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The effects of five years of nocturnal cuirass-assisted ventilation in chest wall disease

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ABSTRACT: We investigated the long-term effectiveness of cuirass-assisted ventilation, and examined whether mortality and morbidity could have been predicted at the time of admittance.

Twenty five patients were commenced on nocturnal cuirass-assisted ventilation between 1983 and 1985, 10 with scoliosis or kyphosis, 8 with a thoracoplasty and 7 with neuromuscular disease. Mean pretreatment vital capacity was 30% of predicted, and arterial carbon dioxide tension ($Paco_2$) was 8.2 kPa (62 mmHg).

Fifteen patients were alive 5 yrs later. Two had discontinued assisted ventilation, both dying soon afterwards, and three had been changed to intermittent positive pressure ventilation. Survival could not have been predicted from age, severity of disease, lung volumes or arterial blood gases at presentation. $Paco_2$ in the survivors had risen from a mean of 6.1 kPa (46 mmHg) after one year to 6.8 kPa (52 mmHg) after 5 yrs ($p<0.05$), but remained significantly less than at presentation. There were no significant change in arterial oxygen tension (Pao_2), lung volumes, respiratory muscle strength, haemoglobin, right heart failure, exercise tolerance, mental function and symptom scores after 5 yrs, compared to after 1 yr. The median amount of time spent in hospital declined from 15 days per patient in the first year after initial discharge with cuirass-assisted ventilation, to between 3–5.5 days per patient in subsequent years.

We conclude that nocturnal cuirass-assisted ventilation has a role in long-term management of patients with neuromuscular and skeletal chest wall disorders. A randomized comparison with nasal intermittent positive pressure ventilation is now indicated.

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The technique of external negative pressure ventilation using cuirass-assisted ventilation was first reported over 100 yrs ago, but it was the stimulus of the poliomyelitis epidemics in the middle decades of the present century that lead to the improvements that made it practical for clinical use [1]. A proportion of poliomyelitis victims continued to need assisted ventilation in the long-term [2], and for some of these cuirass-assisted ventilation was an effective and more convenient alternative to a tank ventilator (iron lung), or positive airway pressure through a tracheostomy [3, 4]. It was subsequently realized that the technique of cuirass-assisted ventilation could be of value in managing chronic hypoventilation resulting from other diseases affecting the respiratory muscles [5–12], or chest wall deformities [13–16]. Non-invasive assisted ventilation is increasingly used in the management of these disorders, particularly since the introduction of intermittent positive pressure ventilation (IPPV) *via* a nasal mask [17]. Little information is available, however, about which patients are most likely to benefit, or about the demands that patients commenced on long-term assisted ventilation at home make on health services.

Between 1983 and 1985, we commenced 25 patients with neuromuscular and skeletal chest wall disorders on nocturnal cuirass-assisted ventilation at home [18]. We have reviewed these patients after 5 yrs, to see in which patients cuirass-assisted ventilation remained effective, whether long-term survival could have been predicted at the time of presentation and how often they were admitted to our Assisted Ventilation Unit.

Subjects and methods

Of the 25 patients originally commenced on cuirass-assisted ventilation, 12 were female. The median age was 53 yrs (range 12–68 yrs), and vital capacity (VC) was 31 % predicted (range 8–69% predicted). Twenty three patients had a VC less than 50% predicted, and 21 had an arterial carbon dioxide tension ($Paco_2$) greater than 6 kPa (46 mmHg) at presentation. Five patients had previously required IPPV *via* an endotracheal tube for acute respiratory failure. Nine patients had a thoracic scoliosis (idiopathic in seven and secondary to poliomyelitis in two), and one had a thoracic kyphosis as a result of spinal tuberculosis. Eight patients had had a thoracoplasty

performed for tuberculosis in the 1950s. In addition to the two patients with previous poliomyelitis included in the scoliotic group, seven patients had a neuromuscular disease affecting the respiratory muscles (Duchenne's dystrophy in two, other muscular dystrophy in two, and an unclassified myopathy in three).

Individual cuirass shells were made for each patient [19], and these were used with either a Cape cuirass pump or Newmarket pump (SiPlan Electronics Research Ltd) [20]. Approximately half of the patients used a Cape pump initially, but after 5 yrs all had changed to a Newmarket pump. The cuirass was used overnight, and additionally during the day when necessary, at the most negative pressure the patient could tolerate and at the respiratory rate that they found most comfortable. The ability of cuirass-assisted ventilation to produce adequate ventilation and to rest the respiratory muscles was confirmed by measurements of tidal volume, blood gases and the electromyographic activity of the respiratory muscles. When doubt arose as to whether the patient was using cuirass-assisted ventilation at home, this was verified from a clock on the pump. Upper airway obstruction during cuirass-assisted ventilation was excluded by clinical observation, and monitoring of oxygen saturation and carbon dioxide levels during sleep.

Patients who were alive after 5 yrs were reassessed during a routine visit to the Assisted Ventilation Unit. Arterial blood was sampled from the radial artery with the patient breathing air, and analysed in a Radiometer blood gas analyser. VC was measured using a wedge spirometer (Vitalograph), and functional residual capacity (FRC) by helium dilution (P.K. Morgan). Total lung capacity (TLC) and residual volume (RV) were computed from FRC, inspiratory capacity and expiratory reserve volume. Predicted values were taken from standard equations [21], using arm span as an estimate of height in those patients who were unable to stand or who had a scoliosis [22]. Maximum mouth pressures were recorded using a pressure transducer (SiPlan Electronics Research Ltd) connected to a mouthpiece with a 22 gauge leak. Maximum inspiratory pressure (MIP) was obtained at RV, and maximum expiratory pressure (MEP) at TLC. Maximum voluntary ventilation was estimated by measuring expired volume, whilst the patient breathed for 15 s through a low resistance valve (P.K. Morgan). Ambulant patients performed a 6 min corridor walk. Symptoms were assessed using visual analogue scales and a depression questionnaire, and the patients also performed simple mental function tests consisting of copying, deletion of numbers from a matrix, decoding of letter-number pairs and a trail test. Details of hospital admissions and travel with the cuirass and pump were obtained from hospital records, and from an interview with the patient. In calculating the number of days spent in hospital, the initial admission to initiate cuirass-assisted ventilation was excluded. Clinical signs of right heart failure and electrocardiographic evidence of right heart "strain" were scored by the system used by HOEPPNER *et al.* [23]. For the index of diuretic usage, one unit was arbitrarily defined as 40 mg of frusemide, 1 mg of bumetanide, 5 mg of amiloride or 50 mg of spironolactone.

Statistical comparisons were made using non-parametric tests in the "First" statistical package on an A3000 microcomputer (Acorn), the level of statistical significance being taken as 0.05.

Results

Two patients with scoliosis refused to continue with assisted ventilation, and both died within one year. The survival curve for the other 23 patients is shown in figure 1. Fifteen patients were alive after 5 yrs. Two patients who lived alone, one with a scoliosis and one with a thoracoplasty, died within one week of their initial discharge from the Assisted Ventilation Unit. Two patients with myopathies died during acute respiratory tract infections, one despite being intubated and ventilated, and the other in the ambulance on his way to the Assisted Ventilation Unit. Two patients, one with a myopathy and one with a thoracoplasty, died with gradually worsening right heart failure. One patient with a thoracoplasty and a history of asthma died at home during an acute wheezy episode.

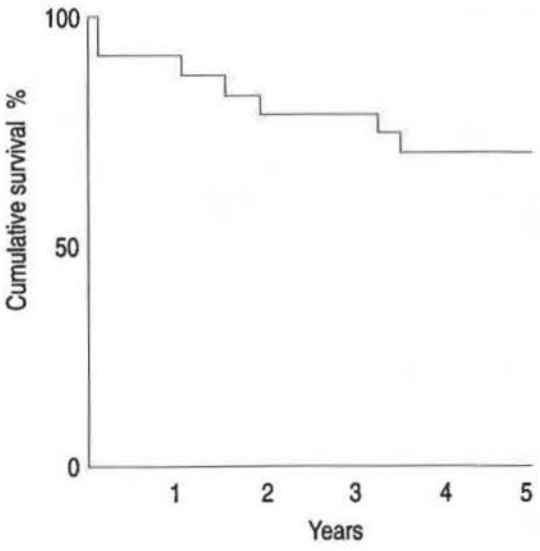

Fig. 1. – Survival of 23 patients with chest wall disease treated with nocturnal cuirass-assisted ventilation at home.

Table 1. – Age, vital capacity and arterial blood gases at the time of initial presentation of patients who died within 5 yrs of commencing nocturnal cuirass-assisted ventilation, compared to those who survived

	Alive n=15	Dead n=10
Age yrs	58 (23–68)	48 (12–64)
VC % pred	26 (8–48)	36 (25–69)
$Paco_2$ kPa	7.8 (5.4–10.9)	7.6 (5.2–9.5)
Pao_2 kPa	7.9 (5.5–11.3)	7.7 (5.2–12.8)

Data are presented as mean and range in parenthesis. VC: vital capacity; $Paco_2$: arterial carbon dioxide tension; Pao_2: arterial oxygen tension.

Table 2. – Arterial blood gases, lung volumes, maximum mouth pressures, 6 min walking distance, haemoglobin concentration, clinical right heart failure scores, electrocardiographic right heart strain scores and diuretic therapy index in 12 patients before, after 1 yr and after 5 yrs of nocturnal cuirass-assisted ventilation

	Before	After 1 yr	After 5 yrs
$Paco_2$ kPa	8.2±1.7	6.1±0.8†	6.8±1.0†*
Pao_2 kPa	8.1±2.0	9.2±1.5	9.4±1.9
VC *l*	1.06±0.52	1.07±0.49	1.00±0.40
TLC *l*	2.00±1.03	2.09±0.59	2.17±0.77
FRC *l*	1.25±0.46	1.40±0.37	1.48±0.56
RV *l*	1.23±0.70	1.17±0.46	1.28±0.66
MIP cmH_2O	44±19	47±12	41±15
MEP cmH_2O	62±28	66±21	81±54
MVV $l \cdot min^{-1}$	20±8	26±9	30±8†
6 min walk m	219±136	317±109†	225±89
Hb $g \cdot dl^{-1}$	15.3±0.9	14.7±0.9†	14.9±1.0
Rt heart failure	1 (0–2)	0 (0–1)	1 (0–2)
Rt heart strain	1 (0–4)	1 (0–3)	0 (0–3)
Diuretic therapy	2 (0–4)	1 (0–4)	2 (0–8)

Data are presented as mean±SD, or median with range in parenthesis. TLC: total lung capacity; FRC: functional residual capacity; RV: residual volume; MIP: maximal inspiratory pressure; MEP: maximal expiratory pressure; MVV: maximal ventilatory volume; †: $p<0.05$ compared to before; *: $p<0.05$ compared to 1 yr. For further abbreviations see legend to table 1.

Table 3. – Mean symptom scores in six patients before, after 1 yr and after 5 yrs of nocturnal cuirass-assisted ventilation

	Before	After 1 yr	After 5 yrs
Breathlessness (0–100)	28	34	40
Tiredness (0–100)	6	25	15
Happiness (0–100)	0	2	2
Depression (0–62)	6	3	7

0 = opitimal score.

Table 4. – Mean time taken to complete mental function tests in six patients before, after 1 yr and after 5 yrs of nocturnal cuirass-assisted ventilation

	Before	After 1 yr	After 5 yrs
Trail test s	41	33	40
Deletion test s	33	38	31
Copying test s	40	30	34
Decoding test s	59	47	49

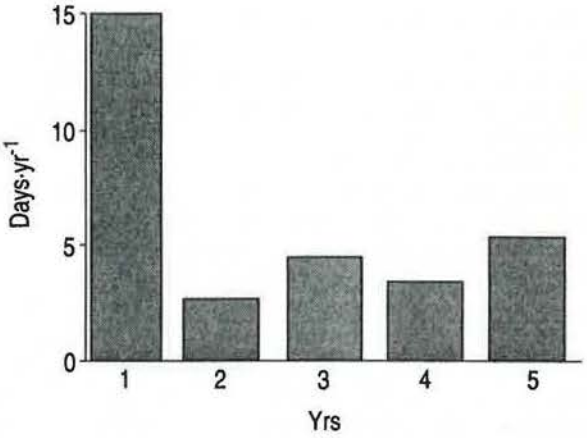

Fig. 2. – Median number of days spent as in-patients on the Assisted Ventilation Unit by 15 patients with chest wall disease after initial discharge from hospital with cuirass-assisted ventilation.

The other death occurred in a patient with scoliosis, in another hospital at night, during cuirass-assisted ventilation, 2 days after an elective tracheostomy had been performed. In total, four of the 10 deaths occurred in hospital. There were no significant differences in age, VC, arterial oxygen tension (Pao_2) or $Paco_2$ at the time of initial presentation, between those patients who died and those who survived for 5 yrs (table 1).

During the five year period of follow-up, two patients were changed to IPPV *via* a nasal mask, in both cases on account of increasing difficulty fitting the cuirass through a combination of limb muscle weakness and either severe thoracic deformity or obesity. After an unsuccessful trial of IPPV *via* a nasal mask, one further patient was changed to IPPV through a tracheostomy, for deteriorating respiratory failure.

Details of changes in physiological variables for the 12 patients still using nocturnal cuirass-assisted ventilation after 5 yrs are given in table 2. Lung volume and maximum mouth pressure measurements were not made in two patients prior to commencing assisted ventilation, and one patient with muscular dystrophy was too weak to perform the tests after 5 yrs. Changes in symptom scores and mental function tests are given in tables 3 and 4, there being no statistically significant difference in the median scores after one or 5 yrs. Figure 2 shows the median number of days spent in hospital each year, including planned check-up admissions, and admissions precipitated by an acute illness. Only one of the 15 patients had been admitted to another hospital during the 5 yrs, and she was the only patient to live more than 100 km from the Assisted Ventilation Unit. One patient had undergone an operation in Newmarket General Hospital, spending the postoperative period on the Assisted Ventilation Unit. Five of the 15 patients had been on holiday in the United Kingdom with their cuirass, as had one of the 10 patients who were not alive at 5 yrs. Two had also been abroad. Only one patient remained in employment. Two patients who lived alone had managed nocturnal cuirass-assisted ventilation successfully for 5 yrs.

Discussion

In the absence of a control group, any conclusions about the effect of nocturnal cuirass-assisted ventilation on the natural history of our patients must be made with caution. Although it would have been preferable from a scientific point of view to withhold treatment from half of the patients in a random manner, we did not feel that this was ethically acceptable. The patients who discontinued assisted ventilation do not constitute an adequate control group, and although their death could be interpreted as showing that assisted ventilation was necessary for their survival, an alternative explanation is that the patients' rejection of assisted ventilation reflected the absence of any beneficial effect.

Table 5 gives details of other studies of assisted ventilation in patients with chest wall disease, in which a 5 yr survival rate is given or can be calculated [5, 8, 13, 24–29]. The overall figure of 60% in our patients is rather less than previous reports, although in some of these studies it is not entirely clear if all relevant deaths were reported. Our patients showed a similar variation in survival pattern to previous reports [24], with 80% of scoliotic and kyphotic patients surviving 5 yrs, but only 60% of patients with a thoracoplasty. Differences in case mixture make comparison with other series difficult, particularly in patients with progressive neuromuscular diseases, and we do not feel that sufficient evidence is available to conclude that cuirass-assisted ventilation is less effective than other methods of ventilatory assistance.

We were unable to predict which patients would survive from data available at the time of their initial presentation in either acute or chronic respiratory failure. We do not feel that assisted ventilation should be denied patients merely on the grounds of their age, the extent of their disease, or the severity of respiratory failure at the time of presentation. As in previous studies [30], even patients with progressive neuromuscular diseases appeared to derive considerable benefit from assisted ventilation. The demand of these patients on acute medical services was low, declined with time, and was probably less than it would have been had they not received assisted ventilation.

In retrospect, some of the deaths could probably have been prevented. One patient died after surgery in another hospital, emphasizing the importance of close monitoring and surveillance by staff from the Assisted Ventilation Service after any operative procedure. The deaths shortly after initial discharge of two patients living alone might have been prevented by closer supervision, although their discharge home in each case followed a lengthy admission, with arrangement of the maximum home support available. One patient, who had been doing well on cuirass-assisted ventilation, developed what was thought to be a minor upper respiratory tract infection. He deteriorated and died in the ambulance on the way to hospital. This illustrates the importance of emphasizing to the patient, their carers and local medical practitioner that prompt treatment should be given for any respiratory tract infection.

The physiological benefits seen after one year of nocturnal assisted ventilation in these patients [18] were less evident in the smaller number of patients still using cuirass-assisted ventilation after 5 yrs. Although the mean $Paco_2$ had risen compared to the level after one year, it remained significantly less than prior to commencing assisted ventilation. In the absence of any significant change in lung volumes or respiratory muscle strength, the increase in maximum voluntary ventilation between one and 5 yrs is likely to represent a learning effect, although an improvement in respiratory muscle function, which was not picked up by maximum mouth pressures, or a reduction in airway resistance are alternative explanations.

In normal subjects and patients with chronic airflow obstruction, nasal IPPV has been shown to be more effective than external negative pressure ventilation in unloading the respiratory muscles during wakefulness [31], but the relevance of this in the long-term has not yet been

Table 5. – Five year survival rates of patients with chest wall disease using nocturnal assisted ventilation

First author	[Ref.]	Patients n	Disease	VC mean pre treatment % pred	Method of ventilation	5 yr survival %
Weirs	[13]	3	P, S	19	Cuirass	67
Alexander	[5]	5	NM	?	Cuirass	40
Splaingard	[26]	40	NM	?	Tank	76
Garay	[8]	6	P, S	32	Tank/Jacket	100
Curran	[25]	18	NM	13	Tank	76
Gerard	[24]	80	S, TB	<55	TIPPV	74
Baydur	[27]	9	NM	13	TIPPV	89
Bach	[28]	92	P	30	MIPPV	92
Leger	[29]	255	NM, S, TB	<20	NIPPV	88*
Jackson	Present study	25	P, NM, S, TB	30	Cuirass	60

P: poliomyelitis; NM: other neuromuscular; S: scoliosis; TB: sequelae of tuberculosis (including thoracoplasty); TIPPV: intermittent positive pressure ventilation *via* a tracheostomy; MIPPV: via a mouthpiece; NIPPV: *via* a nasal mask; VC: vital capacity. *: 4 yr survival rate.

established. At the time our patients were commenced on assisted ventilation, we were unable to offer nasal IPPV as an alternative. Whilst a few patients currently using external negative pressure ventilation may need to be transferred to nasal IPPV [32], the majority of our patients are in a stable clinical state, and are happy with cuirass-assisted ventilation. Some patients prefer external negative pressure ventilation to nasal IPPV [33], and our current practice is to try both techniques in most cases, and to choose whichever is more effective and more acceptable for the patient. External negative pressure ventilation may be better tolerated by young children than nasal IPPV [34], and at present it is more reliable and cheaper. Difficulties in fitting cuirass shells to patients with severe thoracic deformities, obesity, or immobility have been described previously, but were uncommon in patients presenting to our Assisted Ventilation Unit. We did not encounter the problems with recurrent aspiration which have been reported previously [27], possibly because of the semi-recumbent position in which most of our patients slept with their cuirass respirators. Similarly, upper airway obstruction was less of a problem than previously described with external negative pressure ventilation [17, 26, 30, 35], and none of the patients were prescribed protriptyline.

The use of nocturnal assisted ventilation is likely to increase in the future, as patients present with respiratory failure, as a late complication of poliomyelitis [36, 37], or thoracoplasty [38, 39], and as the beneficial effects for patients with nocturnal hypoventilation as a result of scoliosis or progressive neuromuscular diseases are recognized. Our experience suggests that cuirass-assisted ventilation will continue to have a role in the management of these patients. A randomized comparison of this technique with nasal IPPV is now indicated.

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