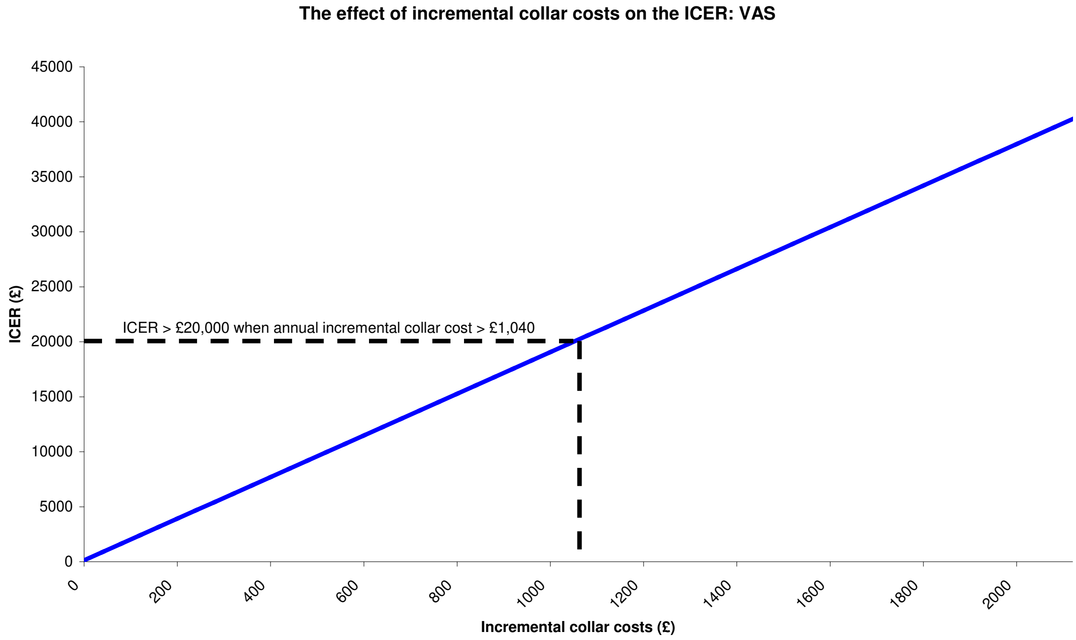


Scenario 3

In this scenario we estimate that if the new collar had an annual incremental cost of £800 or less it would generate an incremental cost effectiveness ratio (ICER) of less than £20,000.



Scenario 4
In this scenario we estimate that if the new collar had an annual incremental cost of £1,040 or less it would generate an incremental cost effectiveness ratio (ICER) of less than £20,000.



For ease of comparison, the results of our analysis are presented in Table 5.

Table 5: Results of the scenario analyses

Scenario	Scenario summary*	Allowable annual incremental collar cost, producing ICER < £20,000
1	Moderate impact on adequacy, no impact on use	£720
2	Large impact on adequacy, no impact on use	£960
3	Moderate impact on adequacy, large impact on use	£800
4	Large impact on adequacy, large impact on use	£1,040

* For full details see Table 1.

As expected, the largest allowable annual incremental collar costs are associated with scenarios 2 and 4, in which it is assumed that the new collar improves adequacy rates to 50% for both moderate and severe patients (as opposed to 30% for severe patients in scenarios 1 and 3). Also as expected the allowable annual incremental cost is higher in scenario 3 than in scenario 1, and in scenario 4 compared to scenario 2. This is expected because scenarios 1 and 3 and scenarios 2 and 4 are identical other than scenarios 2 and 4 assuming that the new collar will increase the use of neck collars from 81% to 100% in moderate patients (both sets of scenarios assume 100% use in severe patients, in line with current situation estimates).

Conclusions

Our analysis suggests that there is considerable scope for a new neck brace for MND patients with moderate and severe neck pain to represent a cost effective use of NHS resources. Even with a brace costing around £1,000, it is possible that it will be cost-effective. However, these results should be interpreted with great caution: the potential brace has not yet reached the final design stage and although we do not assume in this analysis that it will prove adequate for all MND patients with neck weakness, we do assume that it provides a very substantial increase in adequacy rates (18% to 50% for moderate patients, and 0% to 30-50% for severe patients).

In addition, the results presented here are dictated importantly by the health state values that we have elicited from a small convenience sample, which provided us with substantial quality of life score differences for the different health states. If the true differences between these health states was found to be much smaller (for example through a larger sample of MND patients), this would reduce the allowable annual incremental collar costs significantly.

Despite these weaknesses, the results also include some conservative assumptions/estimates. The estimates are based on the VAS health state valuation tool, which gave the smallest between health state differences. Therefore from this perspective the results presented are pessimistic. In addition, no other resource use savings are included, which – assuming that a more suitable neck collar would reduce other resource use – also biases our results against the new collar.

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