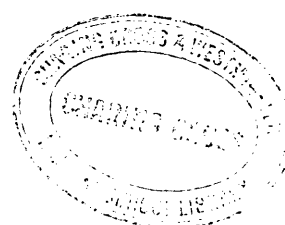


CUIRASS-ASSISTED VENTILATION
IN PATIENTS WITH
CHEST WALL DISEASE

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ABSTRACT

Cuirass-assisted ventilation was studied in twenty-five patients with chest wall disease. All patients had a vital capacity of less than 50% predicted, and twenty-three had daytime hypercapnia.

Tidal volume was proportional to the peak external negative pressure used, but in some subjects increasing this above 20cmH₂O reduced tidal volume. Positive external pressure had little effect on ventilation and produced a fall in end-expiratory volume. Minute ventilation increased with respiratory rate except in the presence of airflow obstruction. Cuirass-assisted ventilation corrected paradoxical chest wall motion. Comparison of cuirass respirators with jacket respirators showed little difference in ventilation.

An improvement in PaCO₂ and PaO₂ was seen during daytime cuirass-assisted ventilation. Oxygenation was improved by supplementary oxygen without inducing a rise in carbon dioxide levels. Cuirass-assisted ventilation could rest the respiratory muscles, but this did not result in any change in oxygen consumption, and respiratory muscle strength was unaffected by one hour of rest.

Cuirass-assisted ventilation during sleep reduced transcutaneous carbon dioxide tensions and improved oxygenation, although four subjects continued to have periods of hypoxia.

Of the twenty-five patients commenced on home nocturnal cuirass-assisted ventilation, four died within the first year. The surviving patients showed improvement in daytime arterial gases, lung volumes and right heart failure. Respiratory muscle strength, respiratory drive and pulmonary artery pressure were unchanged and the patients remained breathless. The cost of maintaining a patient on nocturnal cuirass-assisted ventilation was estimated to be in the region of £3000 per year.

Studies of cuirass-assisted ventilation using a new fan pump showed considerable leakage around the rim of the cuirass shell. The servo system in this pump performed well in limiting excessive peak external negative pressures, but was less good at compensating for increased air leakage. The feasibility of patient-triggered cuirass-assisted ventilation was demonstrated with this pump.

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ABBREVIATIONS

CAV	cuirass-assisted ventilation
cmH ₂ O	centimetres of water
ECG	electrocardiograph
EEV	end-expiratory volume
EMG	electromyograph
FEV ₁	forced expiratory volume in 1 second
FER	forced expiratory ratio
FRC	functional residual capacity
FVC	forced vital capacity
Hb	haemoglobin
Hz	Hertz
IMV	instantaneous minute volume
IPPV	intermittent positive pressure ventilation
kPa	kilopascals
l	litres
MEP	maximum expiratory pressure
min	minute
MIP	maximum inspiratory pressure
mls	millilitres
mmHg	millimetres of mercury
MVV	maximum voluntary ventilation
p	probability
PaCO ₂	arterial carbon dioxide concentration
PaO ₂	arterial oxygen concentration
PtcCO ₂	transcutaneous carbon dioxide tension
REM	rapid eye movement
RV	residual volume
SaO ₂	oxygen saturation
sec	second
SV	spontaneous ventilation
TLC	total lung capacity

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CHAPTER 1

THE HISTORY OF CUIRASS RESPIRATORS

The use of external negative pressure to assist ventilation was introduced over a century ago. There are three main types of external negative pressure device. The cuirass respirator consists of a rigid shell covering the patient's thorax, connected to a negative pressure pump. In the tank (or cabinet) respirator, the patient's body is enclosed up to the level of the neck in a rigid box, which is connected to a similar negative pressure pump. In this thesis, the term "jacket respirator" will only be used for the third type of external negative pressure ventilator in which a rigid shell, which covers but does not touch the thorax, is enclosed by an airtight garment.

This chapter describes developments in the design of the cuirass respirator shell from its invention to the present day, including the introduction of the jacket type of external negative pressure ventilator. The development of the pump units used to provide negative pressure will be described in Chapter 11.

1:1 EARLY MODELS OF CUIRASS

Von Hauke's "Pneumatische Panzer"

The first documented attempts to produce ventilation by applying negative pressure to the chest wall were made in the last century by Von Hauke, who postulated that negative pressure around the ribcage would produce better

expansion than the manual methods then in use. In 1874 he described a cuirass shell constructed from sheet iron with air filled rubber seals round the edges (Von Hauke 1874). This fitted over the anterior half of the thorax. By producing a negative pressure inside the shell Von Hauke found that he was able to produce effective ventilation in children with respiratory muscle paralysis, deformities of the chest wall, asphyxiation and atelectasis. The device was found to be unsuitable if the patient was agitated and Von Hauke went on to design a tank respirator. This proved to be better tolerated by the patients, and was more effective than the cuirass.

Alexander Graham Bell's Cuirass

The next record of cuirass respirators refers to one designed by Alexander Graham Bell in 1882 (Bell 1910). The cuirass he designed consisted of two metal shells which were bolted together around the thorax. A smaller model of this cuirass was shown to produce adequate ventilation in drowned cats. Bell was unable to convince the medical profession of its clinical value, but the principle of a rigid metal shell completely encircling the thorax was subsequently to be adopted in the Burstall jacket and London County Council cuirass. Bell's cuirass was sealed at the edges by inflatable rubber tubes, and this method of producing a seal was also used in later models of cuirass respirator.

Eisenmenger's "Biomotor" Cuirass

In the early years of this century in Germany, Rudolf Eisenmenger designed a cuirass in the form of a rigid dome which fitted over the patient anteriorly (Eisenmenger 1904). Eisenmenger realised the importance of enclosing the abdomen under the cuirass shell, and he incorporated a rubber balloon inside the shell which could be inflated to aid expiration.

Eisenmenger described several studies of the use of his "Biomotor" cuirass. It produced effective ventilation when used in resuscitation and in patients with restrictive pleural disease, pleural effusion, pneumonia, silicosis, asthma, emphysema, chronic bronchitis, bronchiectasis and post diphtheritic diaphragmatic paralysis (Eisenmenger 1942). Physiological studies revealed that the inspiratory flow waveform was closer to the normal pattern than that during other methods of resuscitation (Fischer and Engeser 1938). The rhythmic alternation of positive and negative pressure over the chest and abdomen was even found to produce circulation in the absence of cardiac contraction in animals (Eisenmenger 1929). This led to the use of the cuirass in the treatment of right heart failure (Eisenmenger 1936 & 1939, Fruhman 1951). It was also used in metabolic acidosis and coronary artery disease, whilst the rhythmic abdominal pressure was thought to be beneficial in cases of chronic constipation (Eisenmenger

1939).

In the 1930's it was discovered that Eisenmenger's Biomotor cuirass could maintain adequate ventilation in patients with respiratory muscle paralysis as a result of poliomyelitis. A poliomyelitis epidemic in South Africa had previously stimulated Steuart to develop a cuirass for use in cases of respiratory paralysis in 1918 (Steuart 1918), but the epidemic was over before it could be brought into use. The development in Germany of the Biomotor cuirass for use in poliomyelitis was interrupted with the outbreak of war, but research into this promising application continued in Scandinavia.

Sahlin-Stille Cuirass

The Sahlin-Stille cuirass consisted of a rigid dome which covered the thorax anteriorly. There were three sizes of shell with rubber strips to provide seals (Flaum 1936). The cuirass shell was placed over the patient and bolted onto an operating table. The major disadvantage was that the patient was required to be in the same position on the table and thus was liable to develop pressure sores. Flaum (1936), Bergman (1948) and Jungner et al (1956) used the Sahlin-Stille cuirass successfully in acute poliomyelitis. In the course of further studies, Engstrom found that positive airway pressure ventilation was superior to cuirass-assisted ventilation and he subsequently used the cuirass to assist expiration only (Engstrom and Svanborg 1950,

Engstrom 1954).

The Burstall Cuirass

Poliomyelitis epidemics also stimulated the development of cuirass respirators elsewhere. In 1937 following a poliomyelitis epidemic in Australia, Burstall designed a metal cuirass which was made in one piece and was fitted by passing it over the patient's head (Burstall 1938). There were three sizes of the "Burstall jacket" and the design was subsequently modified to include the abdomen in the metal casing. The cuirass took two people several minutes to apply and seal. As in Steuart's case earlier in the century, delay in developing the cuirass meant that it did not become available for clinical use until the epidemic was almost over. It was never widely used.

Turner's Cuirass

Turner developed a similar cuirass independently in England around the same time as Burstall's work in Australia. Turner's cuirass was constructed in two halves joined together by clips. Like Burstall's later design, Turner's cuirass also included the abdomen inside the shell. The cuirass could be used with a splint frame in cases of poliomyelitis with limb involvement (Medical Research Council Report 1938).

The London County Council Cuirass

A cuirass similar to Turner's was adopted for wider use by the London County Council. Fitting the cuirass

and bolting together the two halves still required several people (Menzies 1938, Blackwell 1949). It was used in the treatment of poliomyelitis, diphtheria, polyneuritis, progressive muscular atrophy, carbon monoxide poisoning and post operative atelectasis.

1:2 LATER MODELS OF CUIRASS

The types of cuirass available in the United Kingdom were reviewed by Menzies in 1938 (Menzies 1938) and considered in a report by the Medical Research Council (Medical Research Council Report 1938). In North America, experience of epidemics of poliomyelitis enabled the Council on Physical Medicine to produce a list of basic requirements for cuirass respirators in 1947. They stipulated that the cuirass should produce adequate ventilation without undue chafing of the skin or discomfort to the patient; it should be available in various sizes to fit different patients and should be simple for unskilled persons to apply. Five such respirators were considered by them and four of these were approved for use. The Council recommended that cuirass-assisted ventilation be used primarily to maintain ventilation whilst transporting poliomyelitis victims to a centre where tank ventilators were available, or to facilitate weaning from dependence on cabinet respirators (Council on Physical Medicine Reports 1947-1950).

The Mullikin Cuirass

The Mullikin portable respirator resembled the London County Council cuirass. It consisted of two aluminium shells which were bolted together by wing nuts. This was found to be uncomfortable and mechanically imperfect and was not accepted (Council on Physical Medicine 1949).

The Blanchard Cuirass

The Blanchard portable plastic respirator consisted of plastic anterior and posterior shells which surrounded the chest and were sealed round the neck and axillae with rubber sheeting. Huddleston (1947) reported the use of this cuirass to treat atelectasis after thoracotomy. A similar design of cuirass with plastic anterior and posterior shells has been used more recently in Japan for the treatment of patients with muscular dystrophy and hypoventilation (Saoyama et al 1984).

The Chestspirator cuirass was similar in design to the Blanchard cuirass. It was accepted by the Council 1949 (Council on Physical Medicine 1949) but no reports of its use were published.

The Huxley Cuirass

The Technicon-Huxley cuirass consisted of a plastic anterior shell made in several sizes to cover both the chest and abdomen. The lower edges of the cuirass were fitted with small legs to prevent downward movement of the shell onto the patient (Mollaret et al 1958). This cuirass design was used successfully in patients with

neuromuscular disease (Cornwell and Davis 1979) and Wallace et al (1961) used the Technicon-Huxley cuirass to maintain ventilation during anaesthesia for endoscopy.

The Drager cuirass and the "Cuirasse Materiel Medical et Sanitaire Francais" were similar in design to the Technicon-Huxley cuirass and were reported by Mollaret et al (1958) to be as effective as their American counterpart.

The Monaghan Cuirass

The Monaghan portable respirator consisted of a plastic dome which fitted over the chest and abdomen and was initially available in six sizes (Council on Physical Medicine 1949). The Monaghan cuirass was less efficient than tank respirators, was not able to maintain respiration reliably in acute or chronic poliomyelitis, and could produce abrasions when used for long periods (Plum and Lucas 1951, Plum and Wolff 1951, Bryce-Smith and Davis 1954, D'Avignon et al 1956). Nevertheless, Peterson and Ward (1948) found this cuirass useful in maintaining respiration during the operation of tracheostomy in patients normally managed in tank respirators.

The Kifa Cuirass

A similar design of anterior cuirass shell made of an aluminium shell with inflated rubber tubing at its edges called the Kifa cuirass was used in the United Kingdom. The seals at the edges were unsatisfactory and

the cuirass was less effective than the Monaghan (Kelleher et al 1952, Jungner et al 1956). The Kifa-Gulberg cuirass was used in conjunction with endotracheal positive pressure (Mollaret et al 1958).

Both the Monaghan and Kifa cuirasses could compress the lower ribcage and produce paradoxical inward motion during inspiration (Scales et al 1953). Downward movement of the cuirass shell with consequent restriction of anterior ribcage expansion was reduced in the Kifa cuirass by legs attached to the edge of the cuirass.

The Poliospirator cuirass, like the Kifa-Gulberg design, combined the use of negative pressure within a cuirass shell with positive pressure applied to the airway. The use of a cuirass which covered both the chest and abdomen resulted in more effective ventilation than the Kifa-Gulberg model (Mollaret et al 1958).

The Medical Research Council Cuirass

A detailed study of cuirass respirators undertaken at the Institute of Orthopaedics, Stanmore and the Electromedical Research Unit of the Medical Research Council at Stoke Mandeville was reported in 1953 (Scales et al 1953). A cuirass consisting of anterior and posterior shells was developed, similar to the Chestspirator, London County Council and Blanchard cuirasses, but with the shells individually moulded to fit the patient. Problems with the seals, particularly at the lower rim, and pressure sores led to this design

being abandoned. A version of the Sahlin Stille cuirass which was clamped to the couch on which the patient lay proved to be efficient but clumsy. The most satisfactory cuirass design developed in this study consisted of a moulded polythene shell covering the thorax anteriorly from the manubrium to the symphysis pubis. The lateral edges of the shell extended down onto the bed, and seals were made of foam rubber covered with rubber sheeting.

Further development of this cuirass design by the Medical Research Council led to the Spirashell. The shell had shorter sides than its prototype described above, and was fitted with deep foam rubber seals which could be removed from the shell (Figure 1). It was designed for patients with poliomyelitis to assist weaning from cabinet respirators during recovery, and for long term use in patients left with permanent respiratory muscle weakness.

FIGURE 1

The Spirashell Cuirass

(reproduced by permission of
the Medical Research Council)



1:3 CUIRASS DESIGN AFTER THE INTRODUCTION OF POLIOMYELITIS IMMUNISATION

The decline of poliomyelitis after the introduction of effective immunisation and refinement of intermittent positive airway pressure ventilation reduced the demand for cuirass respirators.

Cuirass respirators were still used to maintain ventilation during bronchoscopy and laryngoscopy (Peterson and Ward 1948, Green and Coleman 1955, Toker 1955, Bayuk 1957, Sleath and Graves 1958, Wallace et al 1961), and to aid weaning from intermittent positive pressure ventilation (Mollaret et al 1958).

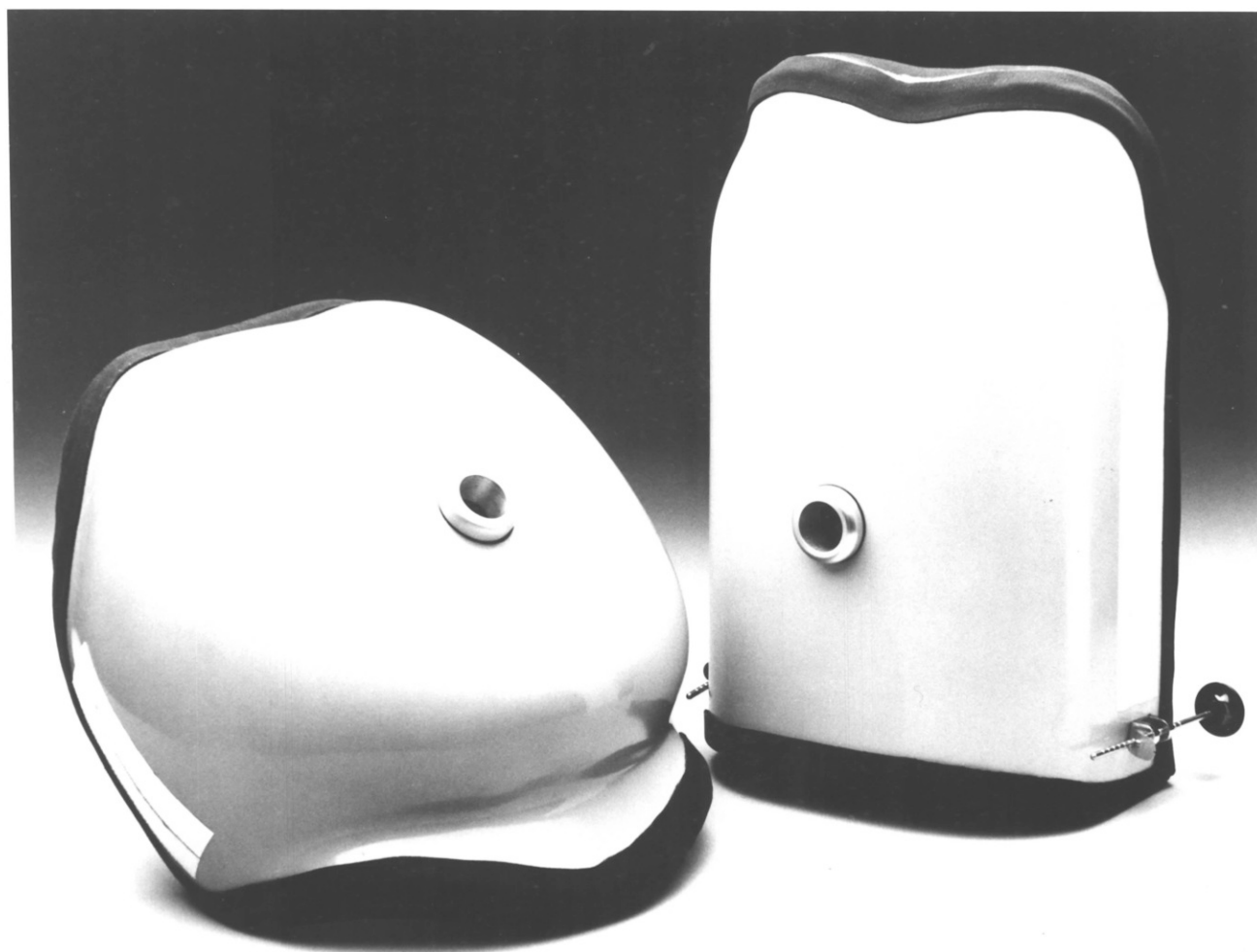
In the 1960's the Vital Capacitor cuirass was studied in both dogs and man, particularly with respect to its effect on the circulation, but its clinical value seems to have been limited (Fountain et al 1973, Getzen et al 1968).

Examples of modern cuirass shells (Lifecare, 655 Aspen Ridge Drive, Lafayette, Colorado), similar to the Monaghan and Huxley designs, are shown in Figure 2.

FIGURE 2

Modern Versions of the Monaghan and Huxley Cuirass Shells

(reproduced by permission of Lifecare)



Individually Moulded Cuirass Respirators

In 1961, Binet and Bochet described the manufacture of their thoraco-abdominal cuirass from moulded plastic (Binet and Bochet 1961). This could be made in a few hours to fit the individual patient, and extended from the clavicles to the iliac crests. The cuirass was fastened by a strap passing round the back of the patient. Their cuirass was used in the correction of hypoventilation resulting from mechanical deformities of the chest wall or respiratory muscle weakness. The development of techniques for moulding cuirass shells to fit the shape of the individual patient's thorax has overcome many of the difficulties in producing an adequate seal and avoiding abrasions during cuirass-assisted ventilation, particularly in patients with chest wall deformity (Weirs et al 1977). A plaster cast of the patient's thorax can be sent to a manufacturer to produce a moulded cuirass shell (Spencer 1981), or the shell can be produced using local resources (Powner and Hoffman 1978, Fulkerson et al 1984, Newman and Wilkins 1988).

Jacket Respirators

As an alternative solution to the problem of differences in chest wall shape, standard shells which do not touch the patient can be covered by an airtight garment to provide the seals around the neck, arms and waist. The shell does not restrict chest wall expansion,

and external negative pressure can be applied to a larger area of the chest wall than with a cuirass shell (Simonds and Branthwaite 1985). Examples of such jacket respirators are the Tunncliffe jacket (Spalding and Opie 1958) and Emerson respirator. These respirators require little adjustment for each patient, but help is usually required to get into the jacket.

CHAPTER 2

THE PATHOPHYSIOLOGY OF RESPIRATORY FAILURE IN PATIENTS WITH CHEST WALL DISEASE

2:1 DAYTIME RESPIRATORY FAILURE IN PATIENTS WITH CHEST WALL DISEASE

2:1:1 Patients with Idiopathic Scoliosis

Scoliosis of the thoracic spine results in deformity of the chest wall. This is associated with a reduction in the compliance of the chest wall (Bergofsky et al 1959, Ting et al 1960, Turino et al 1965, Kafer 1975). In the absence of muscle weakness, most studies have found that the degree of restriction of static lung volumes is related to the angle of spinal curvature (Mankin et al 1964, Westgate 1967, Bjure et al 1968, Weber et al 1975, Kafer 1975). Alveolar ventilation is also inversely related to the angle of spinal curvature (Kafer 1976), and as expected this leads to an inverse relationship between PaCO₂ and vital capacity (Turino et al 1965). A spinal curvature of more than 100 degrees or a reduction in vital capacity below 60% of predicted is likely to be associated with breathlessness (Bake et al 1972) and, less commonly, with respiratory failure of sufficient severity to cause cor pulmonale (Bergofsky et al 1959).

Lung compliance is also low in patients with scoliosis, even when the reduction in lung volumes resulting from the abnormal compliance of the chest wall is taken into account (Bergofsky et al 1959, Cook et al

1960, Ting et al 1960, Caro and DuBois 1961, Turino et al 1965, Cooper et al 1984). Although altered alveolar surface tension forces as a result of underinflation has been proposed as a possible mechanism for the reduced lung compliance, failure of hyperinflation during intermittent positive pressure breathing to correct the low compliance supports the alternative hypothesis of atelectasis in dependent areas of lung.

Deformity of the chest wall and defective mechanical coupling of the respiratory muscles may explain the inability of patients with thoracic scoliosis to generate normal inspiratory and expiratory pressures (Cook et al 1960, Cooper et al 1984). The mechanically disadvantaged respiratory muscles are faced with an increased work of breathing and must use a higher proportion than normal of their maximum strength during tidal breathing. It is therefore not surprising that these muscles become fatigued, with the result that PaCO_2 is inversely related to measures of respiratory muscle strength (Lisboa et al 1985).

The ventilatory response to carbon dioxide is impaired in patients with thoracic scoliosis (Bergofsky et al 1959, Kafer 1975, Mezon et al 1980). Although the ventilatory response is related to the degree of reduction in vital capacity, total lung capacity and chest wall compliance (Kafer 1975), the maximum ventilation achieved by carbon dioxide stimulation is

much less than the maximum voluntary ventilation, suggesting that central respiratory centre insensitivity may contribute to hypercapnia.

The age of onset of respiratory failure in patients with scoliosis is variable. In a survey of idiopathic scoliosis, Branthwaite (1986) found that respiratory failure was more common if the onset of scoliosis occurs under the age of 5 years, and that progressive loss of vital capacity with time was commoner in subjects who presented with a vital capacity of less than 50% predicted. This may explain the rise in PaCO₂ with increasing age seen in these patients and the higher incidence of respiratory failure after the age of 30 years (Godfrey 1970, Bjure et al 1970, Kafer 1976).

2:1:2 Patients with Poliomyelitis

During the poliomyelitis epidemics in the middle of this century, acute poliomyelitis involving the respiratory muscles was one of the major causes of respiratory failure. Assisted ventilation was usually necessary when the vital capacity fell below 50% of predicted (Ferris et al 1955). Many of the patients who needed assisted ventilation could be weaned back to spontaneous ventilation subsequently, but were often left with some respiratory muscle weakness. These patients are at risk of developing respiratory failure many years later, particularly if they have residual diaphragmatic paralysis or if they develop a thoracic

scoliosis (Lane et al 1974, Howard et al 1988).

Studies of patients with scoliosis as a result of poliomyelitis have shown more severe restriction of static lung volumes than in idiopathic scoliosis (Cook et al 1960, Caro and Du Bois 1961, Godfrey 1970, Kafer 1974), presumably reflecting residual respiratory muscle weakness. Progression of the degree of scoliosis with age could explain the delay before the onset of respiratory failure in these patients, but it has been suggested that late degeneration of motor units in respiratory muscles may also be responsible (Campbell et al 1969, Mulder et al 1972).

In contrast to the patients with idiopathic scoliosis Kafer (1974) found that the ventilatory response to carbon dioxide in patients with scoliosis following poliomyelitis was not related to the vital capacity. The presence of a poor ventilatory response to carbon dioxide in a patient with relatively normal lung volumes suggests a central respiratory centre defect, possibly as a result of previous poliomyelitis infection, which may contribute to hypercapnia occurring as a late sequel to poliomyelitis (Shannon et al 1970, Solliday et al 1974).

2:1:3 Patients with other Neuromuscular Diseases

Respiratory failure is a common development in the late stages of many progressive neuromuscular diseases. Respiratory muscle involvement at an earlier stage when the patient is less severely handicapped is

characteristic of some neuromuscular diseases, whose rate of progression may be slow. The degree of respiratory muscle weakness which leads to respiratory failure has been investigated by Braun et al (1983), who studied 37 patients with proximal myopathy. Vital capacity was linearly related to respiratory muscle strength when the latter was more than 30% of predicted, with greater loss of vital capacity in the few patients with respiratory muscle strength less than 30% of predicted. There was an inverse linear relationship between vital capacity and PaCO₂ given by the equation:

$$\text{PaCO}_2 \text{ kPa} = 7.85 - (0.038 \times \text{VC \% predicted})$$

From this equation, a PaCO₂ of 6 kPa or more will occur when vital capacity falls below 49% of predicted. A marked increase in PaCO₂ with decline in respiratory muscle strength (RMS) was seen when the latter was less than 50% of predicted, given by the relationship:

$$\text{PaCO}_2 \text{ kPa} = 7.24 - (0.049 \times \text{RMS \% predicted})$$

From this equation, a PaCO₂ of 6 kPa or more will occur when respiratory muscle strength falls below 25% of predicted.

Patients with neuromuscular diseases also have decreased lung compliance (Gibson et al 1977, De Troyer et al 1980) and decreased chest wall compliance (Estenne et al 1983), both of which contribute to an increase in the work of breathing. Impaired respiratory centre sensitivity to carbon dioxide has been documented

in neuromuscular disease (Riley et al 1977), but is likely to be more important as a cause of nocturnal rather than daytime hypoventilation.

2:1:4 Patients with a Thoracoplasty

The operation of thoracoplasty produced a fall in vital capacity (McIntosh 1935, Lindskog and Freidman 1936, Landis and Weisel 1954), which was later found to be proportional to the number of ribs resected (Gaensler et al 1955, Little 1956).

Thoracoplasty resulted in more pronounced loss of static lung volumes if scoliosis developed subsequently (Cournand and Richards 1941, Powers and Himmelstein 1951, Woodruff et al 1953,). The scoliosis could progress over a number of years (Loynes 1966), and this may account for the progressive ventilatory defect following thoracoplasty (Zifroni et al 1972), and for the development of respiratory failure as a late consequence of the operation (Bergofsky 1979).

Huang and Lyons (1977) studied twenty patients with respiratory failure developing seven to twenty three years after surgery for tuberculosis. None of the patients had a thoracoplasty without lung resection, but ten out of twelve patients with a thoracoplasty in addition to lung resection developed respiratory failure, compared to eight patients with lung resection alone. The patients with lung resection alone had a mean vital capacity of 38.7% predicted compared to 60.2% predicted

for the patients with a thoracoplasty and lung resection. This would suggest that factors other than loss of lung volume are involved in the development of respiratory failure. Bredin (1984) in a study of eleven patients a mean of thirty one years after thoracoplasty found a reduction in vital capacity to a mean of 46% predicted, with a mean PaCO₂ of 5.6 kPa.

In patients with a thoracoplasty pleural, lung parenchymal and bronchial scarring may also increase the work of breathing. As with scoliosis, the pressures which the respiratory muscles must generate during tidal breathing will therefore be greater, whilst the available muscle force may be reduced as a consequence of resection of intercostal muscle, residual phrenic nerve damage, or mechanically disadvantageous insertion of respiratory muscles resulting from scoliosis.

2:2 SLEEP AND GAS EXCHANGE IN PATIENTS WITH CHEST WALL DISEASE

2:2:1 Introduction

In normal subjects, sleep is associated with a decrease in tidal volume, an increase in breathing frequency, a fall in minute ventilation and a rise in either end-tidal or arterial carbon dioxide (Birchfield et al 1959, Gothe et al 1981, Douglas et al 1982a). An increase in upper airway resistance occurs with sleep (Berger 1961, Lopes et al 1983) which is partly compensated for by an increase in respiratory muscle activity (Tadachnik et al 1981).

The pattern of respiratory muscle activity varies with the stage of sleep. During non-rapid-eye-movement (non-REM) sleep, the ribcage contribution to ventilation is greater compared to the awake state, suggesting predominantly intercostal muscle contraction (Tadachnik et al 1981, Gothe et al 1981, Tusiewicz et al 1977, Muller et al 1980). In REM sleep, there is a decrease in the tonic electromyographic activity of the intercostal muscles (Muller et al 1980) with an increase in the phasic activity recorded from the diaphragm. This results in a decrease in the contribution of the ribcage to ventilation compared to non-REM sleep (Tadachnik et al 1981). REM sleep is also associated with irregularity of respiratory muscle contraction (Phillipson 1978, Douglas et al 1982a) and an increase in oxygen consumption (Brebba and Altshuler 1965, Stabell et al 1977, Stothers and Warner 1978, Henderson-Smart and Read 1979).

These changes which occur with sleep have a minimal effect on gas exchange in normal subjects. This is unlikely to be the case for patients with chest wall disease. Even the change from the upright to the supine posture will reduce alveolar ventilation in patients with diaphragmatic weakness, independently of any change in conscious level (Newsom Davis et al 1976, Loh et al 1979). In patients with chest wall disease, an increase in the work of breathing associated with an increase in

upper airway resistance could precipitate inspiratory muscle fatigue and lead to hypercapnia during sleep. The awake functional residual capacity of such subjects is low, and may fall during sleep if respiratory muscle tone is reduced as in normal subjects. A transient decrease in ventilation would therefore produce more profound hypoxaemia than it would in normal subjects, since the effect of apnoea on oxygen saturation is greater at lower lung volumes (Findley et al 1983). An increase in oxygen consumption would also result in a fall in oxygen saturation unless accompanied by an increase in ventilation. The effect of these factors on gas exchange during sleep will be considered separately for the subdivisions of chest wall disease used in the previous section.

2:2:2 Patients with Idiopathic Scoliosis

Mezon et al (1980) studied sleep in five patients with idiopathic scoliosis. Four patients had significantly lower mean oxygen saturations during REM sleep compared to the awake state, and in all five the lowest oxygen saturation was recorded during REM sleep. Two patients had central apnoeic episodes during REM sleep. One patient had periodic respiration, associated with oxygen desaturation only when it occurred during REM sleep. The remaining two patients, the only females in the group, had a normal respiratory rhythm during sleep, although one showed oxygen desaturation. Guilleminault et

al (1981) reported similar findings in series of five patients with kyposcoliosis with spinal curves of 90-115 degrees, all of whom hypoventilated during sleep, with the lowest oxygen saturations occurring during rapid eye movement sleep.

Midgren et al (1986) investigated 13 patients with severe scoliosis with a mean vital capacity of 39% predicted and daytime PaCO_2 of 6 kPa. During sleep, oxygen desaturation coincided with a rise in transcutaneous carbon dioxide tension. The subjects with mean nocturnal oxygen saturations of under 90% had significantly smaller vital capacities with a larger fall on assuming the supine position than the patients with a mean nocturnal saturation of over 90%. All the subjects with a mean nocturnal saturation of under 90% had daytime hypercapnia.

Sawicka and Branthwaite (1987) studied sleep in detail in eleven subjects with non-paralytic scoliosis, ten subjects with paralytic scoliosis and nine normal subjects. All the scoliotic subjects had a forced vital capacity of less than 56% of predicted. Mean ear oxygen saturation was lower in the scoliotic subjects during both REM and non-REM sleep compared to controls. Falls in oxygen saturation were most pronounced during REM sleep. Oxygen desaturation during sleep was related to age in the non-paralytic scoliotic patients. Although an increase in end-tidal carbon dioxide tension from

wakefulness to non-REM sleep was seen in both the normal and scoliotic subjects, a further increase during REM sleep was seen only in the scoliotic subjects. Episodes of hypopnoea in the non-paralytic scoliotic subjects occupied only a small proportion of non-REM sleep and were most pronounced during REM sleep. Hypopnoea occupied a larger proportion of non-REM sleep in the paralytic scoliotic compared to the non-paralytic scoliotic subjects, but was also more severe during REM sleep.

2:2:3 Patients with Poliomyelitis

Changes in respiratory muscle strength, chest wall compliance and central respiratory drive in patients with poliomyelitis make it likely that these patients will be at risk of developing respiratory abnormalitis during sleep. Hill et al (1983) and Goldstein et al (1987) have both reported patients with poliomyelitis who developed severe oxygen desaturation at night, particularly during REM sleep. These episodes of oxygen desaturation occurred mainly during central apnoeas, but with some additional obstructive apnoeic episodes. In a review of two hundred and nine patients with previous poliomyelitis, Howard et al (1988) found that eighty six patients needed assisted ventilation to correct nocturnal hypoventilation, which developed a mean of 28.5 years after the acute illness.

2:2:4 Patients with other Neuromuscular Disease

Solliday et al (1974), Bellamy et al (1975), Newsom

Davis et al (1976), Loh et al (1979), Skatrud et al (1980), Hill et al (1983), Goldstein et al (1987), Veale et al (1986), Chan et al (1987) and Stradling and Warley (1988) have all described abnormal gas exchange during sleep in patients with respiratory muscle weakness. Sleep was staged by Skatrud, Hill, Goldstein and Stradling, and in each case the severest abnormalities of gas exchange were detected during REM sleep. Skatrud suggested that intercostal inhibition during REM sleep was particularly likely to produce hypoventilation in patients with diaphragmatic weakness.

In a larger study of sleep in patients with neuromuscular disease, Bye et al (1986) described 10 patients with neuromuscular disease with a mean daytime PaCO₂ of 7.1 kPa, vital capacity of 48% predicted and inspiratory muscle strength of 26% predicted. Nine of these patients showed oxygen desaturation during rapid eye movement sleep.

Bradley et al (1984) reported 18 patients with central apnoeic episodes during sleep. Five of the subjects had daytime hypercapnia, and three patients with respiratory muscle weakness were included within this subgroup. The hypercapnic patients had apnoeic episodes during non-REM and REM sleep, but the mean duration of apnoea was slightly longer during REM sleep. Although these patients had low oxygen saturations at night, they had daytime hypoxaemia and were polycythaemic. The fall

in oxygen saturation with the change from wakefulness to sleep is not stated.

Smith et al (1988) have studied fourteen patients with Duchenne muscular dystrophy without daytime respiratory failure. Nine of these patients developed oxygen desaturation during sleep, with hypoventilation occurring predominately in REM sleep. Daytime respiratory function tests could not distinguish these nine patients from those who did not desaturate.

2:2:5 Patients with a Thoracoplasty

Although the value of assisted ventilation at night in patients with a thoracoplasty has been reported (Sawicka et al 1983), the only study of gas exchange during sleep in these patients is that of Phillips (1987). He studied twenty five patients with a thoracoplasty overnight using transcutaneous monitoring of oxygen and carbon dioxide tensions. The maximum transcutaneous carbon dioxide tension recorded overnight was a median of 0.7 kPa above awake levels (range 0.1 to 6.5 kPa), and twenty of the subjects had at least one hypoxaemic episode during the night.

2:3 INDICATIONS FOR NOCTURNAL ASSISTED VENTILATION

2:3:1 Nocturnal Hypoventilation

The presence of nocturnal hypoventilation in patients with chest wall disease may be suggested by a poor sleep pattern, nocturia or daytime symptoms such as sleepiness, poor concentration, confusion or morning headache. If

abnormal gas exchange during sleep is confirmed, assisted ventilation at night should improve these symptoms.

Assisted ventilation at night is also indicated when complications of abnormal gas exchange during sleep have developed, such as polycythaemia, pulmonary arterial or systemic hypertension.

Studies of gas exchange during sleep in patients thought to be at high risk of nocturnal hypoventilation will sometimes detect severe hypoxaemia and hypercapnia during sleep in the absence of established daytime complications or symptoms. It is usually appropriate to correct nocturnal hypoventilation in these patients if the severity is such that it is considered likely that symptoms, complications or death will occur if it is left untreated.

2:3:2 Daytime Alveolar Hypoventilation

Patients with chest wall disease who have daytime alveolar hypoventilation are likely to have more severe hypoventilation during sleep. O'Leary et al (1979) have suggested that patients with neuromuscular disease who have a daytime PaO_2 of less than 6.6 kPa or a PaCO_2 of greater than 7.2 kPa should be considered for nocturnal assisted ventilation. Rochester and Martin (1985) include a daytime PaCO_2 of greater than 5.9 kPa as one of the indications for assisted ventilation at night. In most cases, confirmation of abnormal gas exchange during sleep is possible prior to instituting assisted

ventilation.

2:3:3 Respiratory Muscle Fatigue

Rochester and Martin (1985) also suggested that assisted ventilation should be instituted in patients with clinical evidence of severe respiratory muscle fatigue in whom the PaCO₂ has not yet become elevated. Clinical indicators of fatigue include tachypnoea of greater than thirty breaths per minute, alternating chest and abdominal expansion and paradoxical inward inspiratory movement of the abdomen (Cohen et al 1982). The efforts of such patients to maintain normal arterial blood gases are unlikely to succeed for much longer, and nocturnal cuirass-assisted ventilation offers the possibility of resting the muscles at night to improve their daytime function.

2:3:4 Patients at Risk of Developing Respiratory Failure

Daytime respiratory failure is likely to develop in patients with thoracic scoliosis who have a vital capacity of less than 50-60% of predicted (Bergofsky et al 1959), in patients with poliomyelitis who have a vital capacity of less than 30% of predicted (Lane et al 1974), and in patients with other neuromuscular diseases who have a vital capacity of less than 50% of predicted or a reduction of respiratory muscle strength to less than 25% of predicted (Braun et al 1983).

Rochester and Martin (1985) suggested that assisted ventilation should be considered in patients with chest

wall disease who are at high risk of developing respiratory muscle failure, using the following criteria: maximum inspiratory mouth pressure of less than 25 cmH₂O in subjects with neuromuscular disease or less than 50 cmH₂O in patients with scoliosis, or an FVC less than 25% of predicted values. The studies mentioned in the previous paragraph would support a figure of 50% of predicted for vital capacity rather than 25% as the level below which there is an increased risk of respiratory failure. The limits for respiratory muscle strength take no account of the difference in the normal respiratory muscle strength between the sexes (Black and Hyatt 1969, Leech et al 1983), and maximum inspiratory pressures of less than 50 cmH₂O may be recorded even in normal subjects (Leech et al 1983). A maximum inspiratory mouth pressure of less than 50 cmH₂O should therefore be interpreted with caution, and should only be used as an indication for assisted ventilation when other indications are present.

2:3:5 Patients who Cannot be Weaned from Ventilators

In some subjects, assisted ventilation is instituted as an emergency and evaluation of the underlying pathophysiological mechanisms is subsequently impossible. These patients may be impossible to wean from nocturnal assisted ventilation, and cuirass-assisted ventilation may be substituted for intermittent positive pressure ventilation (O'Leary et al 1979).

2:4 CONTRAINDICATIONS TO NOCTURNAL CUIRASS-ASSISTED VENTILATION

Cuirass-assisted ventilation is inappropriate if it fails to correct abnormalities of gas exchange during sleep. Like other methods of external negative pressure ventilation (Scharf et al 1978), cuirass-assisted ventilation may be ineffective if upper airway obstruction develops.

In patients in whom nocturnal cuirass-assisted ventilation is commenced in the absence of nocturnal hypoventilation, alternative methods of assisted ventilation should be used if gas exchange during sleep is worse during cuirass-assisted ventilation than spontaneous ventilation. This may occasionally be caused by restriction of chest wall movement by the cuirass.

When nocturnal cuirass-assisted ventilation is used to rest the respiratory muscles at night, it would be logical to use an alternative method of assisted ventilation if a reduction in respiratory muscle activity is not confirmed by electromyographic studies.

In patients who are unable to protect their own airway from inhalation of oral secretions or regurgitated gastric contents, intermittent positive pressure ventilation using a cuffed treacheostomy tube is more appropriate than cuirass-assisted ventilation.

CHAPTER 3

METHODS USED IN THE STUDY OF CUIRASS ASSISTED VENTILATION AND DETAILS OF SUBJECTS PARTICIPATING IN THESE STUDIES

3:1 Manufacture of Cuirass Shells

A ready made cuirass shell selected from a range of sizes may be suitable for assisted ventilation in a subject with a normal chest wall shape. The deformity seen in the majority of patients with respiratory failure due to skeletal or neuromuscular chest wall disease will, however, usually prevent an adequate fit with a ready made cuirass shell (Weirs et al 1977, Curran 1981). A plaster cast can be taken of the patient's chest and sent to a manufacturer of cuirass shells for a cuirass to be constructed (Spencer 1981). Some delay in obtaining the cuirass is inevitable, and it may still need to be adjusted to fit the patient. Manufacture of cuirass shells in the same hospital as the patient offers advantages in terms of the speed with which a moulded cuirass can be obtained, and the ease with which minor adjustments can be made. The technique used at Newmarket to construct cuirass shells is described below.

The patient is positioned supine on a couch lying as flat as is comfortable. Two layers of Stockinette are placed over the chest anteriorly to prevent the plaster cast from sticking to the patient. Two strips of 5 cm square foam rubber are then placed across the patient. These foam strips are positioned where the inferior and superior borders of the finished cuirass will fit onto

the patient. The inferior strip is positioned to lie in the suprapubic region centrally, running laterally to the iliac crests and then vertically down onto the couch. The superior strip is positioned to lie over the manubriosternal angle centrally, running laterally below the clavicles to the axilla and then vertically down onto the couch.

Layers of plaster of Paris are applied to the patient over the area bounded by the superior and inferior foam strips and the couch laterally. When sufficient layers have been applied to make a rigid shell, the plaster is allowed to set for a few minutes and the cast carefully removed from the patient. It is left for several hours to set fully and then the central portion is built up with more plaster of Paris strips to a depth of 7-10cm to allow room for expansion of the thorax and abdomen when the finished cuirass respirator is in use. Before building up the cast, careful observation of the patient is required to determine which portions of the chest and abdomen move and in which direction during respiration. Attention is paid during the shaping of the cast to allow most room over the most mobile portions of the chest wall and also over any potential pressure points. The lateral edges of the cast and the portion overlying the foam strips are not altered since these areas are where the cuirass shell will seal onto the patient.

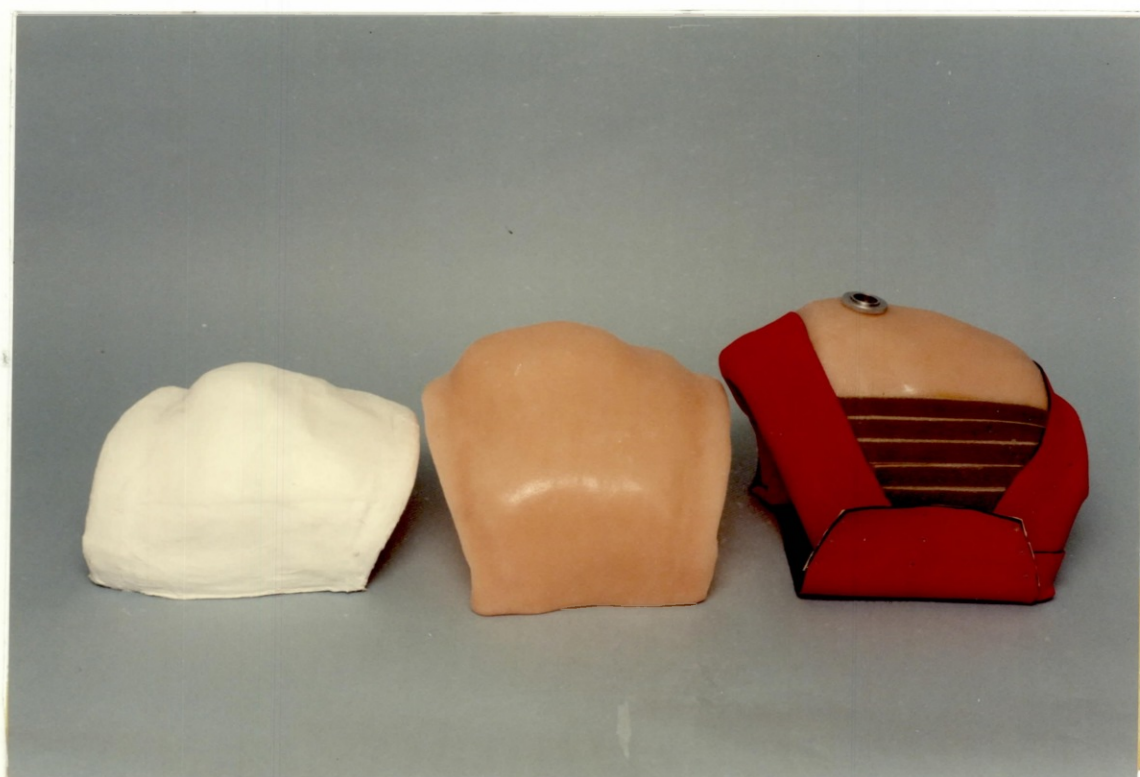
A rectangular piece of Vitrathene plastic 6mm thick is cut to the maximum length and width of the plaster cast and placed in a sheet of Stockinette to aid handling when it is hot. It is heated in an oven at 140 degrees Celsius for one hour, placed over the plaster cast and moulded to the shape of the cast. The excess Vitrathene is trimmed with a sharp knife whilst still warm, and the trimmed and moulded shell is carefully pressed down onto the shell for a few minutes until it begins to harden. It is then allowed to cool and removed from the cast.

The shell is placed over the patient again and any areas along the rims where pressure points might develop are trimmed. The areas where particular attention to this point is needed are the axillae and the iliac crests. Flexion of the hips to an angle of at least 45 degrees should be possible before the anterior aspect of the thigh touches the rim of the shell. Strips of 5 cm square foam are applied with glue to the inside edges of the cuirass. The shell is fitted to the patient again and if the foam does not touch the patient along all the borders of the shell, more foam is inserted. The foam is then covered with strips of fabric-backed Neoprene rubber. These strips are glued to the inside of the cuirass shell adjacent to the foam, pulled taught over the foam and the rim of the cuirass and then glued to the outside of the shell. A strip of Neoprene covered with material on both sides is cut to form a strap which will

pass round behind the patient. This is glued to one edge of the outside of the cuirass shell, and a Velcro fastening is glued to the shell and the strap to fix the other side. A connector is fitted centrally to the anterior portion of the cuirass.

Figure 3 shows the plaster cast after building up, the Vitrathene shell after moulding and the finished cuirass.

FIGURE 3

Stages in the Manufacture of a Cuirass Shell

3:2 Pumps Used for Negative Pressure Ventilation

Two pumps were used in these studies. The Cape cuirass pump (Figure 4) was studied as an example of a bellows pump and the Newmarket pump as an example of a fan pump (Figure 5). The Cape cuirass pump has a stroke volume of 17.6 litres. The inspiratory/expiratory ratio is fixed at 1/2 and the cycle rate can be 13, 16, 19, 22 or 25 per minute. The Newmarket pump was used exclusively in all the studies described in this thesis, with the exception of the studies of the pumps themselves which are described in chapter 11. This pump uses an industrial fan rotated by an electric motor. A valve intermittently opens the negative pressure end of the fan chamber to the inside of the cuirass. The motor altering the state of the valve is electronically controlled to produce the desired waveform. Respiratory rate, peak negative pressure, peak positive pressure and inspiratory/expiratory ratio can be altered on the pump control panel. An external signal can be fed into the pump for generating waveforms other than a simple half sinewave and for triggering of the pump by the patient.

FIGURE 4

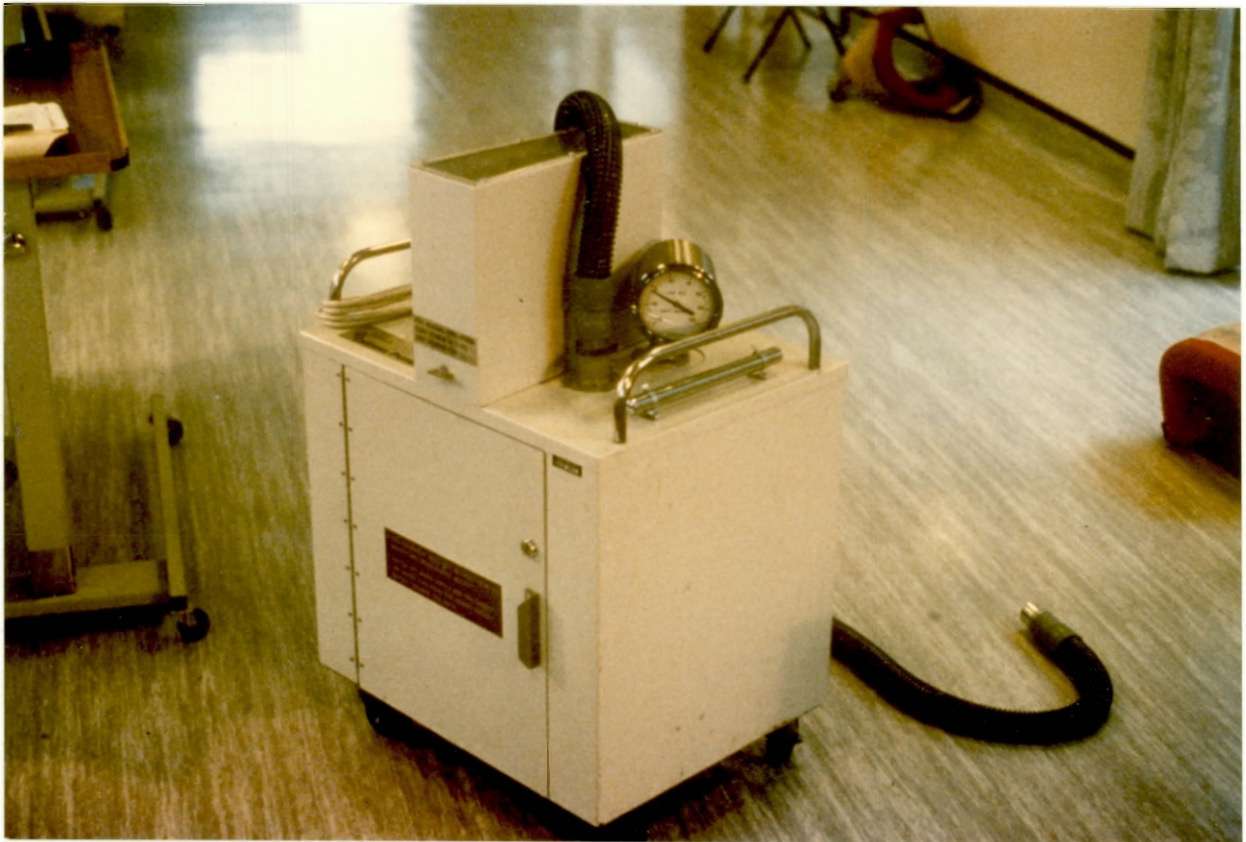
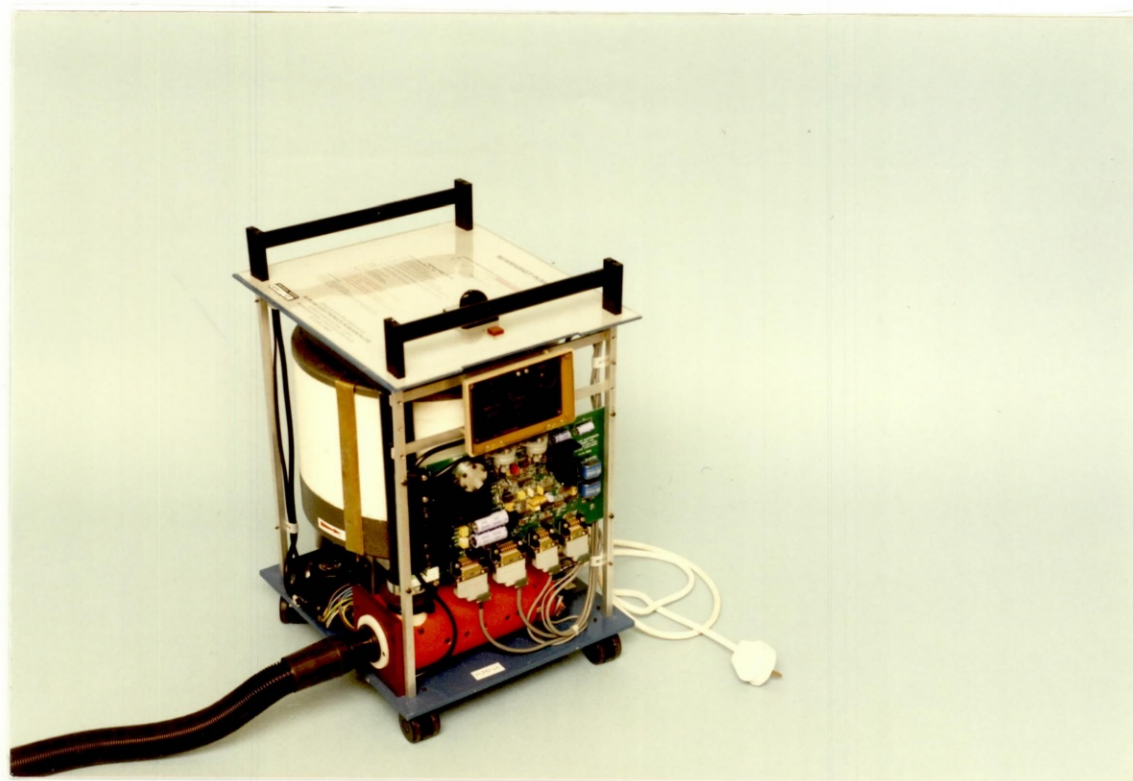
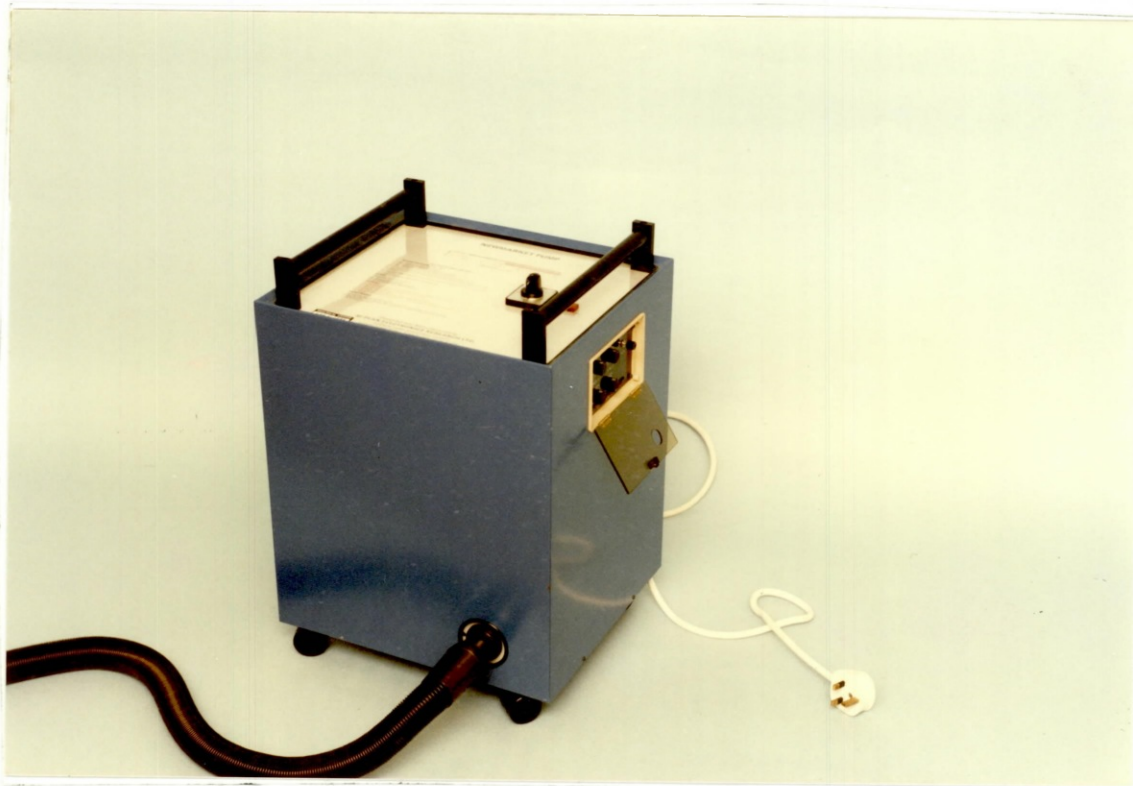
The Cape Cuirass Pump

FIGURE 5

The Newmarket Cuirass Pump

3:3 Static Lung Volumes

All lung volumes are expressed at body temperature, pressure and water saturation. Forced expiratory volume in 1 second (FEV1) and forced vital capacity (FVC) were measured with a Vitalograph wedge spirometer.

Functional residual capacity (FRC) was determined using the helium dilution technique. At the end of a tidal expiration, a tap was turned to connect the patient to a spirometer containing 2.7 litres of air, 0.3 litres of oxygen and 1 litre of helium. The rate at which oxygen was added to this closed system was adjusted to maintain a constant end-expiratory volume on the spirometer. After equilibration between the spirometer mixture and the gas in the patient's lungs, end expiratory volume was calculated automatically by the PK Morgan lung function analyser. The mean of two measurements of end-expiratory volume was taken as functional residual capacity. When these measurements were made during cuirass-assisted ventilation, the volume obtained is referred to as end expiratory volume (EEV).

Total lung capacity (TLC) was taken as the sum of inspiratory capacity and FRC, and residual volume (RV) as the difference between vital capacity (VC) and total lung capacity.

3:4 Dynamic Lung Volumes

Tidal volumes were measured using a PK Morgan wet spirometer. The resistance of this spirometer was approximately 0.01 cmH₂O/l/min. The mean of 5 breaths was used during all measurements of tidal volume during spontaneous or assisted ventilation. The electrical signal from a PK Morgan drum spirometer was used to calibrate the Respitrace, details of which are given in section 3:9.

Maximum voluntary ventilation and minute volumes were measured by integration of flow measured with a Mercury Electronics pneumotachograph head, pressure transducers and integrator. Comparison of the wet spirometer and pneumotachograph for the measurement of volume in litres revealed the following relationship:

$$\text{Pneumotachograph volume} = .871 \times \text{Spirometer Volume} + .137$$

Maximum voluntary ventilation was measured as volume inspired through a PK Morgan low resistance valve. The patient breathed at a rate chosen by themselves as deeply as possible for 15 seconds. The results were expressed as litres per minute. The inspired and expired minute volumes used in the measurement of oxygen consumption were measured over two consecutive minutes with the patient breathing through a PK Morgan low resistance valve (see section 3:11).

3:5 Pump Flow Rates

Flow was measured with a Mercury Electronics pneumotachograph flow head and pressure transducers. Factory calibrated flow heads appropriate to the flow being measured were used.

3:6 Pressures

Maximum mouth pressures were measured using a Si Plan pressure transducer calibrated with a water manometer. The response of the pressure transducer was linear up to a pressure of 110 cm of water. Maximum inspiratory pressure was recorded at the mouth with the patient making a maximal inspiratory effort from residual volume. The mouthpiece was occluded apart from a standard 22 gauge leak to reduce the effect of cheek muscles (Black and Hyatt 1969, Leech et al 1983). Maximum expiratory pressure was recorded in a similar fashion with a maximal expiratory effort from total lung capacity. The best of three attempts was recorded, each attempt consisting of a maximum pressure maintained for at least 1 second.

The Si Plan pressure transducers were also used to record the pressures generated within the cuirass shells during assisted ventilation.

3:7 Pulmonary Artery Pressure and Cardiac Output

Written informed consent was obtained for right heart catheterisation in eight subjects. A flow-directed balloon catheter was introduced under local anaesthesia through the brachial, subclavian or femoral vein and

passed into the pulmonary artery under X ray screening control. Pulmonary artery pressures were recorded using Druck pressure transducers. Thermodilution cardiac output was measured were taken as the mean of 3 injections of cool saline at each stage. All injections were made during expiration. The cardiac output was calculated by an Instrumentation Laboratories computer.

3:8 Arterial Blood Gas Analysis

Arterial samples were obtained from the radial artery. Analysis of the samples was performed using a Radiometer AB 2 Acid Base Laboratory analyser.

3:9 Ribcage and Abdominal Motion

Rib cage and abdominal motion was studied using the RespiTrace inductance plethysmograph. This system consists of two coils which are positioned round the chest and abdomen. The assumption is made that the respiratory system behaves as a two compartment model with two degrees of freedom. The gain of the signal from each coil is adjusted so that the sum of the two signals is equivalent to tidal volume.

There are several ways of calibrating the device. The first consists of utilising the difference in breathing pattern between the upright and supine positions. Using a reference volume signal from a spirometer, the volume-motion coefficients for the ribcage and abdominal signals are calculated using simultaneous equations (Sackner et al 1980, Guyatt et al

1983). The second method utilises the isovolume manoeuvre, whereby the subject moves the abdomen in and out with no net flow of air at the mouth. The gains of the ribcage and abdominal signals are adjusted until an XY plot shows no net change in volume of the Respitrace sum signal during the isovolume manoeuvre. The volume motion coefficients are then calculated by comparison of the Respitrace sum signal with a spirometer reference signal (Connellan et al 1981). A third method utilises the smaller differences between ribcage and abdominal breathing during tidal breathing. During tidal breathing in one position, Respitrace ribcage and abdominal signals and a spirometer signal are collected. The ribcage/spirometer and abdomen/spirometer factors for each point are then calculated and plotted against each other. Linear regression of these two factors allows the ribcage and abdomen volume motion coefficients to be calculated. This method is known as the indirect isovolume method (Stradling et al 1985).

The accuracy of the Respitrace system is dependent on posture, and since the two position method utilises change in posture, the direct and indirect isovolume methods are more accurate (Connellan et al 1981). The direct isovolume method underestimates the abdominal coefficient, and the indirect isovolume method appears to be superior (Stradling et al 1985). In the patients studied, the indirect isovolume method did not

produce accurate calibration values, since there was insufficient separation of ribcage and abdominal signals during tidal breathing. The isovolume manoeuvre was learnt easily by all patients, and could be performed even by patients with diaphragmatic weakness and paradoxical abdominal motion. Calibration was checked against the electrical signal from a PK Morgan drum spirometer. Statistical analysis was performed on 100 paired Respitrace and spirometer data points, sampled at a rate of 20 samples per second. Satisfactory calibration was considered to have been obtained if 95% of the Respitrace points were within 15% of the spirometer signal. If accurate calibration was not obtained, the Respitrace bands were re-positioned and the isovolume manoeuvre repeated. Calibration checks were performed at each suction pressure in the cuirass.

3:10 Inspiratory Muscle Activity

The activity of the inspiratory muscles was assessed by electromyograms of the diaphragm recorded with surface electrodes. After cleaning the the skin, five mm diameter silver cup electrodes coated in silver chloride electrode jelly were placed in the right 6th and 7th intercostal spaces in the mid-clavicular line and secured with adhesive tape. A third electrode was placed on the right wrist. The electromyographic signals were passed through an Ormed 4880 preamplifier with a frequency bandwidth of 40-10000 Hz. The raw signal was rectified

and integrated using a leaky integrator with a time constant of 0.2 s. The resultant signal was recorded on an Ormed Multi 6 chart recorder. An estimate of diaphragmatic activity was made by measuring the height of the increase in the integrated electromyogram signal during inspiration, ignoring the electrocardiographic artefact.

3:11 Oxygen Consumption

A Servomex 570A analyser was used to measure oxygen concentration and a Gould Capnograph 4 was used to measure carbon dioxide concentration. These were calibrated using air and cylinders containing either oxygen-free nitrogen or 7% carbon dioxide. Patients were studied in the non-fasting state breathing air through a mouthpiece attached to a PK Morgan low resistance valve. Inspired volume was measured over two minutes by integration of the flow signal of a pneumotachograph attached to the inspiratory side of the valve. The pneumotachograph was then switched to the expiratory side of the valve and expired volume was measured for two minutes. The expired gas was passed through a mixing chamber and the oxygen and carbon dioxide concentrations were recorded at the mid-point of the expired volume measurement. Oxygen consumption was calculated from the mixed expired oxygen concentration and inspired volume and corrected to standard temperature and pressure.

3:12 Respiratory Drive

Respiratory drive was estimated by measuring the ventilatory response to carbon dioxide by a rebreathing technique (Rebuck et al 1973, Rebuck and Campbell 1974). Patients breathed from a bag containing 6-8 litres of oxygen and the carbon dioxide was allowed to accumulate until the end-tidal level had increased by approximately 2%. Flow was measured by a Fleisch pneumotachograph head attached to a Validyne pressure transducer and integrated to give tidal volume. Carbon dioxide concentration was measured by a Centronics MGA 200 mass spectrometer and the maximum during each breath recorded. Analogue signals from the pressure transducer and mass spectrometer were passed through an Interactive Structures AI 02 analogue to digital converter into an Apple II computer. The computer sampled flow rate at 8kHz for integration and carbon dioxide concentration at 5kHz. From this data the computer derived instantaneous minute volume (IMV) in litres per minute as the product of tidal volume in litres and total breath time in seconds divided by sixty, and calculated regression equations for the relationship between end-expiratory carbon dioxide concentration and IMV.

3:13 Exercise Tolerance

After a practice walk on a separate day, the distance

walked along a flat corridor in 6 minutes was recorded. The patients were asked to walk for as far as possible in the time at their own pace, stopping when necessary. The distance was expressed in metres.

3:14 Haematology

Haemoglobin concentration and packed cell volume were measured in the haematology laboratory at Newmarket General Hospital using a Coulter model S automatic analyser.

3:15 Sleep Studies

Sleep oxygen saturation was estimated using a Hewlett Packard 47201A ear oximeter. The response of this oximeter to a step change in saturation is exponential, with a time constant of three seconds (Douglas et al 1979). Transcutaneous carbon dioxide tension (PtcCO₂) was measured by a Radiometer TCM 20 transcutaneous carbon dioxide electrode. The relationship between PaCO₂ and PtcCO₂ in kPa for this electrode was

$$\text{PtcCO}_2 = 1.04 \times \text{PaCO}_2 + 1.21$$

Patients were studied under standard conditions in the assisted ventilation unit. Their own hospital bed was moved into the monitoring area adjacent to the main ward area of the assisted ventilation unit, and they were allowed to commence the sleep study at their normal bedtime. The light from the monitoring apparatus was screened from the bed, and the area was illuminated only by ceiling night lights. Some noise from the main ward

area of the assisted ventilation unit was audible. Patients were attached to the monitoring equipment for one night prior to the sleep study to become accustomed to the apparatus. For initial studies, SaO₂ and PtcCO₂ were recorded on a chart recorder running at 1 mm per minute.

More detailed investigation of ventilation and oxygenation during sleep was undertaken in patients who showed hypoxic dips. Airflow was monitored by a Lectromed nasal thermistor in conjunction with a preamplifier (Lectromed 3543). Diaphragmatic activity was recorded by surface electromyography using the apparatus described in section 3:10. The pressure inside the cuirass shell was measured by Si Plan electronics pressure transducers.

3:16 Symptoms and Mental Function

Symptoms were assessed by a set of visual analogue scales. The scales were 10 cm long and the patients were asked to mark on the line how they felt at that time. The contrasting words at either end of the scale were as follows:

Extremely short of breath/Not short of breath at all

Not at all tired/Very tired

Happy/Sad

Where possible, patients were assessed on the morning of admission and after a night of unmonitored assisted ventilation. The distance of the patient's mark on the

line from the end of the line which denoted the ideal state was measured.

Depression was assessed by an adapted depression questionnaire which is shown in Appendix 2. Four tests were used to assess mental function, the time taken to complete them being recorded:

1 The trail test: Subjects were asked to draw a line between consecutive numbers 1 to 25 scattered at random over the page.

2 The deletion test: Subjects were asked to delete a specified letter from a page of columns of letters in random order.

3 The copying test: Subjects were asked to copy columns of figures on the top half of a page onto the bottom half.

4 The decoding test: From a code of paired numbers and letters, subjects were asked to choose the appropriate letter to pair with a set of numbers.

3:17 Recording Apparatus, Statistics and Computing

Recordings were made on the six channels of a Lectromed MT 6 chart recorder in conjunction with a MT6 PX preamplifier cabinet. Digital storage of selected signals onto floppy disc was made using a Si Plan electronics universal interface analogue to digital converter attached to the 1 MHz bus of a BBC B microcomputer.

All statistical tests were carried out on the BBC B

microcomputer using the statistical package for the BBC microcomputer released by the Department of Applied Statistics at the University of Reading (INSTAT). The significance of data was evaluated by using Wilcoxon rank sum, Mann Whitney, Spearman rank correlation and Fisher's exact tests (Broughton Pipkin 1984). Repeatability was assessed by calculation of coefficients of repeatability (Bland and Altman 1986).

3:18 Anthropometric Measurements

Arm span was measured with a tape measure in each subject. It was used in preference to height in the calculation of predicted values for lung volumes from published regression equations (Quanjer et al 1983). The angle of spinal curvature was calculated from lateral chest radiographs using the method of Cobb (Cobb 1948). Ideal weights for age, sex and height were taken from standard tables (Geigy Scientific Tables 1970).

3:19 Subjects

Twenty five subjects with chest wall disease participated in the studies described in this thesis. All had been referred to the Assisted Ventilation Unit at Newmarket General Hospital. Fourteen of the patients were female. Table 1 shows the anthropometric details of the subjects. Satisfactory radiographs for the calculation of the Cobb angle were not available for one of the patients. The age when cuirass-assisted ventilation was started ranged from 15 to 68 with a median of 53 years.

Six subjects had an idiopathic thoracic scoliosis, and one had a thoracic kyphosis following tuberculosis of the spine at the age of 3 years.

Three patients were referred on account of respiratory symptoms occurring following poliomyelitis. None had required assisted ventilation during the acute childhood infection. Two of these subjects also had a significant thoracic scoliosis. All 3 had limb muscle weakness, one being wheel chair bound, one walking with a brace and stick, and one walking unaided.

Seven subjects had neuromuscular diseases affecting their respiratory muscles. Four had a proximal myopathy, one had a familial limb girdle muscular dystrophy, one had motor neurone disease and one had Duchenne muscular dystrophy. The patient with Duchenne muscular dystrophy had a scoliosis, and three of the patients with proximal myopathies had scoliosis sufficiently severe to require spinal fusion. The patients with Duchenne muscular dystrophy, familial limb girdle muscular dystrophy and one of the patients with an unclassified proximal myopathy were confined to wheelchairs, whilst the other patients with neuromuscular disease were able to walk unaided.

Eight patients had had a thoracoplasty performed previously for tuberculosis, on the left side in four patients, on the right side in three patients, and bilaterally in one patient (KN). In only one patient was

thoracoplasty accompanied by lung resection (NW). All 8 patients had received artificial pneumothorax treatment prior to the thoracoplasty, and three had been treated by phrenic nerve crush. The median number of ribs resected was 7.5 (range 6 to 9).

TABLE 1

Anthropometric details of subjects.

Subject	Disease	Age (y)	Sex	Arm Span (m)	Spinal Curvature (degrees)	Weight (% ideal)
HH	IS	60	M	1.90	120	90
SS	IS	50	F	1.68	75	69
AP	IS	46	M	1.46	70	60
MB	IS	59	F	1.45		57
SG	IS	34	F	1.08	90	19
BC	IS	45	M	1.68	135	71
WW	K	53	M	1.73	0	75
AV	P	53	F	1.48	100	57
BS	P	41	M	1.70	70	73
YH	P	51	F	1.14	10	69
MS	NM	42	F	1.70	120	68
JG	NM	52	F	1.62	0	65
GP	NM	23	M	1.67	5	64
RB	NM	36	M	1.65	0	68
TC	NM	21	F	1.64	70	56
RR	NM	15	M	1.25	85	24
JL	NM	66	M	1.73	0	74
WB	T	68	M	1.83	20	83
MH	T	63	F	1.62	20	66
DH	T	54	F	1.52	50	59
IJ	T	57	F	1.51	25	58
NW	T	68	F	1.51	70	59
DS	T	64	F	1.56	50	61
KN	T	59	M	1.74	0	76
HC	T	62	F	1.68	15	69

IS=Idiopathic scoliosis

K=Kyphosis

P=Poliomyelitis

NM=Neuromuscular

T=Thoracoplasty

Lung volumes are given in Table 2 and expressed as percentage of values predicted from age, sex and arm span in Table 3. All subjects had a vital capacity of under 2 litres, and 15 subjects had a vital capacity of under 1 litre. Vital capacity was less than 50% of predicted in all subjects and less than 25% of predicted in 9. The 9 patients with a vital capacity of less than 25% predicted included all 3 patients with poliomyelitis, 3 of the 6 patients with other neuromuscular disease, 1 with scoliosis, 1 with a thoracoplasty and the patient with kyphosis secondary to spinal tuberculosis.

The reduction in total lung capacity was similar to the reduction in vital capacity except in 2 patients (SG and RB). Both these subjects had residual volumes greater than predicted, although neither had clinical evidence of airflow limitation and their ratios of FEV₁ to FVC were high. Residual volume and functional residual capacity were less than predicted values in all other subjects, but the reduction in residual volume and functional residual capacity was less than the reduction in vital capacity and total lung capacity.

TABLE 2

Observed Lung Volumes of Subjects.

Subject	FEV1 (l)	FVC (l)	TLC (l)	RV (l)	FRC (l)	FER (%)	RV/TLC (%)
MS	.93	1.05	1.62	.57	.85	88	35
HH	.85	1.10	2.30	1.15	1.42	77	50
SS	.40	.77	1.62	.85	.85	51	52
AP	.70	.90	1.20	.33	.51	77	27
MB	.60	.85	1.99	.83	1.29	71	41
SG	.26	.26	1.19	.75	.86	100	63
BC	.93	1.27	2.30	1.23	1.60	73	53
WW	.35	.65	1.49	.84	.88	53	56
AV	.35	.41	1.16	.72	.75	85	62
BS	.90	.95	1.60	.65	.99	94	40
YH	.44	.50	1.06	.84	1.06	88	80
JG	.39	.39	1.24	.76	.85	100	61
GP	.39	.39	1.62	1.30	1.34	100	80
RB	1.65	1.70	4.20	2.50	2.20	97	59
TC	1.00	1.10	1.84	.74	1.18	90	40
RR	.30	.44	.85	.41	.52	68	48
JL	.80	1.60	3.30	2.40	3.40	50	72
WB	.70	1.75	3.59	1.50	2.10	40	42
MH	.25	.55	1.80	1.25	1.28	45	69
DH	.60	1.05	2.05	1.00	1.39	57	48
IJ	.30	.99	2.30	1.31	1.53	30	56
NW	.65	.91	1.80	.89	1.11	71	49
DS	.29	.66	1.80	1.14	1.25	43	48
KN	.55	1.30	2.32	1.07	1.44	42	46
HC	.45	1.20	2.70	1.52	1.73	46	56

Mean	.61	.91	1.96	1.06	1.30	69	53
Median	.57	.93	1.80	.89	1.25	71	57

TABLE 3

Lung Volumes of Subjects Expressed as
Percentage of Predicted Values.

Subject	FVC (%)	TLC (%)	RV (%)	FRC (%)
MS	31	29	32	29
HH	21	28	44	36
SS	25	30	46	30
AP	31	26	19	18
MB	47	52	52	55
SG	26	88	150	59
BC	30	43	62	49
WW	15	22	38	25
AV	19	29	47	31
BS	21	24	34	30
YH	17	21	47	39
JG	14	25	43	31
GP	8	25	91	47
RB	40	68	145	71
TC	30	36	56	45
RR	17	34	55	26
JL	40	58	96	95
WB	39	47	56	55
MH	22	36	64	47
DH	46	48	61	56
IJ	46	55	79	62
NW	49	43	48	45
DS	30	40	62	48
KN	32	24	47	40
HC	44	51	75	62

Mean	30	39	62	45
Median	30	36	55	45

Table 4 shows the arterial blood gases sampled at rest with the subjects breathing air. Twenty three of the subjects had a PaCO₂ greater than 6 kPa, and only two had a PaO₂ of 10 kPa or greater. The maximum mouth pressures for the subjects are given in Table 5. Maximum inspiratory pressures were less than 50 cmH₂O in nineteen subjects and less than 25 cmH₂O in five subjects.

Satisfactory studies of gas exchange during sleep were obtained prior to commencing nocturnal cuirass-assisted ventilation in only thirteen subjects. These subjects were studied after several nights in the Assisted Ventilation Unit, during at least one of which they had been connected to the ear oximeter and transcutaneous carbon dioxide electrode. In two subjects, nocturnal assisted ventilation had already been commenced at another hospital using intermittent positive pressure ventilation through a tracheostomy. In five subjects, it was necessary to commence assisted ventilation as soon as the patients were referred to the Assisted Ventilation Unit. In the remaining five subjects, it was not possible to record satisfactory sleep studies either because the subjects were unable to sleep for more than a few minutes at a time in the environment of the Assisted Ventilation Unit, or because the ear oximeter and transcutaneous carbon dioxide electrode were needed to monitor other subjects admitted with acute respiratory failure. Of the thirteen subjects studied, all showed a

rise in $P_{tc}CO_2$ to levels greater than 7 kPa. If the relationship between P_aCO_2 and $P_{tc}CO_2$ in the awake state described in Chapter 3 applies during sleep, this would indicate a rise in P_aCO_2 to levels of greater than 6 kPa. In twelve of the subjects, $P_{tc}CO_2$ remained elevated above 7 kPa for a proportion of the sleep time sufficient to result in a mean $P_{tc}CO_2$ of 7 kPa or greater. The oxygen desaturation which accompanied this hypoventilation was variable. One subject (KN) maintained his oxygen saturation throughout the night with a minimum saturation of 79%. At the other extreme, two subjects (DH,DS) spent more than half of their sleep time with saturations less than 60%. Both these subjects were severely hypoxaemic during the daytime. The remaining ten subjects showed episodes of oxygen desaturation during sleep, with minimum saturations falling below 50% in six of the subjects.

TABLE 4

Daytime arterial blood gases of subjects
breathing air.

	PaCO ₂ (kPa)	PaO ₂ (kPa)
MS	7.3	9.0
HH	8.6	8.1
SS	7.9	6.8
AP	9.5	5.3
MB	7.9	8.1
SG	10.9	5.5
BC	6.4	5.3
WW	8.5	6.1
AV	7.2	8.4
BS	7.6	5.5
YH	7.7	6.4
JG	7.9	7.4
GP	10.3	7.9
RB	7.1	10.0
TC	6.1	8.5
RR	9.4	7.5
JL	8.7	6.2
WB	7.5	7.2
MH	8.6	8.1
DH	7.7	7.3
IJ	6.6	7.8
NW	6.3	9.6
DS	8.9	5.2
KN	5.4	11.2
HC	5.9	6.4

Mean	7.8	7.4
Median	7.7	7.4

TABLE 5

Maximum inspiratory (MIP) and maximum expiratory (MEP) mouth pressures of subjects.

Subject	MIP cmH ₂ O	MEP cmH ₂ O
MS	59	54
HH	33	60
SS	23	10
AP	46	16
MB	19	34
SG	20	36
BC	25	42
WW	37	53
AV	43	59
YH	32	36
JG	25	46
GP	64	33
RB	30	58
TC	33	39
RR	25	44
JL	9	37
WB	34	110
MH	29	31
DH	32	53
IJ	51	53
NW	16	21
DS	32	37
KN	81	106
HC	53	74

Mean	35	48
Median	32	40

TABLE 6

Mean and maximum transcutaneous carbon dioxide tension (PtcCO₂) during sleep without assisted ventilation.

Subject	Mean PtcCO ₂ (kPa)	Maximum PtcCO ₂ (kPa)
MS	8.7	10.4
SS	10.5	11.7
AP	9.1	10.5
WW	8.2	8.9
AV	7.9	8.8
JG	8.2	8.8
GP	8.4	9.5
DH	11.6	12.9
IJ	7.0	8.1
DS	8.5	8.9
NW	10.8	15.7
KN	8.8	9.3
HC	6.5	7.6
Mean	8.8	10.2
Median	8.7	9.3

TABLE 7

Ear oxygen saturation during sleep without assisted ventilation.

Subject	Sleep time (min)	Percentage of sleep time at saturation:					Minimum Saturation
		<90%	<80%	<70%	<60%	<50%	
MS	355	92	18	5	2	<1	45
SS	430	100	97	29	3	1	28
AP	280	100	91	34	4	1	29
WW	180	100	91	19	<1	<1	59
AV	425	15	4	<1	0	0	66
JG	235	97	3	1	0	0	61
GP	305	93	22	5	0	0	66
DH	355	99	90	81	74	12	21
IJ	375	100	41	4	<1	0	53
DS	425	100	100	99	99	94	5
NW	445	100	80	4	1	0	54
KN	145	54	<1	0	0	0	79
HC	230	100	100	23	1	<1	45
Mean	322	88	57	23	14	8	47
Median	355	100	80	5	1	0	53

Table 8 shows the indications for nocturnal cuirass-assisted ventilation in each subject. Symptoms of nocturnal hypoventilation and clinical evidence of respiratory muscle fatigue were not recorded in a sufficient number of subjects to warrant inclusion as indications. Polycythaemia and clinical evidence of pulmonary hypertension could be explained by daytime respiratory failure in the majority of subjects, and could not therefore be considered as complications of nocturnal hypoventilation alone. All of the thirteen subjects studied showed nocturnal alveolar hypoventilation, including the two subjects who were normocapnic during the daytime.

Twenty three of the twenty five patients had daytime hypercapnia, and all patients had a vital capacity of less than 50% predicted. As discussed in the previous chapter, Rochester and Martin suggested a maximum inspiratory mouth pressure of less than 50 cmH₂O be taken as an indication for assisted ventilation in subjects with idiopathic scoliosis and subjects with neuromuscular disease complicated by scoliosis, or less than 25 cmH₂O in subjects with neuromuscular disease uncomplicated by scoliosis (Rochester and Martin 1977). Applying the same critical level of respiratory muscle weakness for scoliosis to patients with a thoracoplasty, sixteen of the twenty four subjects studied fulfilled these criteria of inspiratory muscle weakness as an

indication for assisted ventilation. Two subjects had been commenced on intermittent positive pressure ventilation through a tracheostomy prior to referral, and attempts to wean them from nocturnal assisted ventilation had failed.

TABLE 8

Indications for nocturnal cuirass-assisted ventilation.

Subject	Sleeping PtcCO ₂ >6.9 kPa	Daytime PaCO ₂ >5.9 kPa	MIP <25cmH ₂ O (neuromuscular) or <50cmH ₂ O (kyphoscoliosis or thoracoplasty)	FVC <50% predicted	Failure to wean from IPPV
MS	+	+		+	
HH		+	+	+	
SS	+	+	+	+	
MB		+	+	+	
AP	+	+	+	+	
SG		+	+	+	
BC		+	+	+	
WW	+	+	+	+	
AV	+	+	+	+	
BS		+		+	
YH		+		+	
JG	+	+		+	
GP	+	+		+	
RB		+		+	+
TC		+	+	+	
RR		+	+	+	
JL		+	+	+	+
WB		+	+	+	
MH		+	+	+	
DH	+	+	+	+	
IJ	+	+		+	
NW	+	+	+	+	
KN	+			+	
HC	+			+	
DS	+	+	+	+	

CHAPTER 4

CUIRASS-ASSISTED VENTILATION AND DAYTIME VENTILATION

4:1 INTRODUCTION

The ability of cuirass respirators to provide effective ventilation will depend on the design of the cuirass shell, the respiratory mechanics of the patient, the contribution of the patient's own respiratory muscles to ventilation, and the setting of the cuirass respirator pump. Although studies of ventilation using different cuirass designs were described in chapter 1, these were mainly conducted on patients with acute poliomyelitis (Kelleher et al 1952, Scales et al 1953, Collier and Affeltdt 1954, Cardus et al 1966) or normal subjects (Bryce-Smith and Davis 1954, Wallace et al 1961). The ventilation which can be achieved with an individually moulded cuirass in a patient with chest wall deformity has only been studied in isolated cases (Fulkerson et al 1984).

Fitting a cuirass, the rims of which rest on the upper chest and lower abdomen, to a patient with chest wall disease may restrict expansion and decrease total respiratory system compliance. If cuirass-assisted ventilation is to be effective, the assistance provided by external negative pressure must outweigh any restriction of chest wall expansion. Some early designs of cuirass were fitted with legs or a gantry in an attempt to try and reduce downward movement of the

cuirass onto the patient during the negative pressure phase of the cycle. This downward motion is most apparent when larger peak external negative pressures are used.

The individually moulded cuirass studied in this thesis was constructed with sides extending down onto the bed to try to reduce downward movement of the cuirass. The extent to which restriction of chest wall expansion occurred was studied by recording the tidal volumes achieved with increasing peak external negative pressure, comparing these tidal volumes to those obtained with jacket respirators which do not touch the chest wall, and measuring the end-expiratory volume during cuirass-assisted ventilation compared to that during spontaneous ventilation. The motion of the chest and abdomen was studied by inductance plethysmography, and arterial blood gas analysis was performed to ascertain whether any change in overall ventilation was accompanied by a change in alveolar ventilation.

Positive pressure can be used to aid expiration during cuirass-assisted ventilation (Bryce-Smith and Davis 1954, Binet et al 1961, O'Leary et al 1979, Holtackers et al 1982). The value of this in terms of additional ventilation achieved has not been studied other than in patients with poliomyelitis (Binet et al 1961). This chapter also describes the change in tidal volume and the effect on end-expiratory volume of external positive

expiratory pressure during cuirass-assisted ventilation in patients with chest wall disease.

The rate of respiration during spontaneous ventilation in patients with chest wall disease is usually increased compared to normal subjects. Coordination between the patient's own respiratory efforts and the applied external negative pressure cycle during cuirass-assisted ventilation is often best achieved by setting the rate of the negative pressure pump close to the patient's spontaneous respiratory rate. The effect of respiratory rate on the ventilation achieved with cuirass-assisted ventilation is also examined in this chapter.

4:2 SUBJECTS and METHODS

4:2:1 Subjects

Seventeen subjects were studied using individually moulded cuirass shells. All subjects had been using cuirass-assisted ventilation at night for at least two weeks, and were in a clinically stable state.

4:2:2 The Effect of Peak External Negative Pressure, External Positive Expiratory Pressure and Respiratory Rate on Tidal Volume

Tidal volume was taken as the mean of five consecutive breaths measured using a mouthpiece attached to a PK Morgan wet spirometer. The subject lay supine for 15 minutes, and spontaneous tidal volume was then recorded. The cuirass was applied and connected to a Newmarket pump running at a rate of 15 breaths per minute

with an inspiratory:expiratory ratio of 1:1. The peak negative pressure was measured inside the cuirass shell using a Si Plan pressure transducer. A period of five minutes was allowed between each change in the setting of the pump to enable the tidal volume to stabilise. Tidal volume was measured with a peak external negative pressure of 10 cmH₂O. The peak negative pressure was then increased in steps of 10 cmH₂O up to 40 cmH₂O with repeat measurements of tidal volume at each setting. The maximum external positive pressure which could be generated by the pump was then introduced into the expiratory phase of the respiratory cycle, and the tidal volume measurements repeated in conjunction with peak external negative pressures of 10, 20, 30 and 40 cmH₂O in turn. External positive pressure was then removed and the peak external negative pressure reset to 30 cmH₂O. Tidal volume was measured at respiratory rates of 10, 15, 20, 25 and 30 breaths per minute. The tidal volumes obtained were multiplied by the respiratory rate to derive minute volume.

4:2:3 Arterial Blood Gases

On a different day from the measurement of tidal volume, arterial blood was sampled from the radial artery with the subject breathing spontaneously in the supine position. The sample was analysed immediately using a Radiometer blood gas laboratory analyser. Cuirass-assisted ventilation was then commenced using

an inspiratory:expiratory ratio of 1:1 with a peak external negative pressure of 30 cmH₂O, without external positive pressure, at a respiratory rate of 15 breaths per minute. After 30-60 minutes arterial blood was sampled again and analysed in the same way.

4:2:4 End-expiratory Volume

On a separate day, resting end-expiratory volume in the supine position was measured by helium dilution. The subject's cuirass was then fitted and cuirass-assisted ventilation commenced at a respiratory rate of 15 breaths per minute with an inspiratory:expiratory ratio of 1:1. A peak external negative pressure of 30 cmH₂O was used, initially without external positive pressure. After fifteen minutes, end-expiratory volume was measured again. External positive pressure was then applied up to the maximum positive pressure which could be achieved, or the maximum which could be tolerated by the patient. After a further fifteen minutes, end-expiratory volume was measured again.

4:2:5 Ribcage and Abdominal Motion

The motion of the ribcage and abdomen was measured over 5 breaths using the Resptrace inductance plethysmograph, calibrated as described in chapter 3. Inductance coils were placed around the chest immediately below the level of the manubrio-sternal angle and around the abdomen at the level of the umbilicus. Recordings were made during spontaneous ventilation after the

subjects had been resting in the supine position for fifteen minutes. Cuirass-assisted ventilation was commenced at a rate of fifteen breaths per minute with an inspiratory:expiratory ratio of 1:1 and peak external negative pressure of 30 cmH₂O. After a further fifteen minutes, RespiTrace recordings of ribcage and abdominal motion were repeated.

4:2:6 Comparison with Jacket Respirators

The tidal volumes achieved with the cuirass were compared in some subjects with three other external negative pressure ventilators. The Tunnickliff jacket was described in Chapter 1. It consists of a backplate and anterior shell which covers the chest and upper abdomen. The shell does not touch the subject, and an airtight seal is obtained by a plastic overgarment. The Emerson respirator also consists of an airtight overgarment and a rigid shell which does not touch the subject. The shell of the Emerson respirator is larger and covers the thorax from the region of the clavicles to the suprapubic region.

Since both of these jacket respirators cover a different area of the thorax to the cuirass, a third type of jacket respirator was constructed. A shell covering an identical area of the thorax to the cuirass was manufactured, using the same technique as described in section 3:1. The plaster cast of the patient was built up further than for the cuirass, so that the moulded

shell did not touch the patient at any point. This shell was covered by a plastic airtight overgarment similar to that used with the other two jacket respirators.

The cuirass and these three jacket style respirators were used in random order in each patient. Tidal volumes were measured at peak external negative pressures of 10, 20, 30 and 40 cmH₂O with each jacket, using a respiratory rate of 15 breaths per minute with an inspiratory:expiratory ratio of 1:1.

4:3 RESULTS

4:3:1 The Effect of Peak External Negative Pressure, External Positive Expiratory Pressure and Respiratory Rate on Tidal Volume

The tidal volumes obtained using peak external negative pressures between 10 and 40 cmH₂O are given in Table 9. The changes in tidal volume on adding expiratory external positive pressure are given in Table 10. There was no significant increase in tidal volume with the addition of external positive expiratory pressure. The effect of respiratory rate on tidal volume is shown in Table 11 and on minute volume in Table 12.

4:3:2 Arterial Blood Gases

Table 13 gives the arterial blood gas tensions during spontaneous and cuirass-assisted ventilation. PaCO₂ was significantly lower (Wilcoxon signed rank sum 14, $p < 0.01$) and PaO₂ significantly higher (Wilcoxon signed rank sum 15.5, $p < 0.05$) during cuirass-assisted ventilation compared to spontaneous ventilation.

4:3:3 End-expiratory volume

End-expiratory volumes during spontaneous and cuirass-assisted ventilation are listed in Table 14. End-expiratory volume during cuirass-assisted ventilation using external negative pressure alone was not significantly different from that recorded during spontaneous ventilation. The addition of external positive expiratory pressure produced a significant fall in end-expiratory volume (Wilcoxon signed rank sum 10, $p < 0.05$).

4:3:4 Ribcage and Abdominal Motion

During spontaneous ventilation, four subjects (SS,AP,WB,RB) had paradoxical inward motion of the abdomen during inspiration. Two subjects with thoracoplasties (HC,IJ) had paradoxical inward motion of the ribcage when breathing spontaneously. During cuirass-assisted ventilation, paradoxical motion was corrected in all but one subject (IJ). Two subjects (JG,MB) had synchronous ribcage and abdominal expansion during spontaneous respiration, but developed paradoxical inward motion of the ribcage during cuirass-assisted ventilation. The remaining three subjects studied had synchronous expansion of ribcage and abdomen during both spontaneous and cuirass-assisted ventilation.

4:3:5 Comparison with Jackets

The tidal volumes obtained with the different types

of jacket compared to the tidal volumes obtained with the cuirass at the same peak external negative pressure are given in Tables 15 to 17. None of the differences in tidal volume were significantly different from those obtained during cuirass-assisted ventilation, except for the Tunncliffe jacket using a peak external negative pressure of 20 cmH₂O (Wilcoxon signed rank sum 6, $p < 0.05$).

TABLE 9

Tidal volumes (mls) obtained during spontaneous ventilation (SV) and cuirass-assisted ventilation (CAV) using peak external negative pressures of 10 to 40 cmH₂O at a respiratory rate of 15 breaths per minute.

Subject	SV	CAV			
		10	20	30	40
MS	200	378	394	477	
HH	304	418	418	449	388
AP	403	346	296	313	
MB	224	262	274	289	
WW	459	489	501	495	489
BS	246	231	242	298	340
AV	220	307	433	412	453
RB	266	384	433	597	758
JG	202	245	318	319	
TC	349	303	340	426	354
MH	305	340	340	316	
WB	490	543	591	656	716
IJ	292	411	418	442	457
DH	245	303	332	310	
DS	378	315	349	396	
KN	495	650	698	794	817
HC	320	287	340	322	
Mean	317	365	395	430	530
Median	304	340	394	412	457

TABLE 10

Change in tidal volume (mls) during cuirass-assisted ventilation on adding maximum expiratory external positive pressure compared to tidal volumes obtained using the same peak external negative pressure (cmH₂O) alone.

Subject	Peak external negative pressure:			
	10	20	30	40
MS	+60	+66	+39	
HH	+134	+90		
MB	+99	+60	+36	
AP	-24	+68	+63	
WW	+140	+100		
AV	+100	-1	+57	+52
BS	+20	+30	+51	+93
RB	+242	+397	+361	+176
JG	-4	-73	-70	-1
RR	+63	-33		
GP	+16	-30		
WB	-27	-36	-29	
IJ	-25	+10	-20	-30
HC	+98	+169	+132	-3
KN	+32	+18	-36	-51
DS		-42	-20	-24

Mean	+62	+50	+47	+26
Median	+60	+24	+37	-2

TABLE 11

Tidal volumes (mls) obtained using a peak external negative pressure of 30 cmH₂O with a rate between 10 and 30 breaths per minute.

Subject	Respiratory rate:				
	10	15	20	25	30
HH	503	522	507	427	408
MB	424	358	322	245	289
AP	331	385	301	289	259
AV	429	433	372	352	337
BS	534	448	298	260	233
JG	296	318	216	218	211
RR	218	248	167	155	158
GP	289	305	234	178	285
WB	740	612	445	322	361
IJ	537	418	310	324	246
HC	588	346	316	334	352
KN	889	698	612	594	510
DS	388	355	322	300	257
Mean	474	419	340	307	300
Median	429	385	316	300	285

TABLE 12

Minute volumes (l/min) obtained using a peak external negative pressure of 30 cmH₂O with an inspiratory/expiratory ratio of 1/1 with a respiratory rate between 10 and 30 breaths per minute.

Subject	Respiratory rate:				
	10	15	20	25	30
HH	5.03	7.83	10.14	10.67	12.24
MB	4.24	5.37	6.44	6.12	8.67
AP	3.31	5.77	6.02	7.22	7.77
AV	4.29	6.49	7.44	8.80	10.11
BS	5.34	6.72	5.96	6.50	6.99
JG	2.96	4.77	4.32	5.45	6.33
RR	2.18	3.73	3.34	3.87	4.74
GP	2.89	4.57	4.68	4.45	8.55
WB	7.40	9.18	8.90	8.05	10.83
IJ	5.37	6.27	6.20	8.10	7.38
HC	5.88	5.19	6.32	8.35	10.56
KN	8.89	10.47	12.24	14.85	15.30
DS	3.8	5.32	6.44	7.50	7.71

Mean	4.74	6.28	6.80	7.69	9.00
Median	4.29	5.77	6.32	7.50	8.55

TABLE 13

Arterial blood gases during spontaneous ventilation (SV) and after 30-60 minutes of cuirass-assisted ventilation (CAV).

Subject	PaCO ₂ (kPa)			PaO ₂ (kPa)		
	SV	CAV	change	SV	CAV	change
MS	6.1	5.2	-0.9	9.4	11.2	+1.8
HH	6.5	5.5	-1.0	10.2	11.9	+1.7
MB	7.9	7.2	-0.7	8.0	8.2	+0.2
SS	7.2	8.0	+0.8	12.9	13.1	+0.2
BC	6.3	4.6	-1.7	5.6	6.2	+0.8
WW	7.1	6.2	-0.9	8.3	9.2	+0.9
AV	6.3	5.7	-0.6	9.5	10.0	+0.5
JG	7.9	7.7	-0.2	7.4	7.9	+0.5
JL	5.6	5.1	-0.5	8.0	8.0	0
MH	6.5	6.7	+0.2	8.8	8.1	+0.7
WB	7.2	6.1	-1.1	7.6	8.3	+0.7
DH	6.6	6.5	-0.1	13.4	12.6	-0.8
IJ	7.4	7.1	-0.3	7.2	8.1	+0.9
DS	8.9	9.0	+0.1	5.2	4.8	-0.4
HC	6.0	4.8	-1.2	7.8	7.9	+0.1
KN	5.2	4.3	-0.9	10.0	12.1	+2.1
Mean	6.8	6.2	-0.6	8.7	9.2	+0.6
Median	6.5	6.1	-0.65	8.1	8.2	+0.6

TABLE 14

End-expiratory volume (l) during spontaneous ventilation (SV), cuirass-assisted ventilation (CAV) using external negative pressure alone (ENP) with a peak external negative pressure of 30 cmH₂O, and in conjunction with maximum expiratory external positive pressure (EPP).

Subject	End-expiratory volume:		
	SV	CAV:ENP	CAV:ENP+EPP
HH	1.23	1.41	1.08
MB	.66	.59	.63
RB	2.09	2.33	1.91
WW	1.08	1.34	1.14
JG	.92	.92	.87
TC	.99	.92	.89
SS	2.09	2.18	2.10
RR	.62	.60	.49
MS	.63	.60	.57
BS	.77	.74	.90
WB	1.73	2.49	1.62
BC	1.00	1.52	.99

Mean	1.15	1.30	1.10
Median	.99	1.13	0.94

TABLE 15

Difference in tidal volume (mls) using the Tunncliffe jacket compared to the cuirass at the same peak external negative pressure.

Subject	Peak external negative pressure (cmH ₂ O):			
	10	20	30	40
MS	+41	+89	-6	
HH	-60	+119	+88	
RB	-92	-51	-161	-271
TC	+72	+170	+138	
MH	-5	+33	+83	
IJ	-7	+30	+24	-28
DH	+20	+15	+33	
DS	+139	+39	+96	
KN	+90	+24	-66	
HC	+20	+117	+125	+8

Mean	+22	+58	+35	-97
Median	+20	+36	+58	-28

TABLE 16

Difference in tidal volume (mls) using the Emerson respirator compared to the cuirass at the same peak external negative pressure.

Subject	Peak external negative pressure (cmH ₂ O):			
	10	20	30	40
MS	+89	+107		
WW	+14	-54		
RB	+167	+181	+149	
TC	-67	+149	+159	
WB	+24	+41		
IJ	+4	+5	+31	+15
DH	-56	-5	-1	
DS	+52	+22		
HC	+56	+14	+167	-1

Mean	+31	+51	+101	
Median	+24	+22	+149	

TABLE 17

Difference in tidal volume (mls) using a moulded jacket compared to the cuirass at the same peak external negative pressure.

Subject	Peak external negative pressure (cmH ₂ O):			
	10	20	30	40
MS	+1	+19		
WW	-10	-54		
RB	+125	+175	+208	
TC	+144	+255	+213	
MH	-66	-36	-18	
WB	+65	0		
IJ	-25	-13	-75	-55
DH	-15	+62	+84	
DS	+80	+20	+61	
HC	+26	+27	+79	

Mean	+32	+45	+79	
Median	+13	+19	+79	



4:4 DISCUSSION

Previous studies of the tidal volumes achieved during cuirass-assisted ventilation involved patients with severe respiratory muscle weakness following poliomyelitis (Collier and Affeldt 1954) or normal subjects paralysed during general anaesthesia (Bryce-Smith and Davis 1954, Wallace et al 1961) in whom little spontaneous respiratory muscle activity was possible. The extent to which spontaneous respiratory muscle activity influenced the tidal volumes obtained in the studies reported in this chapter was not measured. As demonstrated by Rochester et al (1977) and in the studies described later in this thesis, spontaneous respiratory muscle activity during cuirass-assisted ventilation is reduced compared to spontaneous ventilation. Although this is true when an adequate peak external pressure is used at a comfortable respiratory rate for each subject, spontaneous respiratory muscle activity is likely to be greater when a smaller peak external pressure is used, when the respiratory rate is uncomfortable for the patient, or when they are distressed, for instance by an uncomfortably high external positive pressure. Even using a comfortable respiratory rate and adequate peak external pressure, the use of a mouthpiece to make the recordings of tidal volume may produce an increase in spontaneous respiratory muscle activity (Rochester et al 1977).

For these reasons, the tidal volumes achieved during the studies described in this chapter are likely to be considerably larger than those which would have been obtained if the patients had been paralysed. They are also likely to be larger than those which would have been obtained in the presence of the patients' normal respiratory muscle activity, unaugmented by voluntary effort induced by the study. In only one subject (AV) was a tidal volume achieved during cuirass-assisted ventilation which was greater than spontaneous vital capacity.

Cuirass-assisted ventilation is used principally during sleep. Even if spontaneous respiratory muscle activity during sleep remains in phase with the applied negative pressure cycle, the tidal volumes will probably be smaller than those obtained in these awake studies. Loss of the voluntary augmentation of resting respiratory muscle activity and an increase in upper airway resistance associated with sleep will both reduce the tidal volumes. If asynchrony develops between the respiratory muscles and the applied negative pressure cycle, or if spontaneous respiratory muscle activity ceases, the cuirass will be acting alone in a manner analogous to the studies in paralysed patients mentioned above. The tidal volumes produced in this situation may be inadequate to maintain normal gas exchange. These points will be considered further in the discussion of

sleep in chapter 8.

In most subjects, increasing the peak external negative pressure produced larger tidal volumes. Five subjects showed a smaller tidal volume using a peak external negative pressure of 30 cmH₂O than at 20 cmH₂O. In only nine subjects could a peak external negative pressure of 40 cmH₂O be obtained and tolerated by the subject, but three of these had a lower tidal volume using this pressure than at 30 cmH₂O. Kelleher et al (1952) and Collier and Affeldt (1954) also found that, with the Monaghan cuirass, increasing the peak external negative pressure above 20 cmH₂O produced little additional ventilation and in some cases reduced the tidal volume obtained. This problem was not seen with the Huxley cuirass. This would suggest that the effect of peak external pressure on tidal volume should be evaluated in each patient prior to selecting the pressure to be used for nocturnal cuirass-assisted ventilation. It is likely that any reduction in tidal volume seen with an increase in peak external negative pressure with the patient awake will also occur during sleep.

Collier and Affeldt (1954) found that in subjects with poliomyelitis external positive pressure decreased end-expiratory volume but did not increase tidal volume. In contrast, Binet et al (1961) reported a marked increase in one patient with the use of external positive expiratory pressure of 10cmH₂O. The effect of external

positive expiratory pressure on tidal volume in the present study was small, and the tendency to increase tidal volume at lower peak external negative pressures could reflect a greater contribution of spontaneous respiratory muscle activity to tidal volume with smaller peak external negative pressures.

Ten of the twelve subjects in the present study showed a decrease in end-expiratory volume when external positive pressure was applied during expiration. In all twelve subjects in whom the effect of cuirass-assisted ventilation on end-expiratory volume was studied, cuirass-assisted ventilation increased PaO_2 , irrespective of the direction of change in end-expiratory volume. Nevertheless, in patients with spontaneous end-expiratory volumes less than predicted, an increase in end-expiratory volume with cuirass-assisted ventilation is likely to be advantageous in preventing collapse of underventilated regions of the lungs. The small increase in tidal volume with external positive expiratory pressure is offset by the abolition of the increase in end-expiratory volume seen when external negative pressure is used alone.

One subject with neuromuscular disease (RB) showed a much larger decrease in end-expiratory volume with external positive pressure than the other subjects, and he showed the largest increase in tidal volume. His spontaneous functional - residual capacity was high,

suggesting that weak expiratory musculature might explain the increase in tidal volume with external positive expiratory pressure, although his maximum expiratory mouth pressures were not particularly low compared to the other subjects. When positive pressure is created inside the cuirass shell, the shell is lifted away from the patient with a consequent reduction in the efficiency of the seal. This not only restricts the positive pressure which can be created, but also creates draughts around the patient. For this reason, all the patients found external positive pressure uncomfortable, and none elected to continue using it at night. This intolerance, the small increase in tidal volume and the decrease in end-expiratory volume argue against the use of external positive expiratory pressure during cuirass-assisted ventilation.

As might be expected, tidal volumes fell as respiratory rate increased. At faster respiratory rates the spirometer may have underestimated the tidal volume which would have been achieved without a mouthpiece. Increasing respiratory rate produced a corresponding change in minute volume, except in two subjects. These two subjects (IJ, WB) had the lowest ratios of FEV₁ to FVC of all the subjects, and airway resistance may have limited the change in minute volume. The ratio of dead space to tidal volume may change with respiratory rate, but arterial blood gas analysis was not undertaken to study

the relationship between alveolar ventilation and respiratory rate.

In thirteen subjects, cuirass-assisted ventilation lowered PaCO_2 . One subject (DS) had a higher PaCO_2 during cuirass-assisted ventilation compared to spontaneous ventilation, and the studies of tidal volume also showed that cuirass-assisted ventilation was ineffective in increasing ventilation in this subject. The increase in PaCO_2 seen in one subject (MH) despite an increase in ventilation could be explained either by a mouthpiece effect increasing spontaneous ventilation when measured by the spirometer but not acting when arterial blood was sampled, or by an increase in the dead-space to tidal volume ratio on changing from spontaneous to cuirass-assisted ventilation. An increase in PaCO_2 during cuirass-assisted ventilation was also seen in one subject (SS), who was not included in the study of the effect of peak external negative pressure on tidal volume. Cuirass-assisted ventilation produced an increase in PaO_2 in thirteen subjects. This may reflect the increased ventilation, combined with improved ventilation-perfusion matching as a result of the larger tidal volumes during cuirass-assisted ventilation. O'Leary et al (1979) demonstrated a fall in PaCO_2 and an increase in PaO_2 with cuirass-assisted ventilation in all six of their patients with neuromuscular disease, although in one of the patients the PaCO_2 during

spontaneous ventilation may have been elevated on account of the use of supplemental oxygen. Goldstein et al (1986) reported an increase in oxygen saturation and a decrease in end-tidal carbon dioxide concentration in six subjects with neuromuscular disease after thirty minutes of cuirass-assisted ventilation. Similar changes were seen in the patient reported by Fulkerson et al (1983).

Cuirass-assisted ventilation corrected paradoxical chest wall motion in five out of six subjects. The induction of paradoxical chest wall motion in two subjects can be explained by the predominantly abdominal pattern of ventilation produced by a cuirass shell which covers the upper abdomen, or by direct pressure from the upper rim of the cuirass shell which rests on the upper ribcage, thus limiting ribcage motion in the antero-posterior direction. Scales et al (1953) showed radiographically that Monaghan cuirasses could produce paradoxical ribcage or abdominal motion, although this was not seen with the Huxley cuirass. In an attempt to assess the limitation of chest wall motion produced by the cuirass, this was compared with a moulded jacket respirator which covered a similar area of the chest wall. In most of the subjects studied, the moulded jacket respirator produced tidal volumes which were only slightly greater than those obtained during cuirass-assisted ventilation at the same peak external negative pressure. In two subjects (RB, TC) the tidal

volumes were much greater with the moulded jacket compared to cuirass, suggesting that the cuirass was restricting chest wall expansion.

Both the Emerson and Tunncliffe jackets produced larger tidal volumes than the cuirass in RB and TC, but also in one other subject (HC), suggesting that she benefited from application of negative pressure to a larger area of the chest wall. Leakage of air around the edge and through the seams of the jacket was such that a peak external pressure of 30 cmH₂O could be obtained in only 70% of the subjects with the moulded jacket respirator and in only 50% with the Emerson respirator. Since the jacket style of external negative pressure ventilator is more cumbersome for the patient to use, it should probably only be used when the cuirass does not produce effective ventilation.

CHAPTER 5

CUIRASS-ASSISTED VENTILATION AND THE RESPIRATORY MUSCLES

5:1 INTRODUCTION

In patients with skeletal deformities of the chest wall, the mechanical work which the respiratory muscles must perform during tidal breathing is increased, and respiratory muscle fatigue may develop. Fatigue of abnormal muscle may occur even when the mechanical work required of them is normal. The contribution of respiratory muscle fatigue to hypoventilation in patients with chest wall disease is not known, but assisted ventilation at night leads to improvement in daytime respiratory muscle function (Marino and Braun 1982). Resting the respiratory muscles at night may account for this, but nocturnal assisted ventilation also improves daytime and sleeping arterial oxygen and carbon dioxide levels. Since hypoxia and hypercapnia impair respiratory muscle performance (Koulouris et al 1984, Jardim et al 1981, Juan et al 1984), improved arterial blood gases may partly explain the increase in respiratory muscle strength seen with nocturnal assisted ventilation. A striking improvement in patients well-being has been noted with the use of assisted ventilation at night, and better motivation might account for some of the changes in voluntary tests of muscle strength.

Celli et al (1987) have demonstrated that periods of

external negative pressure ventilation using a jacket respirator during the daytime lead to an improvement in respiratory muscle strength in patients with bilateral idiopathic diaphragmatic paralysis. The present study aimed to determine whether cuirass-assisted ventilation is able to rest the respiratory muscles in patients with chest wall disease, and whether a period of rest independent of the effects of sleep results in an increase in respiratory muscle performance.

5:2 METHODS

The electromyographic activity of the diaphragm was recorded using surface electrodes, placed in the sixth and seventh intercostal spaces in the mid-clavicular line on the right, with a ground attached to the right wrist. The electromyographic signal recorded from these electrodes was amplified, rectified and integrated using a leaky analogue integrator with a time constant of 0.2 seconds (Lectromed Ltd). The resultant signal was recorded on a chart recorder and the maximum change in the signal during five breaths, ignoring electrocardiograph artefact, was averaged to obtain an index of the electrical activity of the diaphragm. Recordings were made with the subject breathing spontaneously in the supine position, and in the same position during cuirass-assisted ventilation after a period of fifteen minutes.

Oxygen consumption was measured by the system

described in Chapter 3. The subjects rested in the supine position for fifteen minutes prior to the recordings made during spontaneous ventilation. Repeat measurements were made fifteen to thirty minutes after commencing cuirass-assisted ventilation. The cuirass pump was set at a rate of fifteen breaths per minute with an inspiratory:expiratory ratio of 1:1. The peak external negative pressure was adjusted to 30 cmH₂O.

On a separate day, maximum inspiratory mouth pressure was recorded before and after one hour of cuirass-assisted ventilation with the cuirass pump set to the most comfortable setting for the patient.

5:3 RESULTS

The peak integrated EMG signals recorded during cuirass-assisted ventilation expressed as a percentage of the signal recorded during spontaneous ventilation are given in Table 18. A reduction in inspiratory diaphragmatic activity with cuirass-assisted ventilation was seen in all cases. The oxygen consumption values are given in Table 19. There was no significant difference between the values obtained during spontaneous and cuirass-assisted ventilation.

The maximum inspiratory pressures before and after one hour of cuirass-assisted ventilation are shown in Table 20. There was no significant difference before and after assisted ventilation.

TABLE 18

Peak tidal integrated electromyographic activity during cuirass-assisted ventilation recorded with surface electrodes, expressed as a percentage of tidal electromyographic activity during spontaneous respiration.

Peak tidal EMG (% spontaneous ventilation)	
MS	76
SS	66
SG	28
WW	57
RB	14
TC	50
MH	75
IJ	48
WB	44
DH	7

Mean	46
Median	49

TABLE 19

Oxygen consumption measured during spontaneous ventilation (SV) and cuirass-assisted ventilation (CAV).

Subject	Oxygen consumption (mls/min)		
	SV	CAV	Change
SS	109	112	+3
HH	224	236	+12
BC	371	446	+75
WW	259	119	-140
BS	264	268	-4
AV	204	130	-74
MS	181	209	+28
JG	296	270	-26
JL	218	200	-18
WB	254	247	-7
MH	204	199	-5
DH	142	141	-1
IJ	159	211	+52
HC	211	196	-15
KN	280	336	+56
DS	269	259	-10

Mean	228	224	-5
Median	221	210	-4.5

TABLE 20

Maximum inspiratory pressure recorded at the mouth before and after one hour of cuirass-assisted ventilation.

Subject	Maximum Inspiratory Pressure (cmH ₂ O)		
	Before	After	Change
MS	41	48	+7
SS	12	15	+3
SG	32	47	+15
WW	62	55	-7
BS	80	87	+7
RB	19	24	+5
TC	22	28	+6
MH	38	53	+15
DH	34	39	+5
WB	27	29	+2
NW	11	13	+2
HC	60	52	-8
IJ	24	32	+8

Mean	35	40	+4
Median	32	39	+5

5:4 DISCUSSION

Although they may also record activity in the intercostal and abdominal muscles, skin surface electrodes were used to record diaphragm electromyograms in this study on account of their acceptability for the patient. All subjects were well accustomed to cuirass-assisted ventilation and able to relax during the study. A reduction in the amplitude of the inspiratory activity was seen in all subjects during cuirass-assisted ventilation, in contrast to the normal subjects studied by Rodenstein et al (1986) in whom respiratory muscle activity was reduced during external negative pressure ventilation only when the subjects were asked to relax.

In addition to voluntary influences, the decrease in inspiratory muscle activity could be caused by a reduction in respiratory drive secondary to changes in arterial blood gases with cuirass-assisted ventilation. The changes in arterial blood gases documented in chapter 4 were small, and unlikely to explain the magnitude of the fall in inspiratory muscle activity. Inspiratory muscle activity usually declined within a few breaths of commencing cuirass-assisted ventilation before sufficient time had elapsed for any change in arterial blood gases.

Stimulation of pulmonary and chest wall receptors by increasing the tidal volume above spontaneous levels may have induced reflex inhibition of inspiratory muscle activity during cuirass-assisted ventilation. In

sleeping tracheotomised man, lung inflation has been shown to inhibit subsequent inspiration (Winning et al 1988), but the contribution of this and other reflexes to the reduction in inspiratory muscle activity during cuirass-assisted ventilation remains unknown.

Rochester et al (1977) have recorded the electromyographic activity of the diaphragm during external negative pressure ventilation. They studied the effect of a tank respirator and a cuirass in both normal subjects and patients. Only one patient with scoliosis was studied with a cuirass, and they showed a fall in diaphragmatic activity. Gandevia and McKenzie (1987) have shown that the amplitude of the diaphragm EMG recorded during phrenic nerve stimulation in normal subjects is greater in the supine posture compared to the upright posture. Rochester studied his subjects in the seated position during spontaneous ventilation, and in the supine position during assisted ventilation, therefore introducing the possibility that the decrease in EMG activity seen was an underestimate of the true change in diaphragmatic activity. Goldstein et al (1986) comment on a marked reduction in diaphragmatic electromyographic activity in six patients with neuromuscular disease during negative pressure ventilation.

The reduction in inspiratory muscle activity seen during cuirass-assisted ventilation would be expected to

be accompanied by a reduction in the oxygen consumed by the respiratory muscles. In normal subjects, the oxygen consumption of the respiratory muscles during tidal breathing has been estimated as 0.35mls per litre of ventilation (Campbell 1959). The oxygen cost of breathing is likely to be higher in subjects with chest wall disease compared to normal subjects. Bergofsky et al (1959) have measured total oxygen consumption in scoliotic subjects, but the proportion of total oxygen consumption used by the respiratory muscles in patients with chest wall disease has not been determined.

Rochester et al (1977) noted that insertion of a mouthpiece induced a return of respiratory muscle activity during assisted ventilation. In the present study, electromyographic recordings were made without a mouthpiece, whereas oxygen consumption was measured with a mouthpiece. Had inspiratory muscle activity been recorded during the measurement of oxygen consumption, it is possible that no difference would have been seen between spontaneous and cuirass-assisted ventilation on account of the presence of a mouthpiece. This would also explain the failure to document any change in oxygen consumption. The short period over which oxygen consumption was measured and the fact that the subjects were not fasting may also have contributed to the failure to demonstrate any change in oxygen consumption.

Several factors can explain the failure to demonstrate an increase in maximum inspiratory pressure after one hour of cuirass-assisted ventilation. The coefficient of reproducibility (Bland and Altman 1986) of maximum inspiratory pressure measurements by this method was 15 cmH₂O, and therefore this method of assessing respiratory muscle strength cannot reliably detect small changes. All of the subjects studied were in a stable clinical state, were using nocturnal cuirass-assisted ventilation and in contrast to the patients studied by Celli et al (1987) may have had only minimal respiratory muscle fatigue at the time of the present study.

One hour of cuirass-assisted ventilation may not have been sufficient to relieve respiratory muscle fatigue. Edwards et al (1977) reported that the mechanical response of skeletal muscle to supramaximal stimulation at 20 Hz took several hours to recover after induction of fatigue. Aubier et al (1981) studied the mechanical response of the diaphragm to supramaximal phrenic nerve stimulation at 20 Hz before and 30 minutes after breathing through a resistance to produce fatigue. Thirty minutes after the induction of fatigue, the response of the diaphragm was still only 30% of the pre-fatigue values. Moxham et al (1980) found that after the development of fatigue in the sternomastoid muscle, the mechanical response to stimulation at low frequencies was still abnormal after 4 hours of rest. If the time

course for recovery of fatigue is similar when patients with chest wall disease receive assisted ventilation, a longer period of rest would have been necessary to relieve fatigue and improve respiratory muscle strength.

CHAPTER 6

THE EFFECT OF CUIRASS-ASSISTED VENTILATION ON CARDIAC OUTPUT

6:1 INTRODUCTION

In 1942, Eisenmenger reported the advantageous effects of the Biomotor cuirass on the circulation of patients with right heart failure (Eisenmenger 1942). The effects of tank respirators on the circulation were studied during poliomyelitis epidemics (Maloney and Whittenberger 1951), but it was not until the 1960's that cardiovascular function during cuirass-assisted ventilation was studied further. Getzen initially studied dogs using cinefluorography and found that during cuirass-assisted ventilation cardiac size increased. Carotid thermography in man suggested that cardiac output increased during cuirass-assisted ventilation (Getzen et al 1968).

On theoretical grounds, cuirass-assisted ventilation has been advocated as one of the few methods of assisted ventilation which should not impede venous return (West 1982). The aim of the present study was to determine the effects of cuirass-assisted ventilation on cardiac output in man.

6:2 METHODS

Right heart catheterisation was performed in eight subjects and cardiac output measurements were made as described in Chapter 3. Cardiac output was recorded after the subjects has been resting in the supine

position breathing spontaneously for fifteen minutes. Cuirass-assisted ventilation was then commenced at a respiratory rate of fifteen breaths per minute with an inspiratory:expiratory ratio of 1:1 and a peak external negative pressure of 30 cmH₂O. After a further fifteen minutes, cardiac output measurements were repeated.

6:3 RESULTS

The mean of the three cardiac output measurements made during spontaneous and cuirass-assisted ventilation for each subject are given in Table 21. The difference between the lowest and highest cardiac output measurement in each patient ranged from 0.1 to 1.1 l/min, with a median for the sixteen sets of readings of 0.6 l/min. There was no significant difference in cardiac output between spontaneous and cuirass-assisted ventilation.

TABLE 21

The mean of three thermodilution cardiac output measurements for each subject during spontaneous ventilation (SV) and cuirass-assisted ventilation (CAV).

Subject	Cardiac Output (l/min)	
	SV	CAV
AP	4.4	3.6
BC	4.0	4.1
WW	4.3	4.3
BS	3.5	2.6
JG	4.9	5.3
WB	7.5	7.5
DH	3.7	3.3
KN	4.1	3.6

Median	4.2	3.85

6:4 DISCUSSION

There are several theoretical reasons why cuirass-assisted ventilation might increase or decrease cardiac output. During quiet respiration, intrathoracic pressure falls during inspiration and rises again during expiration. Cuirass-assisted ventilation, like deep breathing, exaggerates the normal pattern of intrathoracic pressure swing. During inspiration, lowering of intrathoracic pressure increases venous return to the right heart. Descent of the diaphragm during inspiration creates a positive intra-abdominal pressure, and it has been suggested (Robotham et al 1983) that this may impede venous return from the lower limbs. Inside a thoraco-abdominal cuirass shell, during inspiration the upper abdomen and chest are submitted to negative pressure. The venous return from the head and upper limbs should therefore increase, whereas there will be little pressure differential between the upper abdomen and chest to affect venous return from the inferior vena cava. When there is a negative pressure inside the cuirass shell, the lower rim descends onto the lower abdomen, and this may impair venous return from the legs.

The right ventricle is also affected by changes in afterload occurring as a result of lung inflation. Whittenberger et al (1960) showed in dogs that pulmonary vascular resistance was greater at high lung volumes, although this effect was most marked when pulmonary blood

flow was low. In these experiments, the chest was open. During inspiration with a closed chest, the pressure around the pulmonary artery (intrathoracic pressure) falls, and the transmural pulmonary artery pressure will therefore rise. The increase in pulmonary vascular resistance and the increase in transmural pulmonary artery pressure both increase right ventricular afterload. The external negative pressure during cuirass-assisted ventilation is transmitted to the intrathoracic cavity to produce inspiration, which should affect right ventricular afterload in a similar manner to normal inspiration. Oxygenation was not measured in sufficient numbers of subjects in the present study for conclusions to be drawn about the effect of oxygen on pulmonary vascular resistance. The studies reported in Chapter 4 show a small increase in PaO_2 , which would probably be associated with a small fall in pulmonary vascular resistance.

With an increase in both preload and afterload, an increase in right ventricular size would be expected during inspiration. Goldblatt et al (1963) studied ventricular size in man using myocardial clips inserted at the time of open heart surgery. Cinefluorography demonstrated an increase in right ventricular size during quiet inspiration, exaggerated by deep inspiration. Brinker et al (1980) used echocardiography in man to measure right ventricular size during the Muller

manoeuvre and also found that right ventricular size increased. Goldblatt et al (1963) also found a small decrease in left ventricular size during inspiration. Stooile et al (1974) used the same technique in dogs during partial pulmonary artery occlusion, and found that as right ventricular size increased, so left ventricular size decreased. Bemis et al (1974) studied isolated dog hearts using cinefluorography and found that as right ventricular end-diastolic pressure was increased, the interventricular septum bulged into the left ventricle. Echocardiography has been used to study ventricular size in man by Weyman et al (1976) during right ventricular volume overload, and Brinker et al (1980) during the Muller manoeuvre. Both demonstrated leftward septal displacement as the right ventricular volume increased. If right ventricular changes during normal inspiration are exaggerated during cuirass-assisted ventilation as would be expected, then left ventricular output should decrease in a similar manner.

A decrease in intrathoracic pressure will increase the transmural aortic pressure, and therefore even if intra-arterial pressure remains the same, left ventricular afterload will increase. Scharf et al (1981) used radionuclide ventriculography and Buda et al (1979) used myocardial markers to study the effect of the Muller manoeuvre on left ventricular ejection fraction in man. In Scharf's subjects with a normal myocardium,

there was no change in ejection fraction, but in subjects with ischaemic heart disease ejection fraction decreased. Buda's patients had all had either a heart transplant or coronary artery surgery, and ejection fraction decreased during the Muller manoeuvre. Karam et al (1984) used radionuclide ventriculography in normal subjects to measure ejection fraction. When the mean intrathoracic pressure was decreased by making the subjects breathe through an inspiratory resistance, ejection fraction fell. Cuirass-assisted ventilation should produce a more marked decrease in intra-thoracic pressure than occurs during spontaneous inspiration, and thus left ventricular afterload should be increased.

Lung inflation may also affect the circulation by reflex or humoral mechanisms. De Burgh Daly and Robinson (1968) showed in dogs that lung inflation lowered systemic vascular resistance, this effect being abolished by vagotomy or sympathectomy. Cross perfusion experiments in the dog have shown that blood from one animal in which the lungs are inflated causes a decrease in cardiac output in an animal with uninflated lungs (Grindlinger et al 1979). The mechanism of this effect may be release of prostaglandins from the lungs during inflation (Berry et al 1971).

The results of the present study show no difference in cardiac output between spontaneous and cuirass-assisted ventilation. The small number of subjects studied and

scatter of the individual measurements of cardiac output was such that a small difference in cardiac output between spontaneous and cuirass-assisted ventilation would not have been detected.

CHAPTER 7

CUIRASS-ASSISTED VENTILATION AND SUPPLEMENTARY OXYGEN THERAPY

7:1 INTRODUCTION

In normal subjects, hypoxia and hypercapnia both stimulate an increase in ventilation. Patients with chronic respiratory failure become tolerant to hypercapnia, and the chemical control of breathing is dependent on the response to hypoxia. Correction of hypoxia in these patients by increasing the inspired oxygen concentration will decrease respiratory drive and lead to hypoventilation.

Administration of supplementary oxygen to patients with respiratory failure secondary to scoliosis or neuromuscular disease has been shown to produce a rise in arterial carbon dioxide tensions or a fall in minute ventilation (Bergofsky 1959, Bruderman and Stein 1961, Fulkerson et al 1983, Skatrud et al 1980).

The study described in this chapter examines the ability of cuirass-assisted ventilation to maintain ventilation, despite a fall in respiratory drive, when the inspired oxygen concentration is increased. If no rise in arterial carbon dioxide level occurred, then the combination of oxygen and cuirass-assisted ventilation would be of use in the treatment of hypoxaemia in patients with chest wall disease.

7:2 METHODS

Ten subjects with chest wall disease were studied.

All were in a stable clinical condition and were receiving cuirass-assisted ventilation every night. A Hewlett-Packard ear oximeter and Radiometer transcutaneous carbon dioxide electrode were attached to the patient. After lying supine for thirty minutes breathing air, an oxygen face mask was applied and the flow of oxygen adjusted to deliver an inspired concentration of 24%. If after ten minutes there had not been a rise in $P_{tc}CO_2$ of more than 1 kPa, the inspired oxygen concentration was increased to deliver 30% oxygen. If after a further ten minutes there had not been a rise in $P_{tc}CO_2$ of more than 1 kPa above the starting value, the inspired oxygen concentration was increased to 35% and maintained at this level for forty minutes.

The oxygen mask was then removed and the subject placed in their cuirass. Assisted ventilation was commenced at a comfortable rate for the subject with the maximum peak external negative pressure which could be tolerated. A period of thirty minutes was allowed for gas exchange to stabilise on cuirass-assisted ventilation. The absence of a change in $P_{tc}CO_2$ of greater than 0.1 kPa during the last ten minutes of this period was taken as confirmation of stability. The oxygen mask was then applied and the oxygen flow adjusted to increase the inspired concentration to 35%. Cuirass-assisted ventilation at this inspired oxygen concentration was continued for one hour, and at the end

of this period the ear oxygen saturation and transcutaneous carbon dioxide tension were recorded.

7:3 RESULTS

No subject showed a rise in $P_{tc}CO_2$ of more than 1 kPa with inspired oxygen concentrations of 24 or 28%, and therefore only the data relating to an inspired oxygen concentration of 35% is presented. The ear oximetry and $P_{tc}CO_2$ values during spontaneous ventilation are presented in Table 22. Eight subjects had ear oxygen saturations of less than 95% breathing air. In two subjects this desaturation persisted when breathing 35% oxygen.

All subjects showed an increase in $P_{tc}CO_2$ when the inspired oxygen concentration was increased to 35% during spontaneous ventilation. The increases in $P_{tc}CO_2$ in the two subjects with oxygen saturations of greater than 95% when breathing air were 0.1 and 0.2 kPa. In the eight subjects with oxygen saturations of less than 95% when breathing air, $P_{tc}CO_2$ increased by a mean of 0.7 kPa (range 0.2 to 1.2, median 0.6 kPa).

The three subjects with a $P_{tc}CO_2$ when breathing air of less than 7 kPa showed increases of 0.1, 0.2, and 0.5 kPa when the inspired oxygen concentration was increased to 35%. The seven subjects with a $P_{tc}CO_2$ of greater than 7 kPa when breathing air showed a mean increase in $P_{tc}CO_2$ of 0.7 kPa (range 0.2 to 1.2, median 0.6 kPa) when the inspired oxygen concentration was increased to 35%.

The changes in oxygen saturation and transcutaneous carbon dioxide tension when the inspired oxygen concentration was increased during cuirass-assisted ventilation are shown in Table 23. Ear oxygen saturation increased in all subjects, but remained less than 95% in one subject. There was no significant increase in $P_{tc}CO_2$ with the administration of supplementary oxygen for the ten patients considered as a group. Two subjects showed a small increase in $P_{tc}CO_2$ with the addition of supplementary oxygen during cuirass-assisted ventilation, but in only one of them was this increase as large as that seen during spontaneous ventilation.

TABLE 22

The change in ear oxygen saturation (SaO₂) and transcutaneous carbon dioxide tension (PtcCO₂) on increasing the inspired oxygen concentration to 35% during spontaneous ventilation.

Subject	SaO ₂		PtcCO ₂		Change in PtcCO ₂ with Oxygen
	Air	35% oxygen	Air	35% oxygen	
MS	89	94	6.7	7.2	+0.5
WW	91	98	7.2	8.1	+0.9
BS	90	98	8.0	8.7	+0.7
RB	97	100	6.5	6.7	+0.2
TC	92	100	7.7	7.9	+0.2
MH	99	100	6.0	6.1	+0.1
WB	93	99	7.1	7.7	+0.6
DH	79	90	7.4	8.0	+0.6
IJ	89	99	8.4	9.6	+1.2
NW	94	99	8.4	9.2	+0.6
Median	91	99	7.3	7.95	+0.6

TABLE 23

The change in ear oxygen saturation (SaO₂) and transcutaneous carbon dioxide tension (PtcCO₂) on increasing the inspired oxygen concentration to 35% during cuirass-assisted ventilation.

Subject	SaO ₂		PtcCO ₂		Change in PtcCO ₂ with Oxygen
	Air	35% oxygen	Air	35% oxygen	
MS	90	96	6.1	6.1	0
WW	94	98	7.1	6.5	-0.6
BS	93	98	7.4	7.4	0
RB	98	100	4.9	4.9	0
TC	93	100	7.7	7.9	+0.2
MH	93	100	6.0	6.0	0
WB	94	99	7.1	6.9	-0.2
DH	81	90	7.1	7.2	+0.1
IJ	89	99	8.0	7.8	-0.2
NW	94	99	8.8	8.6	-0.2

Median	93	99	7.1	7.15	0

7:4 DISCUSSION

Patients who become chronically hypercapnic lose their ventilatory response to carbon dioxide and become dependent on hypoxia as the main chemical stimulus to breathing. Increasing the inspired oxygen concentration in such subjects can precipitate a further increase in carbon dioxide levels. In the present study, all ten subjects showed a rise in $P_{tc}CO_2$ when the inspired oxygen concentration was increased to 35% during spontaneous ventilation. The magnitude of the rise in carbon dioxide tension is similar to that documented by Bruderman and Stein (1961) in two patients with scoliosis and by Skatrud et al (1980) in a patient with diaphragmatic paralysis.

The rise in $P_{tc}CO_2$ on increasing the inspired oxygen concentration was inversely related to the initial SAO_2 . This suggests that the more hypoxic subjects were more dependent on hypoxic ventilatory drive. These subjects are most in need of oxygen, but it produced the most deleterious effects.

During cuirass-assisted ventilation, increasing the inspired oxygen concentration produced a rise in oxygen saturation in every subject. This was achieved without a rise in carbon dioxide levels. This suggests that in awake patients with chest wall disease, supplementary oxygen can be administered safely during cuirass-assisted ventilation.

The effect of oxygen administration during sleep was not studied. Simonds and Branthwaite (1985) have described the use of supplementary oxygen during external negative pressure ventilation at night. This was used to reduce spontaneous respiratory muscle activity. Coordination between the external negative pressure cycle and spontaneous respiratory muscle activity improved, but at the expense of a rise in carbon dioxide. This may have reflected inability of the respirators used to maintain adequate ventilation without the full contribution of the respiratory muscles, or upper airway obstruction developing in association with reduced respiratory muscle activity.

The results of the present study cannot be extrapolated to suggest that supplementary oxygen is safe to use at night with cuirass-assisted ventilation, particularly in view of Simonds and Branthwaite's findings.

CHAPTER 8

CUIRASS-ASSISTED VENTILATION AND GAS EXCHANGE DURING SLEEP

8:1 INTRODUCTION

In chapter 2, the abnormalities of gas exchange during sleep in patients with chest wall disease were described. Several reports have documented the ability of cuirass-assisted ventilation to improve gas exchange during sleep in small numbers of subjects (Skatrud 1980 et al, Powner and Hoffman 1978, Bye et al 1986, Goldstein et al 1986 and 1987) whilst Simonds and Branthwaite (1985) have studied the relative efficiency of the cuirass, Tunncliffe jacket and pneumosuit jacket.

In chapter 4 daytime ventilation and arterial blood gases were shown to improve with cuirass-assisted ventilation using individually moulded cuirass shells, but assisted ventilation is often only used at night in these patients. Upper airway obstruction and reduced, irregular or absent respiratory muscle activity during sleep might decrease the ability of the cuirass to maintain adequate ventilation. The purpose of the study described in this chapter was to determine the ability of cuirass-assisted ventilation to correct abnormal gas exchange during sleep in patients with chest wall disease.

8:2 METHODS

The effect of cuirass-assisted ventilation on gas exchange during sleep was investigated in 13 subjects with chest wall disease. The monitoring apparatus used during sleep is described in Chapter 3, and consisted of an ear oximeter, a transcutaneous carbon dioxide electrode, a nasal thermistor and surface electromyographic electrodes. All patients were accustomed to this apparatus and to cuirass-assisted ventilation.

Subjects were studied on two nights in the same week, where possible on consecutive nights. Gas exchange during sleep was studied during spontaneous ventilation on one night and with cuirass-assisted ventilation on the other night, in random order. The period of sleep was ascertained on behavioural criteria only, electroencephalographic monitoring not being available. Tracings were analysed manually, and periods of waking during the night were excluded from the analysis. From the tracing of ear oxygen saturation, the lowest oxygen saturation during each minute of sleep was determined. The total time during the night when this saturation was less than 90, 80, 70, 60 and 50% saturation was expressed as a percentage of total sleep time. The minimum saturation reached was also recorded. From the $P_{tc}CO_2$ trace, the maximum level recorded over each five minute period of sleep was determined. From this data the mean

PtcCO₂ during sleep was calculated. The maximum PtcCO₂ recorded overnight was noted.

Coordination between spontaneous respiratory muscle activity and the external negative pressure cycle was assessed by recording diaphragmatic electromyograms using surface electrodes and the pressure inside the cuirass shell using a Si Plan pressure transducer. A Lectromed nasal thermistor was used to detect periods of upper airway obstruction.

8:3 RESULTS

The ear oxygen saturations and carbon dioxide tensions for each patient are given in Tables 24 and 25 respectively. The grouped data for oxygen saturation are displayed in Table 26. There was no significant difference in the sleep time with spontaneous and cuirass-assisted ventilation. The time spent at saturations below 50% was significantly less with cuirass-assisted ventilation compared to spontaneous ventilation (Wilcoxon signed rank sum 0, $p < .01$), but there was no significant difference in the other parameters of sleep oxygenation. The mean and maximum transcutaneous carbon dioxide tensions were both significantly lower during cuirass-assisted ventilation compared to spontaneous ventilation (Wilcoxon signed rank sum 0, $p < 0.01$).

Of the five subjects in whom diaphragmatic activity and airflow were recorded, three showed incoordination between spontaneous respiratory muscle activity and the external negative pressure cycle, but only one patient had periods of absent airflow detected by the nasal thermistor.

TABLE 24

Ear oxygen saturation (SaO₂) during sleep, spontaneous ventilation without assisted ventilation (SV, upper line), and with cuirass-assisted ventilation (CAV, lower line).

Subject	Sleep time	Percentage of sleep time at SaO ₂ :					Minimum SaO ₂
		<90%	<80%	<70%	<60%	<50%	
MS:	SV 355	92	18	5	2	<1	45
	CAV 220	60	10	7	2	0	46
SS:	SV 430	100	97	29	3	1	28
	CAV 480	100	95	47	10	0	51
AP:	SV 280	100	91	34	4	1	29
	CAV 330	94	2	11	6	1	42
WW:	SV 180	100	91	19	<1	<1	59
	CAV 200	51	2	0	0	0	77
AV:	SV 425	15	4	<1	0	0	66
	CAV 435	21	7	1	0	0	63
JG:	SV 235	97	3	1	0	0	61
	CAV 215	100	86	28	6	0	49
GP:	SV 305	93	22	5	0	0	66
	CAV 430	13	2	0	0	0	64
DH:	SV 355	99	90	81	74	12	21
	CAV 377	98	84	8	3	3	34
IJ:	SV 375	100	41	4	<1	0	53
	CAV 380	100	0	0	0	0	81
DS:	SV 425	100	100	99	99	94	5
	CAV 160	100	100	100	80	17	38
NW:	SV 445	100	80	4	1	0	54
	CAV 390	97	0	0	0	0	82
KN:	SV 145	54	<1	0	0	0	79
	CAV 380	72	4	1	0	0	61
HC:	SV 230	100	100	23	1	<1	45
	CAV 464	100	73	3	0	0	59

TABLE 25

Transcutaneous carbon dioxide tensions (PtcCO₂) at night during spontaneous ventilation (SV) and cuirass-assisted ventilation (CAV).

Subject	SV		CAV	
	Mean PtcCO ₂	Maximum PtcCO ₂	Mean PtcCO ₂	Maximum PtcCO ₂
MS	8.7	10.4	5.6	6.1
SS	10.5	11.7	9.3	10.2
AP	9.1	10.5	7.0	8.1
WW	8.2	8.9	5.4	5.7
AV	7.9	8.8	7.8	8.2
JG	8.2	8.8	4.0	4.3
GP	9.4	10.6	5.3	6.5
DH	11.6	12.9	9.3	10.2
IJ	7.0	8.1	6.4	6.8
DS	8.5	8.9	7.2	8.1
NW	10.8	15.7	6.5	7.1
KN	8.6	9.3	4.0	5.3
HC	6.5	7.6	5.4	5.9
Mean	8.8	10.2	6.4	7.1
Median	8.6	9.3	6.4	6.8

TABLE 26

Sleep ear oxygen saturation (SaO₂) for 13 patients with chest wall disease studied during spontaneous ventilation (SV) and cuirass-assisted ventilation (CAV).

	Sleep time (min)	% of sleep time at SaO ₂ :					Minimum SaO ₂ (%)
		<90%	<80%	<70%	<60%	<50%	
SV:							
Mean	321	88	57	23	14	8	47
Median	355	100	80	5	1	0	53
CAV:							
Mean	343	77	36	16	8	2	57
Median	380	97	7	3	0	0	59

8:4 DISCUSSION

If cuirass-assisted ventilation kept the patients awake, gas exchange would be better during that night compared to the night of spontaneous ventilation when they were able to sleep. In this study, sleep was defined on behavioural criteria only. Although this is likely to lead to an overestimate of true sleep time as defined on electroencephalographic criteria, all patients reported that they had slept for most of the time of the study for both the spontaneous and cuirass-assisted ventilation nights.

Since sleep was not staged, spontaneous ventilation and cuirass-assisted ventilation during different stages of sleep cannot be compared. A difference in the stage pattern of sleep between the two nights may be involved in the difference in gas exchange found. One subject (JG) described a marked subjective improvement in the quality of her sleep with cuirass-assisted ventilation. She may have resembled Bye's patients with sleep disordered breathing (Bye et al 1986) who showed a "rebound" increase in the proportion of time spent in REM sleep on commencing assisted ventilation. Since REM sleep is associated with the most marked sleep breathing abnormalities in neuromuscular disease (Skatrud et al 1980), then cuirass-assisted ventilation may cause a deterioration in gas exchange, as was seen with JG. Electroencephalographic monitoring would be necessary to

confirm this hypothesis.

All subjects had a lower mean PtcCO₂ with cuirass-assisted ventilation compared to spontaneous ventilation during sleep, suggesting an improvement in ventilation. As described in Chapter 3, the transcutaneous electrode differed by approximately 1 kPa from arterial PaCO₂ in the awake state, but the comparison did not include any subjects with an arterial PaCO₂ of greater than 10 kPa. The elevated PtcCO₂ values above this figure seen during sleep must therefore be interpreted with caution. This is particularly the case with DS, in whom the maximum PtcCO₂ recorded was 15.7 kPa, even though the severe hypoxia and reduced nasal airflow seen in this subject would be compatible with profound hypoventilation.

Cuirass-assisted ventilation affected oxygenation during sleep in a number of different ways. Three subjects (AV,KN,GP) spent little time at saturations less than 70% during spontaneous ventilation, and there was little difference in the pattern of oxygen saturation when compared to cuirass-assisted ventilation.

Four subjects (WW,IJ,NW,HC) showed an increase in the amount of time spent at saturations greater than 80% when cuirass-assisted ventilation was compared to spontaneous ventilation, with little or no time spent at lower oxygen saturations during cuirass-assisted ventilation. Sleep oxygenation would appear to have been improved by

cuirass-assisted ventilation in these subjects.

Four subjects (MS,AP,DH,DS) showed an improvement in the overall pattern of oxygenation when cuirass-assisted ventilation was compared with spontaneous ventilation, but all continued to have periods of desaturation below 60%. One subject (DS) was severely hypoxic during the daytime, and a saturation above 70% was never achieved during cuirass-assisted ventilation, with 17% of sleep time spent at a saturation of less than 50%. At such saturations, the oximeter probe used tends to underestimate oxygen saturation. This effect is likely to explain the minimum saturation of 5% recorded in this subject. In the other three subjects in this group, the majority of sleep time was spent at saturation above 80%, the lower saturations occurring as short dips. The oxygen desaturation could reflect inability of the cuirass to maintain ventilation without coordinated inspiratory muscle activity, or the development of upper airway obstruction. Although airflow persisted during these hypoxic dips, partial upper airway obstruction may not have been detected by the nasal thermistor, which provides only a semi-quantitative index of airflow. The periods of desaturation were associated with irregularity of the frequency and amplitude of phasic diaphragmatic activity recorded with surface electrodes. The irregularity of inspiratory muscle activity suggests that the subjects were in REM sleep. Asynchrony between the

respiratory muscles and the pump has been noted by Simonds and Branthwaite (1985) to result in loss of efficiency of external negative pressure ventilation during sleep.

In addition to JG, SS also showed lower oxygen saturations during cuirass-assisted ventilation compared to spontaneous ventilation. During cuirass-assisted ventilation SS had periods when no airflow was detected, indicating upper airway obstruction. Simonds and Branthwaite (1985), Goldstein et al (1986) and Scharf et al (1978) have all described the development of upper airway obstruction during external negative pressure ventilation during sleep. Diaphragmatic activity was reduced during the episodes of upper airway obstruction seen in SS, probably associated with reduction in the activity of upper airway dilator muscles.

CHAPTER 9

THE PHYSIOLOGICAL EFFECTS OF ONE YEAR OF NOCTURNAL CUIRASS-ASSISTED VENTILATION

9:1 INTRODUCTION

Most of the previous studies of the use of nocturnal cuirass-assisted ventilation at home in patients with chest wall disease are difficult to interpret. The number of subjects has been small (Weirs et al 1977, Dull and Sadoul 1981, Dunkin 1983, Fulkerson et al 1984, Goldstein et al 1987, Powner and Hoffman 1978), and often the data has been incorporated with that of patients treated with other methods of assisted ventilation (Marino and Braun 1982). Physiological data is generally scanty, patients with widely differing diseases have been included in some analyses, and the follow up interval has sometimes been short. Loh et al (1979), however, studied six subjects with neuromuscular disease in some detail over several years. Sawicka et al (1988) also reported a series of subjects with chest wall disease using external negative pressure ventilation at home, principally from the point of view of their survival, but with blood gas and lung volume data where these were available.

The present study examined the effects of one year of nocturnal cuirass-assisted ventilation in twenty five subjects with chest wall disease, looking prospectively at various physiological parameters.

9:2 METHODS

The initial assessment of the subjects was made at the time of referral to the assisted ventilation unit, whenever possible prior to commencing assisted ventilation. One subject (MH) required immediate institution of assisted ventilation using a tank respirator, and measurement of lung volumes and maximum mouth pressures was delayed until after this initial period of resuscitation. Two subjects (RB, JL) were referred after intermittent positive pressure ventilation had been commenced at another hospital. In all surviving subjects, the repeat assessments were made twelve to fifteen months later, during a clinically stable period for each patient.

Daytime arterial blood gases, haemoglobin concentrations, lung volumes, maximum mouth pressures, maximum voluntary ventilation, pulmonary artery pressure, the ventilatory response to carbon dioxide and six minute walking tests were performed as described in Chapter 3.

Right heart failure was assessed using the scale described by Hoeppner et al (1984). Jugular venous distension was graded 0 to 3 corresponding to a jugular venous pressure waveform not visible in the neck, visible less than 5, 5 to 15, or more than 15 cm above the sternal angle with the subject positioned at 45 degrees to the horizontal; to this score was added the degree of peripheral oedema (0 for nil, 1 for ankle, 2 for above

ankle and 3 for above knee) to produce a right heart failure score. Electrocardiographic evidence of right heart strain was graded according to the system used by Hoepfner et al (1984), with a point for each of the following: frontal QRS axis greater than 110 degrees, R in AVR of greater than 5mm, R in V1 of greater than 7mm, R in V1 greater than S in V1, P in II greater than 2.5mm, ST depression in V1 to V3 or T wave inversion in V2 or V3. A diuretic index was calculated using one point for a 40mg dose of frusemide, 50mg dose of spironolactone or 5mg dose of amiloride. No subject was receiving any other diuretic.

9:3 RESULTS

9:3:1 Survival

Four deaths occurred during the year. One subject with motor neurone disease (JL) led an active life at home for eight months, with little in the way of respiratory symptoms, but got up at night to go to his bathroom where he collapsed and died. Another subject with a congenital myopathy (RR) was well until seven months after commencing home nocturnal cuirass-assisted ventilation, when he developed pneumonia. He was admitted to the Assisted Ventilation Unit, treated with antibiotics, physiotherapy and continuous cuirass-assisted ventilation but subsequently required intermittent positive pressure ventilation via an endotracheal tube. His condition deteriorated whilst he

was ventilated and he died.

Two other subjects died within one week of commencing nocturnal cuirass-assisted ventilation at home. One of these frequently fell asleep during the daytime whilst in hospital, on occasion becoming deeply cyanosed. The importance of using cuirass-assisted ventilation during periods of daytime sleep was stressed to him. When visited at home a few days after discharge he appeared well, but was subsequently found dead lying on his bed not attached to his cuirass. Another subject (DS) became breathless at night shortly after leaving the Assisted Ventilation Unit. She called a neighbour and her general practitioner but was not readmitted to hospital and died later that night. The results in the remainder of this chapter apply to the surviving twenty one subjects.

9:3:2 Daytime Blood Gases

Daytime arterial blood gases are displayed in Table 27. All were taken with the subject breathing air. After one year of nocturnal cuirass-assisted ventilation, PaCO_2 was lower by a median of 1.3 kPa (range -4.8 to +1.4 kPa), nineteen subjects (90%) having a lower PaCO_2 (Wilcoxon signed rank sum 17, $p < 0.01$). PaO_2 increased by a median of 1.4 kPa (range -1.7 to +5.4 kPa), fourteen subjects (67%) showing an increase in PaO_2 (Wilcoxon signed rank sum 26.5, $p < 0.01$).

TABLE 27

The change in daytime arterial blood gases after one year of nocturnal cuirass-assisted ventilation.

	PaCO ₂ (kPa)			PaO ₂ (kPa)		
	Before	After	Change	Before	After	Change
MS	7.3	7.2	-0.1	9.0	7.3	-1.7
HH	8.6	6.9	-1.7	8.1	10.0	+1.9
SS	7.9	7.2	-0.7	6.8	6.5	-0.3
AP	9.5	6.3	-3.2	5.3	8.1	+2.8
MB	7.9	6.8	-1.1	8.1	7.4	-0.7
SG	10.9	6.1	-4.8	5.5	10.9	+5.4
WW	8.5	5.2	-3.3	6.1	8.6	+2.5
AV	7.2	5.7	-1.5	8.4	11.6	+3.2
BS	7.6	4.7	-2.9	5.5	7.2	+2.2
YH	7.7	7.6	-0.1	6.4	8.0	+1.6
JG	7.9	6.7	-1.2	7.4	8.8	+1.4
GP	10.3	7.4	-2.9	7.9	9.0	+1.1
RB	7.1	5.7	-1.4	10.0	11.9	+1.9
TC	6.1	5.1	-1.0	8.5	9.9	+1.4
WB	7.5	6.2	-1.3	7.2	7.2	0
MH	8.6	5.8	-2.8	8.1	10.6	+2.5
DH	7.7	6.3	-1.4	7.3	6.5	-0.8
IJ	6.6	6.0	-0.6	7.8	8.5	+0.7
NW	6.3	7.2	+0.9	9.6	9.0	-0.6
KN	5.4	5.3	-0.1	11.2	10.6	-0.6
HC	5.9	7.3	+1.4	6.4	7.6	+1.2
Mean	7.7	6.3	-1.4	7.6	8.8	+1.2
Median	7.7	6.3	-1.3	7.8	8.6	+1.4

9:3:3 Lung Volumes

Lung volumes are displayed in Tables 28 and 29. After one year of nocturnal cuirass-assisted ventilation, FVC increased by a median of 0.17 litres (range -0.30 to +0.60 litres), fifteen of the subjects (71%) showing an increase (Wilcoxon signed rank sum 41, $p < 0.01$). TLC increased by a median of 0.32 litres (range -1.91 to +0.91 litres), sixteen subjects (76%) showing an increase after one year of nocturnal cuirass-assisted ventilation (Wilcoxon signed rank sum 57, $p < 0.05$). FRC also increased by a median of 0.12 litres (range -2.15 to +0.45 litres), 18 subjects (86%) showing an increase after one year of nocturnal cuirass-assisted ventilation (Wilcoxon signed rank sum 39.5, $p < 0.01$). There was no significant change in RV.

TABLE 28

The change in lung volumes after one year of nocturnal cuirass-assisted ventilation.

	FVC (l)			TLC (l)		
	Before	After	Change	Before	After	Change
MS	1.05	0.99	-0.06	1.62	1.74	+0.12
HH	1.10	1.15	+0.05	2.30	2.22	-0.08
SS	0.77	0.98	+0.21	1.62	1.99	+0.37
AP	0.90	0.80	-0.10	1.20	1.00	-0.20
MB	0.85	1.20	+0.35	1.99	1.89	-0.10
SG	0.26	0.44	+0.18	1.19	1.48	+0.29
WW	0.65	0.88	+0.23	1.49	1.56	+0.07
AV	0.41	0.75	+0.34	1.16	1.76	+0.60
BS	0.95	1.10	+0.15	1.60	1.90	+0.30
YH	0.50	1.10	+0.60	1.04	1.95	+0.91
JG	0.39	0.72	+0.33	1.24	1.65	+0.41
GP	0.39	0.30	-0.09	1.62	2.00	+0.38
RB	1.70	1.40	-0.30	4.20	2.90	-1.30
TC	1.10	1.13	+0.03	1.84	2.01	+0.17
WB	1.75	2.10	+0.35	5.50	3.60	+0.01
MH	0.55	0.83	+0.28	1.80	2.20	+0.40
DH	1.05	0.99	-0.06	2.05	2.11	+0.06
IJ	0.99	1.16	+0.17	2.30	2.85	+0.55
NW	0.91	0.83	-0.08	1.80	1.89	+0.09
KN	1.30	1.43	+0.13	2.32	2.78	+0.46
HC	1.20	1.40	+0.20	2.70	3.04	+0.34
Mean	0.89	1.03	+0.14	2.03	2.12	+0.09
Median	0.91	0.99	+0.17	1.80	1.99	+0.29

TABLE 29

The change in lung volumes after one year of nocturnal cuirass-assisted ventilation.

	RV (l)			FRC (l)		
MS	0.57	0.75	+0.18	0.85	0.97	+0.12
HH	1.15	1.12	-0.03	1.42	1.45	+0.03
SS	0.85	1.00	+0.15	0.85	1.29	+0.44
AP	0.33	0.33	0	0.51	0.96	+0.45
MB	0.83	1.40	+0.57	1.29	1.54	+0.25
SG	0.75	1.04	+0.29	0.86	1.09	+0.23
WW	0.84	0.68	-0.16	0.88	0.90	+0.02
AV	0.72	0.95	+0.23	0.75	1.14	+0.39
BS	0.65	0.74	+0.09	0.99	1.29	+0.30
YH	0.84	0.80	-0.04	1.06	0.91	-0.15
JG	0.76	0.93	+0.17	0.85	1.10	+0.25
GP	1.62	1.65	+0.03	1.68	1.75	+0.07
RB	2.50	1.92	-0.58	2.20	1.90	-0.30
TC	0.74	0.75	+0.01	1.18	1.24	+0.06
WB	3.45	1.50	-1.95	4.35	2.10	-2.15
MH	1.25	1.40	+0.15	1.28	1.62	+0.34
DH	1.00	1.12	+0.12	1.39	1.45	+0.06
IJ	1.31	1.69	+0.38	1.53	1.86	+0.33
NW	0.89	1.06	+0.17	1.11	1.23	+0.12
KN	1.07	1.35	+0.28	1.44	1.53	+0.09
HC	1.52	1.61	+0.09	1.72	2.05	+0.32
Mean	1.13	1.13	0	1.34	1.40	+0.06
Median	0.85	1.06	+0.12	1.18	1.29	+0.12

9:3:4 Maximum Mouth Pressures

Maximum inspiratory and expiratory mouth pressures were measured before and after one year of nocturnal cuirass-assisted ventilation in twenty subjects. This data is displayed in Table 30. There was no significant increase in MIP or MEP after one year of cuirass-assisted ventilation. Twelve subjects showed an increase in MIP, and eleven of these also had an increase in vital capacity, compared to three of the seven subjects who showed a fall in MIP ($P < 0.05$ by Fisher's exact test).

9:3:5 Maximum Voluntary Ventilation and Walking Distance

Maximum voluntary ventilation was measured before and after one year of nocturnal cuirass-assisted ventilation in sixteen subjects. This data is displayed in Table 31. MVV increased by a median of 5.5 l/min (range -6 to +22 l/min), ten subjects (62%) showing an increase (Wilcoxon signed rank sum 18, $p < 0.01$). Six minute walking distances before and after one year of cuirass-assisted ventilation were measured in thirteen subjects and are also displayed in Table 31. The median increase was 45 metres (range -24 to +184 metres), nine subjects (69%) showing an increase (Wilcoxon signed rank sum 13.5, $p < 0.05$).

TABLE 30

The change in maximum inspiratory and expiratory mouth pressures after one year of nocturnal cuirass-assisted ventilation.

Subject	MIP (cmH ₂ O)			MEP (cmH ₂ O)		
	Before	After	Change	Before	After	Change
MS	59	56	-3	54	68	+14
HH	33	35	+2	60	74	+14
SS	23	53	+30	10	56	+46
AP	46	40	-6	16	60	+44
MB	19	46	+27	34	34	0
SG	20	32	+12	36	36	0
WW	37	66	+29	53	84	+31
AV	43	52	+9	59	31	-28
YH	32	52	+20	36	95	+59
JG	25	20	-5	46	32	-14
GP	64	49	-15	33	48	+15
RB	30	30	0	58	43	-15
TC	33	44	+11	39	27	-12
WB	34	41	+7	110	104	-6
MH	29	45	+16	31	54	+23
DH	32	30	-2	53	20	-33
IJ	51	50	-1	53	60	+7
NW	16	18	+2	21	18	-3
KN	81	58	-23	106	76	-30
HC	53	65	+12	74	98	+24
Mean	38	44	+6	49	56	+7
Median	33	45.5	+4.5	49.5	55	+3.5

TABLE 31

Maximum voluntary ventilation and six minute walking distance before and after one year of nocturnal cuirass-assisted ventilation.

	Maximum Voluntary Ventilation (l/min)			Six Minute Walk Distance (m)		
	Before	After	Change	Before	After	Change
MS	28	43	+15	216	312	+96
SS	18	25	+7	372	348	-24
SG	18	21	+3			
WW	12	25	+13	167	351	+184
AV	22	22	0	17	120	+103
YH	29	29	0			
JG	12	21	+9	17	24	+7
GP	12	10	+2			
TC	33	55	+22	156	201	+45
WB	21	26	+5	189	351	+162
MH	12	22	+10	156	134	-22
DH	27	21	-6	330	348	+18
IJ	15	21	+6	120	252	+132
NW	21	18	-3	288	288	0
KN	29	27	-2	487	480	-7
HC	25	31	+6	336	420	+84
Mean	21	26	+5	219	279	+60
Median	21	23	+5.5	189	312	+45

9:3:6 Ventilatory Drive

There was no significant difference in the mean ventilatory response to carbon dioxide for the ten subjects studied before and after one year of nocturnal cuirass-assisted ventilation (Table 32).

9:3:7 Pulmonary Artery Pressure

The pulmonary artery pressures before and after one year of nocturnal cuirass-assisted ventilation were only measured in five subjects. The results are presented in Table 33. There was no change in pulmonary artery pressure after cuirass-assisted ventilation. In only four of these five subjects were cardiac output measurements possible at the time of both of the pulmonary artery pressure measurements, and in one subject a reliable pulmonary capillary wedge pressure could not be obtained. Since the effect of a year of nocturnal cuirass-assisted ventilation on pulmonary vascular resistance could only be calculated in three subjects, this data is not presented.

TABLE 32

The slope of the ventilatory response to carbon dioxide, measured using the hyperoxic rebreathing technique, before and after one year of nocturnal cuirass-assisted ventilation.

Subject	Ventilatory response to CO ₂ (l/min/kPa):		
	Before	After	Change
HH	-0.49	2.01	+2.50
MB	1.69	-1.05	-2.74
AP	-0.46	0.25	+0.71
MS	1.48	5.44	+3.96
RB	1.01	1.51	+0.50
JG	0.41	-0.79	-1.20
TC	0.96	0.98	+0.02
IJ	-1.13	5.66	+6.79
DH	-0.02	0.15	+0.17
KN	-0.78	0.54	+1.02
Median	0.20	0.76	+0.60

TABLE 33

Mean pulmonary artery pressure (PAP) before and after one year of nocturnal cuirass-assisted ventilation.

Subject	PAP (mmHg)	
	Before	After
HH	55	23
AP	21	40
BS	27	30
WW	44	35
WB	32	38

9:3:8 Right Heart Failure and Electrocardiograph Scores

The electrocardiographic, right heart failure and diuretic requirement scores of the subjects are given in Table 34. There was no difference in diuretic dosage before and after nocturnal cuirass-assisted ventilation, but both the electrocardiographic scores (Wilcoxon signed rank sum 14.5) and the right heart failure scores (Wilcoxon signed rank sum 34) were significantly lower after one year of nocturnal cuirass-assisted ventilation ($p < 0.01$).

9:3:9 Haemoglobin Concentration

Haemoglobin concentrations are displayed in Table 35. There was no significant change after one year of nocturnal cuirass-assisted ventilation.

TABLE 34

Electrocardiographic, clinical right heart failure and diuretic dose requirement scores before and after one year of nocturnal cuirass-assisted ventilation.

Subject	ECG score			RHF Index			Diuretic Index		
	Before	After	Change	Before	After	Change	Before	After	Change
SS	4	2	-2	1	0	-1	2	2	0
HH	0	0	0	1	0	0	4	4	0
AP	0	0	0	1	0	-1	2	4	+2
MB	2	0	-2	1	1	0	3	3	0
SG	1	1	0	2	0	-2	2	0	-2
WW	2	1	-1	0	0	0	2	2	0
BS	4	1	-3	1	0	-1	1	2	+1
AV	1	1	+1	1	0	-1	1	0	-1
YH	0	0	0	1	0	-1	2	1.5	-.5
MS	0	0	0	1	1	0	1	1.5	+.5
TC	5	4	-1	1	2	+1	3	2	-1
GP	0	0	0	0	0	0	0	0	0
JG	1	1	0	2	0	-2	2	2	0
RB	1	0	-1	0	0	0	0	0	0
WB	3	3	0	2	1	-1	4	1	-3
MH	4	3	-1	0	0	0	4	4	0
DH	3	3	0	0	0	0	2	4	+2
IJ	1	1	0	1	0	-1	0	0	0
NW	0	1	+1	0	2	+2	0	0	0
HC	1	1	0	1	0	-1	1	1	0
KN	1	0	-1	0	0	0	4	0	-4
Median	1	1	0	1	0	-1	2	1.5	0

TABLE 35

Haemoglobin concentrations before and after one year of nocturnal cuirass-assisted ventilation.

Subject	Haemoglobin (g/dl):		
	Before	After	Change
SS	13.2	16.5	+3.3
HH	17.2	13.1	-4.1
AP	18.3	15.5	-2.8
MB	12.8	15.4	+2.6
SG	14.9	12.4	-2.5
WW	15.8	15.2	-0.6
BS	14.3	13.8	-0.5
AV	16.2	16.6	+0.4
YH	13.9	11.9	-2.0
MS	14.7	14.6	-0.1
GP	15.8	14.4	-1.4
JG	13.4	13.4	0
RB	15.2	14.4	-0.8
WB	15.8	14.7	-1.1
MH	15.3	13.8	-1.5
DH	16.3	17.2	+0.9
IJ	14.9	14.4	-0.5
NW	13.0	15.0	+2.0
HC	14.4	15.4	+1.0
KN	16.3	15.8	-0.5

Mean	15.1	14.7	-0.4
Median	15.0	14.6	-0.5

9:4 DISCUSSION

The improvement in daytime PaCO₂ seen in this study after commencing nocturnal cuirass-assisted ventilation is in keeping with previous studies of nocturnal assisted ventilation. These are summarised in the next paragraph.

Sawicka et al (1988) have reported a series of fifty one patients treated with assisted ventilation at home, forty eight of whom used external negative pressure assisted ventilation. Excluding five subjects with severe progressive neuromuscular disease, the median duration of follow up was 4.3 years. Daytime PaCO₂ improved in fourteen of seventeen subjects for whom arterial blood gas measurements were available. Goldstein et al (1987) studied three patients with neuromuscular disease before and after twelve weeks of nocturnal cuirass-assisted ventilation. Daytime PaCO₂ fell from a mean of 7.4 to 5.9 kPa. Loh et al (1979) studied the time course of improvement in awake PaCO₂ in patients with neuromuscular disease treated with nocturnal cuirass-assisted ventilation. He found that PaCO₂ continued to fall for up to five years. Fulkerson et al (1984) reported a patient with poliomyelitis in whom five months of nocturnal cuirass-assisted ventilation resulted in a fall in daytime PaCO₂ from 9.9 to 4.5 kPa. Curran (1981) studied the effects of assisted ventilation at night using external negative pressure in 9 patients

with Duchenne dystrophy and hypercapnia. Three patients used individually-moulded cuirass shells and six patients used tank respirators, although two of the latter used cuirass shells when away from home. After a mean period of nocturnal assisted ventilation of 21.3 months, daytime PaCO₂ had fallen from 8.0 to 6.0 kPa. Marino and Braun (1982) reported an improvement in daytime PaCO₂ in 35 patients with respiratory muscle fatigue who were treated with assisted ventilation. The patients had either abnormalities of the respiratory muscles, or increased work of breathing as a result of chest wall or pulmonary disease, and used either positive airway pressure or external negative pressure ventilation as the mode of ventilatory assistance. Buchsbaum et al (1968) reported a patient with a myopathy in whom external negative pressure assisted ventilation at night using a jacket respirator induced a fall in daytime PaCO₂ from 9.5 to 5.8 kPa.

The fall in daytime PaCO₂ may be explained by a number of different mechanisms. In those subjects referred after a relatively short illness, carbon dioxide production may have been temporarily elevated initially. A fall in carbon dioxide production with recovery from the illness would reduce PaCO₂ without any change in ventilation. Thus the improvement in daytime PaCO₂ might have been observed had the subjects not been commenced on assisted ventilation.

A decline in carbon dioxide production is unlikely explain the fall in PaCO_2 in the majority of subjects, and an increase in ventilation would seem more likely. Alveolar ventilation may increase in the absence of any change in overall ventilation if the ratio of dead space to tidal volume decreases, either because of changes in the lungs themselves or a change in the respiratory pattern to a lower frequency with a larger tidal volume. Loh et al (1979) examined the serial changes in alveolar-arterial oxygen tension differences in six patients treated with nocturnal cuirass-assisted ventilation. As described above, daytime PaCO_2 and PaO_2 improved in all subjects, but only some showed a decrease in alveolar to arterial oxygen difference. This suggests that mechanisms other than improved intra-pulmonary gas exchange were involved.

Ventilation can increase in the absence of any change in the energy expenditure of the respiratory muscles if there is a change in compliance or resistance of the chest wall and lungs. One subject with a thoracoplasty (WB) showed a marked fall in TLC, FRC and RV after one year of nocturnal cuirass-assisted ventilation. The high initial values are likely to be the result of reversible airflow obstruction, which had improved by the time of the repeat assessment. In addition to changes in airway resistance, the compliance of the lungs and chest wall is much greater at lower lung volumes (Grassino et al

1973), and the work of breathing in this subject must have been considerably lower after one year of nocturnal cuirass-assisted ventilation.

The contribution of changes in the work of breathing to the improvement in PaCO_2 in the other subjects is less easy to predict. One patient with respiratory muscle weakness (RB) also showed a decline FRC, TLC and RV which would be associated with a more favourable position on the pressure volume curve for both the lungs and the chest wall, but volume changes in the remaining subjects were small and unlikely to have contributed much to changes in the work of breathing.

As discussed in Chapter 6, cuirass-assisted ventilation rests the respiratory muscles. A period of rest at night may result in increased strength of the respiratory muscles during the daytime, and the ventilation produced by these muscles could increase without any change in respiratory drive. The absence of any significant change in maximum mouth pressures for the subjects as a whole and the improvement in PaCO_2 in patients who showed a fall in maximum mouth pressures argues against a major contribution of respiratory muscle rest to the improvement in PaCO_2 .

Although the ventilatory response to carbon dioxide may be restricted by the maximum ventilatory capacity of which the subject is capable (Cameron 1979), the maximum minute ventilation achieved during the assessment

of the response to carbon dioxide was well below the maximum voluntary ventilation both before and after nocturnal cuirass-assisted ventilation. Respiratory muscle fatigue (Moxham et al 1981) and sleep deprivation (Cooper and Phillips 1982, Douglas et al 1982b) impair the ventilatory response to carbon dioxide. Cuirass-assisted ventilation should reduce respiratory muscle fatigue and improve the sleep pattern by correcting abnormalities of gas exchange during sleep (Bye et al 1986).

During the rebreathing assessment of the ventilatory response to carbon dioxide, end-tidal carbon dioxide concentration was allowed to rise by approximately two percent. The change in blood or cerebro-spinal fluid hydrogen ion concentration produced by this increase in carbon dioxide tension will depend on the carbon dioxide pool in the body. Prior to the start of nocturnal cuirass-assisted ventilation, PaCO_2 was higher than one year later. The carbon dioxide pool is also likely to have been smaller, and thus a two percent increase in end-tidal carbon dioxide concentration may have induced a larger change in cerebrospinal fluid hydrogen ion concentration than one year previously.

All these factors could contribute to an improvement in the ventilatory response to carbon dioxide. The absence of any significant change after one year of cuirass-assisted ventilation for the group as a whole may

reflect the small number of patients studied, since eight of the ten patients showed an increase.

Some of the subjects in this study complained of a feeling of breathlessness for a few minutes immediately cuirass-assisted ventilation was discontinued. Smith et al (1962) have demonstrated in normal subjects that an increased tidal volume can be a stimulus to respiration. In addition to resetting the respiratory centre to a lower PaCO_2 , the respiratory centre may become accustomed to the afferent stimuli from the lungs and chest wall. When tidal volume falls after the cessation of cuirass-assisted ventilation, the efferent output is increased in an attempt to maintain the same afferent stimulus, and is accompanied by the sensation of breathlessness. The contribution of this mechanism to the improvement in daytime PaCO_2 in the present study is uncertain.

The fall in daytime PaCO_2 was accompanied by a rise in PaO_2 . A similar increase in PaO_2 has been noted by Loh et al (1979), Sawicka et al (1988), Fulkerson et al (1984) and Goldstein et al (1986). In Sawicka's report, as in the present study, pre-treatment PaO_2 was lower in subjects with skeletal chest wall disease, including thoracoplasty, than in those with neuromuscular disease, but both showed an increase in PaO_2 after assisted ventilation. As with PaCO_2 , in some subjects recovery from the acute illness that precipitated referral for

assisted ventilation may have been accompanied by a fall in oxygen consumption. The fall in PaCO_2 suggests that alveolar ventilation increased, but PaO_2 may also have risen because of an improvement in ventilation-perfusion matching as a result of the increase in FRC. In the study by Loh et al (1979), the increase in PaO_2 in six subjects with neuromuscular disease after commencing nocturnal cuirass-assisted ventilation could not be explained by changes in alveolar-arterial oxygen gradient, which only improved in two of the subjects.

There was no significant overall change in haemoglobin concentration after one year of nocturnal cuirass-assisted ventilation. Fourteen of the subjects showed an increase in daytime PaO_2 accompanied by a fall in haemoglobin concentration, whilst four subjects showed a fall in daytime PaO_2 accompanied by an increase in haemoglobin concentration. Only two subjects showed changes in PaO_2 and haemoglobin concentration which occurred in the same direction. Although the fall in haemoglobin will reduce oxygen carrying capacity, decreased blood viscosity may lead to an improvement in tissue perfusion. Wedzicha et al (1983), Wallis et al (1984), Thomas et al (1977) and Paulson et al (1979) have shown the beneficial effects of correction of polycythaemia on pulmonary and cerebral blood flow.

Few of the previous reports of the effects of nocturnal cuirass-assisted ventilation contain lung

volume data. Curran (1981) found no change in vital capacity after nocturnal external negative pressure assisted ventilation in 9 patients with Duchenne dystrophy and hypercapnia. Fulkerson et al's patient with scoliosis following poliomyelitis showed an increase in vital capacity from 0.53 to 0.88 litres after five months of nocturnal cuirass-assisted ventilation (Fulkerson et al 1984). In Sawicka's study (Sawicka et al 1988), a small increase in vital capacity after assisted ventilation was seen in subjects with skeletal chest wall disease but not in those with neuromuscular disease. In the present study, the fall in lung volumes in one subject with a thoracoplasty (WB) mentioned above has been attributed to improvement in airflow limitation. In one subject with progressive neuromuscular disease (RB), TLC fell by over 1 litre over the year, associated with smaller declines in other lung volumes. The remainder of the subjects showed a small increase in TLC, VC and FRC.

Reproducible measurements of lung and chest wall compliance were obtained in few subjects, and this data is not therefore reported. Previous studies have produced conflicting evidence for alterations in compliance with assisted ventilation. Sinha and Bergofsky (1972) described the effect of five minutes of intermittent positive pressure breathing in subjects with scoliosis. Vital capacity increased after intermittent positive

pressure breathing in all six subjects from a mean of 1.22 to 1.46 litres. Dynamic pulmonary compliance increased and remained elevated after one hour of spontaneous ventilation, but this change is likely to have been influenced by the changes in resistance which also occurred. The vital capacity measurements were not repeated when the subjects were re-assessed after one hour of spontaneous ventilation. On the other hand, De Troyer and Deisser (1981) investigated the effect of thirty minutes of intermittent positive pressure breathing in subjects with neuromuscular disease. They measured pulmonary compliance using the semi-static occluded expiration technique and found no change after assisted ventilation. McCool et al (1986) showed no change in pulmonary compliance in a similar experiment. In his study, total respiratory system compliance was measured using the weighted spirometer technique and this also was unaffected by twenty minutes of intermittent positive pressure breathing. It is possible that the use of the weighted spirometer technique acted in a similar manner to intermittent positive pressure breathing itself, and may have corrected lung collapse prior to intermittent positive pressure breathing. Nevertheless, there is as yet no convincing evidence for prolonged alteration of pulmonary or chest wall mechanics following assisted ventilation.

There was no significant change in maximum

inspiratory and expiratory mouth pressures after one year of nocturnal cuirass-assisted ventilation. In the subjects who showed an increase in MIP, a learning or motivation effect may have been responsible. There was an association between an increase in MIP and an increase in vital capacity, but the latter may also have been affected by learning and motivation effects. In normal subjects, the pressure which can be generated by the inspiratory muscles decreases as lung volume increases (Rinqvist 1966). WB showed a decline in RV of almost 2 litres, which is likely to explain most of the modest increase in MIP. In the other subjects, changes in RV were small. Respiratory muscles fatigue more easily under hypoxic or hypercapnic conditions (Jardim et al 1981, Koulouris et al 1984, Juan et al 1984). Although increased respiratory muscle strength appears not to be involved in the improvement in daytime arterial blood gases after nocturnal cuirass-assisted ventilation, the converse may be true, in that correction of hypercapnia and hypoxia may be responsible for some of the changes in respiratory muscle strength. The increase in maximum voluntary ventilation may also reflect volitional factors, in addition to improved respiratory muscle performance or thoracic mechanics.

In normal subjects, pulmonary vascular resistance rises in response to hypoxia (Westcott et al 1951). Rises in pulmonary artery pressure during hypoxia in sleep have

been documented in subjects with airflow limitation (Boysen et al 1979), but not in subjects with chest wall disease. In the present study, pulmonary artery pressure was measured before and after nocturnal cuirass-assisted ventilation, but in only five subjects. Pulmonary vascular resistance could not be calculated in all subjects, because of methodological difficulties in measuring pulmonary capillary wedge pressure and cardiac output. The improvement in right heart failure seen in this study may nevertheless relate to a fall in pulmonary vascular resistance after correction of hypoxia, together with an improvement in ventricular performance on account of increased PaO_2 , and in some cases decreased blood viscosity. Sawicka et al (1988), Fulkerson et al (1984) and Hoepfner et al (1984) have reported improvement in right heart failure in patients with chest wall disease after commencing nocturnal assisted ventilation. Turino et al (1965) studied the effect of external negative pressure ventilation using a tank respirator on pulmonary artery pressure in a subject with scoliosis. Pulmonary artery pressure fell, but this may also have been affected by the administration of supplementary oxygen.

CHAPTER 10

SOCIO-ECONOMIC ASPECTS OF CUIRASS-ASSISTED VENTILATION

10:1 INTRODUCTION

Previous studies of home nocturnal assisted ventilation have recorded symptomatic improvement (Sawicka et al 1988) and a reduction in the amount of time patients spend in hospital (Kompalitch et al 1979, Levi Valensi et al 1979). The cost of assisted ventilation at home has been estimated for intermittent positive pressure ventilation (Creese and Fielden 1977, Goldberg et al 1980, 1984, Banaszczak et al 1981, Feldman and Tuteur 1982, Burr et al 1983, Sivak et al 1983), but not for cuirass-assisted ventilation.

In the present chapter, the effect of nocturnal cuirass-assisted ventilation on symptoms, hospital admission rates and general practitioner attendances is examined. An estimate is made of the capital and recurrent costs of home cuirass-assisted ventilation.

10:2 METHODS

Details of the visual analogue symptom scores, depression scores and mental function tests used are given in Chapter 3. Patients performed the tests on their first admission and twelve to fifteen months later, at the time of the measurements detailed in Chapter 9.

The number of hospital admissions and the number of days spent in hospital were obtained from the subjects notes. Admissions solely for the purpose of the studies

described in other chapters were not included in this analysis. Details of district nurse, home help and hospital engineer visits during the year were obtained directly from each subject. Questionnaires were sent to the general practitioner of each subject, requesting details of attendances in the year prior to and in the year subsequent to commencing cuirass-assisted ventilation. Completed questionnaires or photocopies of records were received from all but one of the general practitioners, and the subject for whom a reply was not received was able to supply the requisite details from personal records.

The capital costs were calculated on the basis of a construction cost of £36,000 for the assisted ventilation unit, with £20,000 for purchase of monitoring equipment. This cost was distributed between the 60 subjects admitted to the unit each year, assuming a life of 25 years for the assisted ventilation unit and 10 years for the monitoring equipment. The cost of materials for each cuirass was £50 and of a cuirass pump was £2085. These costs were distributed assuming a life of 2 years for each cuirass and 10 years for the pump. The expense of purchasing a new mattress was taken as £200. If a new power point was required in the patient's bedroom for the cuirass pump, the cost of this was taken as £20. No other home alterations were necessary.

The cost of transporting the subjects to hospital was

calculated at a rate of 30p per mile. The expense incurred by subjects providing their own transport was not included, but the cost of rail fares for the two Scottish subjects paid by their own health authorities were included.

Shared in-patient costs per patient per day were taken from health authority statistics as follows: general services £26.77, medical and surgical equipment £3.36, direct treatment services £1.86 and medical staff £9.59. Nursing costs for the assisted ventilation unit were calculated on the basis that one additional nurse was needed on each shift on the respiratory ward to provide nursing on the assisted ventilation unit, resulting in a cost of £41.1 per patient per day. One physiotherapist spent approximately half of her time on the assisted ventilation unit, leading to a cost of £9.33 per patient per day. The cost of investigations was estimated assuming that a chest radiograph, electrocardiograph, lung function tests, full blood count, urea and electrolytes and arterial blood gases were performed on each patient once per admission, leading to an estimated total cost of £42 per patient per admission.

Home care costs were calculated using a rate of £3.50 per general practitioner visit, £4.74 per district nurse attendance and £2.00 per hour for home help. The cuirass pump consumes approximately 2 units of electricity per

hour, the cost of a unit at the time of the study being 5.13p.

10:3 RESULTS

Visual analogue symptom scores and depression scores before and after one year of nocturnal cuirass-assisted ventilation are displayed in Table 36. There was no significant change in breathlessness, temper or tiredness after one year of nocturnal cuirass-assisted ventilation, but depression scores had decreased by a median of 3 points (Wilcoxon signed rank sum 1, $p < 0.01$). The times taken to perform simple mental function tests are given in Table 37. The trail test time decreased by a median of 7 seconds (Wilcoxon signed rank sum 1, $p < 0.01$) after one year of nocturnal cuirass-assisted ventilation, but there was no significant change in the other tests.

TABLE 36

Visual analogue symptom scores and depression score before and after one year of nocturnal cuirass-assisted ventilation.

Symptom:	Happiness		Breathlessness		Tiredness		Depression	
	Before	After	Before	After	Before	After	Before	After
Subject								
HH	20	30	52	35	65	88	4	2
MB	51	26	52	33	47	54	12	4
WW	3	2	69	61	5	3	5	3
AV	4	0	28	3	0	4	6	0
MS	19	2	9	14	82	44	7	4
TC	7	73	0	34	0	49	5	6
JG	0	24	0	41	15	33	8	4
WB	0	12	67	39	0	25	4	1
MH	0	4	6	26	6	44	10	4
HC	13	5	13	25	50	71	4	3
KN	0	0	50	49	28	41	3	1
Median	3.5	5	28	33	15	44	5	3

TABLE 37

Mental function test scores before and after one year of nocturnal cuirass-assisted ventilation.

Test:	Trail		Deletion		Copying		Decoding	
	Before	After	Before	After	Before	After	Before	After
Subject								
HH	37	34	31	36	42	40	122	74
MB	80	53	59	48	41	41	81	70
WW	41	36	32	48	47	40	53	56
AV	30	25	44	38	50	28	56	46
MS	33	25	24	27	40	34	55	97
TC	66	35	35	17	26	29	55	56
WB	51	33	37	38	40	42	67	62
MH	60	44	40	45	55	41	84	47
KN	50	36	28	21	27	30	59	46
JG	30	32	33	31	33	29	77	51
HC	31	37	25	17	29	27	49	34
Median	41	35	33	36	40	34	59	56

The details of hospital admissions over the year after commencing nocturnal cuirass-assisted ventilation are given in Table 38. Admissions were mainly routine for monitoring purposes, only occasionally being precipitated by an acute respiratory illness or equipment failure. The Newmarket hospital engineers visited five subjects at home once during the year (AP,MB,WW,AV,DH) and two subjects twice (HH,GP). Engineers from a local hospital visited TC once during the year. Three subjects were visited by the district nurse once a week and three subjects received home help once a week. The number of general practitioner visits to each subject and the number of antibiotic prescriptions per subject were not significantly different in the year after commencing nocturnal cuirass-assisted ventilation compared to the previous year (Table 39).

Seven subjects needed new mattresses and two subjects had power points moved at the time of commencing home assisted ventilation. The estimated cost of home cuirass-assisted ventilation is detailed in Table 40.

All patients were able to leave their home during the daytime and four had been away on holiday with their cuirass and pump, in two instances outside the United Kingdom. Two patients were in full time and one in part time employment prior to the illness which led to their referral to Newmarket. One patient continued in full time employment after starting cuirass-assisted

ventilation at home, but the other two patients stopped work. Thirteen patients had relatives at home who were actively involved in their care. Eight of these relatives were at work prior to the start of cuirass-assisted ventilation at home, and none stopped work thereafter. Three patients using cuirass-assisted ventilation at home were able to continue caring for relatives at home.

TABLE 38

Admissions to hospital during one year of nocturnal cuirass-assisted ventilation.

Subject	Number of hospital admissions	Hospital transport (Y/N)	Total days in hospital	Distance in miles from hospital
MS	2	Y	11	400
HH	1	Y	8	40
SS	9	Y	36	40
AP	10	Y	28	15
MB	5	Y	12	50
SG	7	Y	38	40
WW	8	Y	40	40
BS	6	N	14	15
AV	4	N	9	35
YH	3	Y	31	200
JG	4	Y	9	30
RB	6	Y	15	30
TC	1	Y	7	400
GP	10	N	28	20
WB	9	N	26	10
MH	9	N	45	25
DH	5	N	21	40
IJ	11	N	59	40
NW	8	Y	31	15
KN	4	N	8	50
HC	4	N	11	45

Mean	6		23	
Median	6		21	

TABLE 39

General practitioner attendances and antibiotic prescriptions during one year of nocturnal cuirass-assisted ventilation (CAV) and for the year prior to commencing nocturnal cuirass-assisted ventilation.

Subject	General practitioner attendances (per year)		Antibiotic prescriptions (per year)	
	CAV	Prior to CAV	CAV	Prior to CAV
MS	4	6	2	4
HH	11	11	4	1
SS	8	8	1	0
AP	10	13	0	2
MB	0	0	0	0
SG	9	1	1	6
WW	20	16	3	4
BS	6	5	1	1
AV	2	3	0	0
YH	6	1	0	2
JG	3	10	3	2
RB	4	7	0	0
TC	4	18	1	4
GP	6	14	0	1
WB	3	1	1	0
MH	1	7	0	1
DH	2	5	0	1
IJ	3	30	1	3
NW	9	10	1	1
KN	22	16	0	0
HC	7	25	0	0
Median	6	7	1	1

TABLE 40

Estimated cost per patient of one year of nocturnal cuirass-assisted ventilation at home.

Capital costs	£
Assisted ventilation unit	24
Monitoring equipment	33
Cuirass pump	208
Cuirass	25
Home alterations	58
Recurrent costs	
Transport between hospital and home	65
Hospital Care	
Shared in-patient costs	
General services	615
Medical & surgical equipment	77
Direct treatment costs	43
Additional assisted ventilation unit costs	
Nursing staff	945
Medical staff	220
Physiotherapy	214
Technical staff	32
Investigations	252
Home Care	
General Practitioner	23
District Nurse	35
Home Help	15
Electricity	37
Technical staff	6
TOTAL (per patient per annum)	2868

10:4 DISCUSSION

Despite the physiological changes described in the previous chapter, one year of nocturnal cuirass-assisted ventilation failed to relieve symptoms as assessed by visual analogue scales. The reduction in depression scores is more likely to reflect the positive approach to the patients taken by the Assisted Ventilation Unit staff, and although the shorter trail test time could indicate improved cerebral function, it may also be due to a learning effect.

The policy of the Assisted Ventilation Unit was to readmit patients for assessment rather than to visit them at home or to see them as outpatients. As a result the number of admissions to hospital per year in this study is greater than that reported by Splaingard (Splaingard et al 1982 and 1983). Many of his patients used tank respirators, which are difficult to transport to hospital.

As might be expected, the amount of support the patients received at home, either from relatives or health care services, was considerably lower than that needed for more severely disabled patients requiring intermittent positive pressure ventilation (Dunnell et al 1972). Since the largest contribution to the cost of maintaining such a patient at home is nursing care (Splaingard et al 1983), it is not surprising that nocturnal cuirass-assisted ventilation is comparatively

inexpensive. In the United Kingdom, the cost of home care for patients using intermittent positive pressure ventilation is 70% of hospital care (Creese and Fielden 1977, Goldberg et al 1984), whereas in the USA the cost of home care is generally less than 50% of hospital care, and can be as low as 10% (Goldberg et al 1980, Banaszczak et al 1981, Feldman and Tuteur 1982, Burr et al 1983, Sivak et al 1983). This may reflect the higher cost of hospital care in the USA. In France home positive pressure ventilation has been estimated to cost 5% to 20% of hospital care (Tourreau et al 1978, Robert et al 1979).

Previous studies of intermittent positive pressure ventilation at home have demonstrated a fall in the number of episodes of acute respiratory failure (Kompalitch et al 1977, Levi Valensi et al 1979, Robert et al 1979). In the present study, none of the patients had been admitted to hospital in the year prior to commencing home assisted ventilation. No reduction was seen in the number of general practitioner attendances, but in many instances the visits by the general practitioners were for routine assessment, and these may have increased in frequency after the patient commenced nocturnal cuirass-assisted ventilation.

In view of the low demands on health care services made by the patients in the previous year, it is difficult to assess what the cost of their care would

have been had they not commenced assisted ventilation. The patients who could not be weaned from intermittent positive pressure ventilation would probably have had to spend every night in hospital, and in these two patients the savings must have been considerable.

It proved impossible to identify a satisfactory control group with which to compare the subjects receiving nocturnal cuirass-assisted ventilation. The survival of these subjects without such intervention is therefore difficult to predict. Bergofsky et al (1959) documented a fifty percent one year survival for subjects with scoliosis in whom respiratory failure was not treated. In contrast, Libby et al (1982) reported twenty patients with scoliosis who presented with acute respiratory failure, seven of whom required assisted ventilation. The mean number of further acute episodes of respiratory failure was 0.4 per patient per year.

The one year survival rate in the present study was 84% and is comparable with previous reports. In Sawicka's study of long term assisted ventilation (Sawicka et al 1988) five subjects with severe progressive neuromuscular disease died after a median period of 13 months assisted ventilation. A further seven deaths occurred in subjects with less severe neuromuscular disease, making a total of 12 deaths in the group of twenty seven subjects with neuromuscular disease included in the study. One subject with skeletal chest

wall disease of the twenty who continued with assisted ventilation died during an acute respiratory tract infection.

The French experience of long term assisted ventilation is wider than in the United Kingdom, although most reports concern intermittent positive pressure ventilation in patients with chronic airflow limitation. Kompalitch et al (1977) described twenty three patients with respiratory failure as a result of scoliosis or pulmonary tuberculosis who received intermittent positive pressure assisted ventilation either via the mouth or via a tracheostomy at home. The five year survival was 77%. Achard et al (1979) reported a group of subjects with scoliosis, the sequelae of tuberculosis or chronic airflow limitation. Forty four subjects were followed for a period between one and eight years. All six deaths occurred in the first eight months.

Hoepfner et al (1984) and Weirs et al (1977) have noted long term survival with nocturnal assisted ventilation in scoliotic patients, using intermittent positive pressure ventilation and cuirass-assisted ventilation respectively. Uncontrolled studies of nocturnal IPPV suggest better survival of patients with thoracoplasty and scoliosis than with airflow obstruction (Robert et al 1979). Survival in patients with neuromuscular disease varies according to the natural history of the disease process. In Duchenne muscular

dystrophy, nocturnal assisted ventilation prolongs life by an estimated 3 years (Alexander et al 1979).

Both of the patients without progressive neuromuscular disease who died lived alone (BC, DS). Although Robert et al (1979) suggested that living alone was a contraindication for home assisted ventilation, two other patients continue to manage alone having used cuirass-assisted ventilation at home for over a year.

CHAPTER 11

THE CUIRASS PUMP

11:1 INTRODUCTION

The history of the pumps used to provide negative pressure for cuirass-assisted ventilation is outlined in the first section of this chapter. The following sections describe studies of the flow rates needed to generate adequate peak external negative pressures with the cuirass respirator, and the ability of the two cuirass pumps most commonly used in the United Kingdom to generate and maintain these pressures. The effect of the cuirass volume and the tubing which connects the pump to the cuirass on the peak negative pressure and the pressure waveform are also examined.

In recent years, a pump with a servo mechanism and the facility for external control of the pressure waveform has become available for cuirass-assisted ventilation. The behaviour of the servo mechanism of this pump and the ability to achieve patient-triggered ventilation are discussed in the later sections of this chapter.

11:2 THE HISTORY OF PUMPS USED FOR EXTERNAL NEGATIVE PRESSURE VENTILATION

A simple bellows pump was used with the earlier models of cuirass. This was initially operated by hand (Von Hauke 1874, Bell 1910) or foot (Eisenmenger 1904). The necessity for an assistant to move the bellows limited the time for which cuirass-assisted ventilation

could be maintained, and Eisenmenger successfully linked the bellows of his pump to an electric motor (Eisenmenger 1904). A bellows or piston pump attached to an electric motor was subsequently used with the Steuart, Kifa, Mullikin, Blanchard and London County Council cuirass designs. The principle behind these pumps of a fixed volume-cycled pump with an adjustable leak was also employed in the Drinker pump unit (Scales et al 1953). A smaller version of this pump was developed by the Medical Research Council for use with the Spirashell cuirass (Medical Research Council Report 1956-1957), and the same design was adopted for the Cape cuirass pump which remains in use to the present day. Later models included a pulley system with which the rate of the pump cycle can be varied. A battery was incorporated in the Monaghan pump so that it could still be used in the event of power failure. The Cape design includes the facility for the bellows to be operated by hand. Scales et al (1953) used a valve with a spring mechanism to modify the pressure waveform produced by the bellows pump.

Breuillard introduced the concept of using a chamber which was maintained continuously at negative pressure for external negative pressure ventilation. A valve in the pipe connecting the negative pressure source to the inside of the cuirass could be opened and closed to produce the required negative pressure cycle in the cuirass (Emerson 1958). Using this principle, Eisenmenger

replaced his electric bellows with a fan powered by an electric motor, and similar pumps were used with the Burstall cuirass, the Tunncliffe jacket, and with Binet's moulded cuirass (Burstall 1938, Spalding and Opie 1958, Binet and Bochet 1961).

11:3 PUMPS USED IN THE PRESENT STUDY

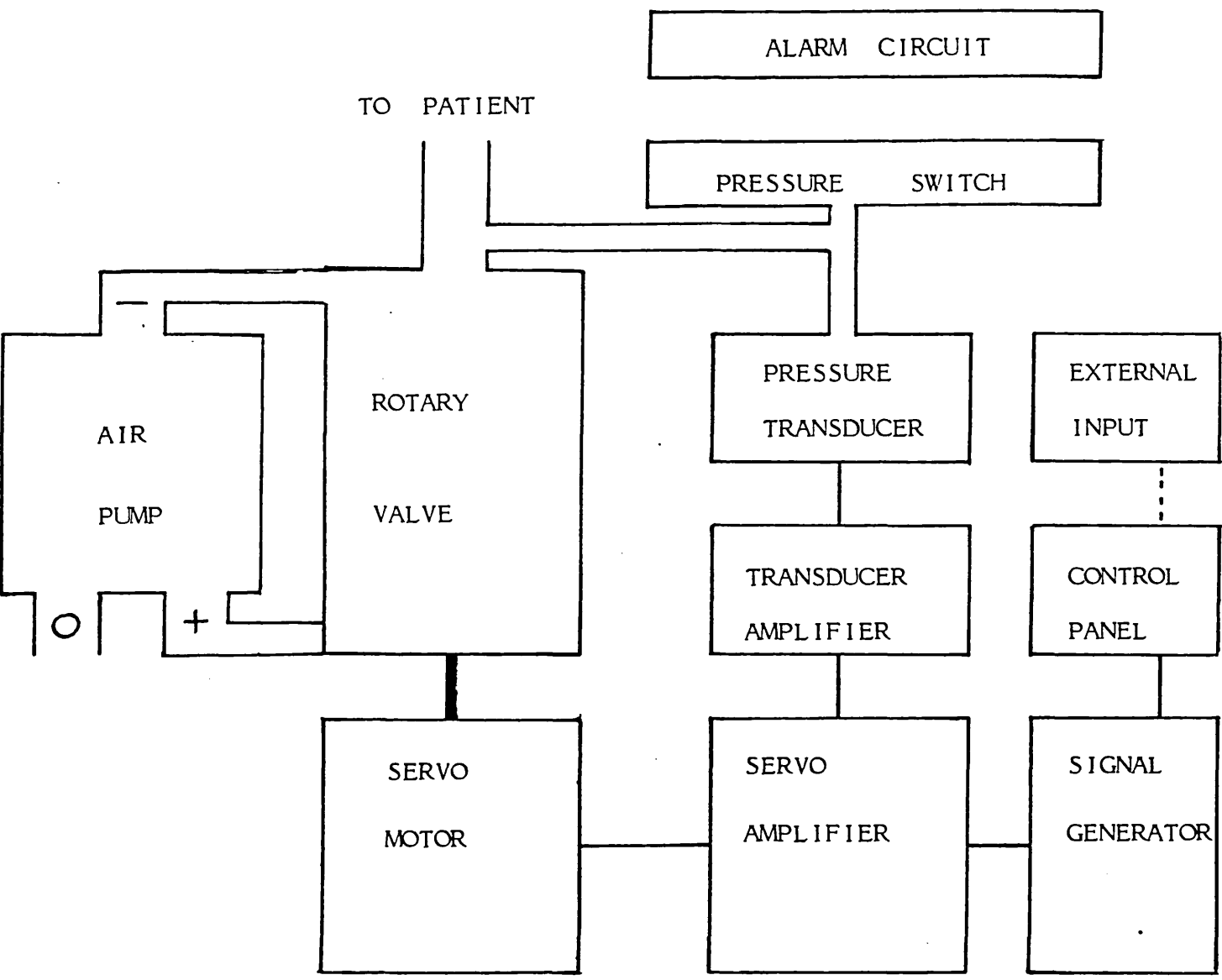
The Cape cuirass pump was used as an example of a bellows pump. This rugged and reliable design of pump has been in use for many years and is the standard pump used for cuirass-assisted ventilation in the United Kingdom. The stroke volume of the bellows is 17.6 litres. Five speeds are available: 13, 16, 19, 22 or 25 cycles per minute. These are changed by moving the position of the belt on a pulley system inside the pump casing. A safety switch ensures that the electric motor stops whenever the door of the pump casing is opened to change the speed. The peak external negative pressure which is produced inside the cuirass shell is controlled by a variable leak valve on top of the pump. Closure of another valve on the top of the pump can be used to produce external positive pressure during the expiratory phase of the pump cycle.

The Newmarket pump was used in the majority of the investigations described both in this chapter and in the rest of this thesis. This is a fan pump with a valve mechanism controlled electronically. The design of the pump is outlined in Figure 6, and the pump itself is

shown in Figure 5. Like the fan pumps described above, an electric motor drives a fan to provide negative and positive pressure sources. A separate motor operates a rotary valve which controls the connection between the tubing which leads to the inside of the cuirass and the negative or positive pressure sources.

FIGURE 6

Block Diagram of the Newmarket Pump



11:4 METHODS

11:4:1 Peak external negative pressure and pump flow

The relationship between pump flow and the negative pressure within the cuirass was investigated in 10 patients using individually moulded cuirass respirators. A square digital waveform with an inspiratory:expiratory ratio of 1:1 was generated by a BBC B computer at a rate 15 cycles per minute. This digital waveform was converted into a voltage waveform by a digital to analogue converter which was connected to the Newmarket cuirass pump. The external input connection on the pump control panel bypasses the signal generator shown in Figure 6, and utilises the servo mechanism to generate the desired waveform. The amplitude of the signal fed into the pump was varied in order to generate an end-inspiratory pressure plateau of between 2.5 and 40 cm of water. Pump flow was measured using a factory-calibrated Mercury Electronics pneumotachograph flow head and pressure transducer with a linear response up to 3000 l/min.

When this waveform was used to control cuirass-assisted ventilation, external negative pressure reaches a plateau towards the end of the inspiratory phase of the cycle. At this time, the pump flow trace also showed a plateau, this being equal to the flow through leaks round the edge of the cuirass shell.

11:4:2 Pump flow rate and pressure waveform

Although a squarewave signal was used in the previous section, the external negative pressure waveform produced was not a square wave. It showed considerable slurring of the initial phase of the waveform when pressure began to fall below atmospheric pressure. This damping of the waveform could be caused by leakage of air round the edges of the cuirass shell, by deformation of the cuirass shell in response to the negative pressure within it, by the tubing connecting the shell to the pump, or it could be inherent in the pump servo mechanism.

In order to examine the cause of the pressure waveform damping, a model was constructed to simulate a cuirass shell using a length of 20cm diameter rigid plastic pipe. The volume of the pipe was 15 litres, this being the estimated volume of air inside a typical cuirass shell when attached to the patient. The connecting tubing from the pump was attached to one end of the pipe and the other end was sealed off. A side hole was made in the tubing to permit monitoring of pressure inside the pipe. A tap was fitted to a larger hole in the side of the pipe which could be opened to produce a variable leak. A square waveform was fed into the pump servo mechanism, and the pressure waveform generated within the cuirass model was recorded whilst the leak was varied.

11:4:3 External negative pressure and the connecting tubing

The pressure drop down the tubing connecting the cuirass to the pump was investigated by the use of the square waveform with the cuirass model described in the previous section. A 2 metre length of 35mm internal bore tubing was used. One side of a differential pressure transducer was attached to the pipe inlet on the cuirass pump. The other side of the pressure transducer was connected to the pipe cuirass model. With the tap adjusted so that at a negative pressure of -30 cm of water a leak flow rate of approximately 1300 l/min was obtained, the amplitude of the square wave was increased to produce flow rates of 300 to 1500 litres per minute. The pressure drop down the tubing was measured at each flow rate. In view of the pressure drop down the connecting tubing shown with the cuirass model, the effect of using the pressure inside the cuirass in the pump servo loop rather than the pressure at the pump inlet was investigated. The pressure transducer in the pump servo mechanism was connected to a 2.5 metre length of tubing of 2 millimetre internal diameter. The other end of this tubing was placed either inside a cuirass shell, or at the pump inlet, the latter being the point where pressure is sensed when the pump is operating normally. The peak external negative pressure within the cuirass was recorded using these two sites of pressure sensing.

11:4:4 Servo control of peak external negative pressure

The ability of the Cape and Newmarket pumps to adjust to variable leaks was studied with the rigid pipe cuirass model described above. The effect of decreasing the leak was investigated by setting the pumps to give -30 cm of water peak suction pressure with the leak tap fully open. The leak was then progressively closed and the peak negative pressure recorded. The effect of increasing leakage was studied by setting the pumps to generate a pressure of 30 cm of water with a leak of less than 10 litres per minute and then gradually opening the leak tap without adjusting the pumps. The amplitude of the pressure waveform was recorded as the leak flow rate increased.

The ability to cope with variable leaks in clinical use was studied by recording the intracuirass pressure overnight. The same subject was studied on two consecutive nights using the Cape pump on the first night and the Newmarket pump on the second. After initially adjusting the pressure to the patient's usual peak suction pressure of -35 cm of water, no further adjustments were made to the pump. Intracuirass pressure was monitored by the BBC C computer using a SiPlan pressure transducer and analogue to digital converter. The peak pressure for each pump cycle was determined by the computer and recorded onto disc.

11:4:5 Patient-triggered cuirass-assisted ventilation

Attempts were made to trigger the Newmarket pump to generate external negative pressure to coincide with spontaneous respiratory effort. The onset of inspiration was sensed using the Respitrace system to detect ribcage motion, a nasal thermistor to detect nasal airflow, and surface electromyograms to detect inspiratory muscle contraction. These analogue signals were converted into digital form and fed into a BBC B microcomputer. The computer was programmed to wait until the input signal reached a preset threshold, after which a half sinewave was fed out through a digital to analogue converter to the pump. The amplitude of the half sinewave was adjusted to produce a satisfactory peak external negative pressure. If the input signal did not reach the threshold value within five seconds of the end of the previous breath, the computer was programmed to generate the half sine waveform irrespective of the state of the input signal.

Ear oxygen saturation during sleep was studied in two subjects during sleep using untriggered and triggered cuirass-assisted ventilation for alternate hours, with sternomastoid electromyographic activity as the trigger stimulus.

11:5 RESULTS

11:5:1 Peak external negative pressure and pump flow

The shape of the pressure and flow waveforms obtained with the square pressure waveform external signal generator are shown in Figure 7a and 7b respectively. These demonstrate the plateau of pressure and flow towards the end of inspiration. The relationship between end-inspiratory flow rate and pressure using this square waveform is shown in Figure 8.

FIGURE 7

Pump Flow and Pressure Waveforms
(See text for details)

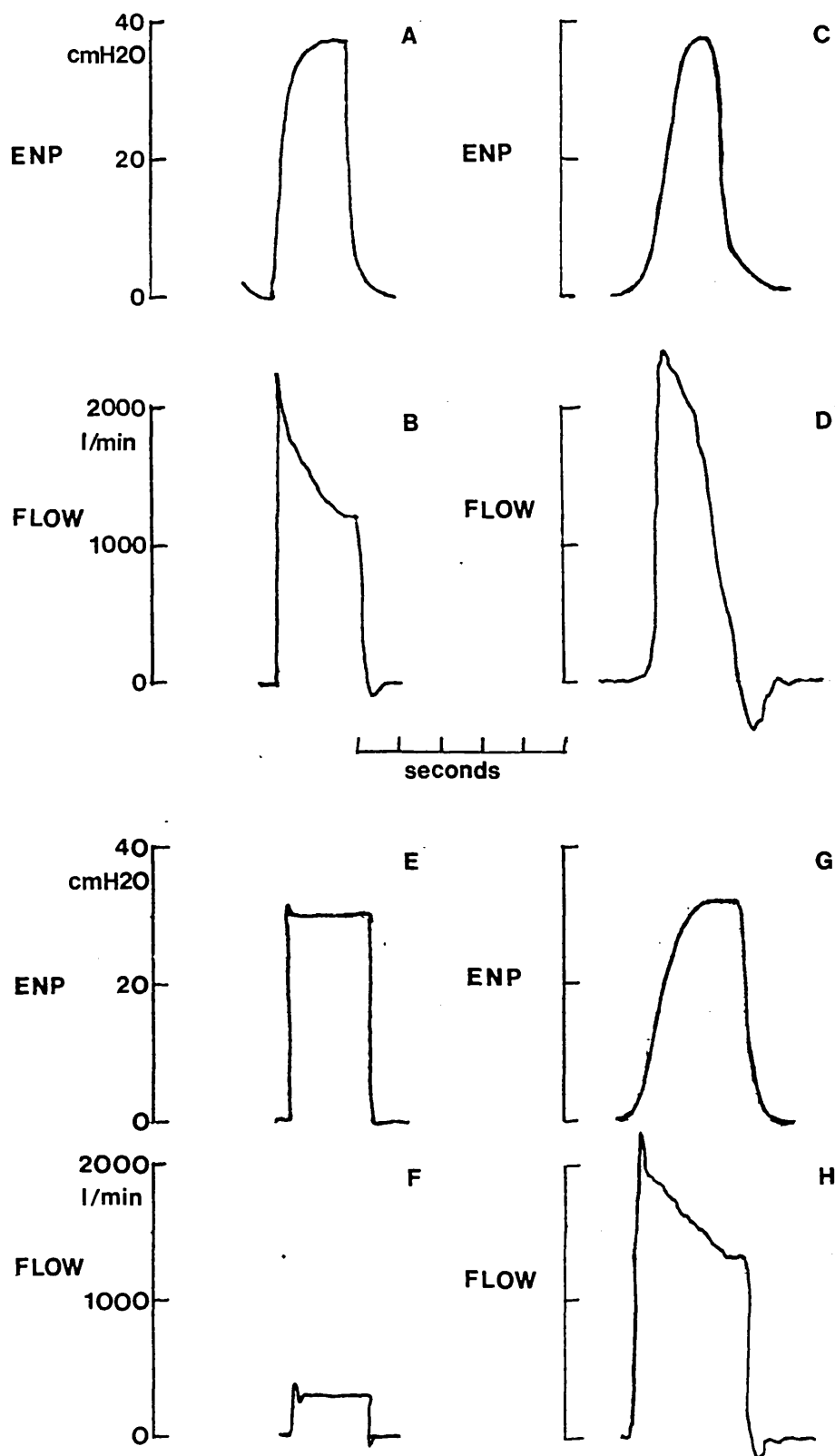
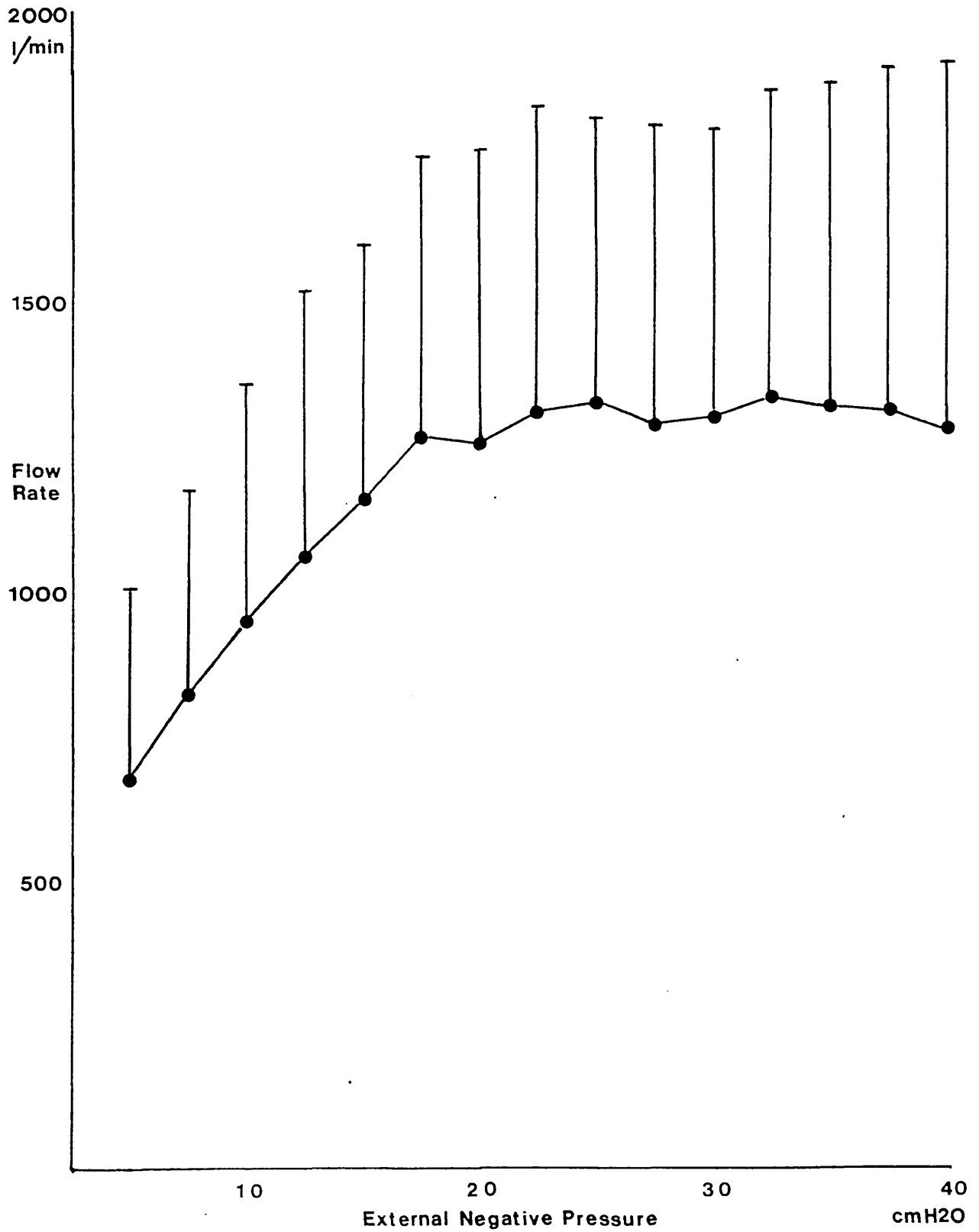


FIGURE 8

The Relationship Between Pump Flow and
Peak External Negative Pressure



11:5:2 Pump flow rate and pressure waveform

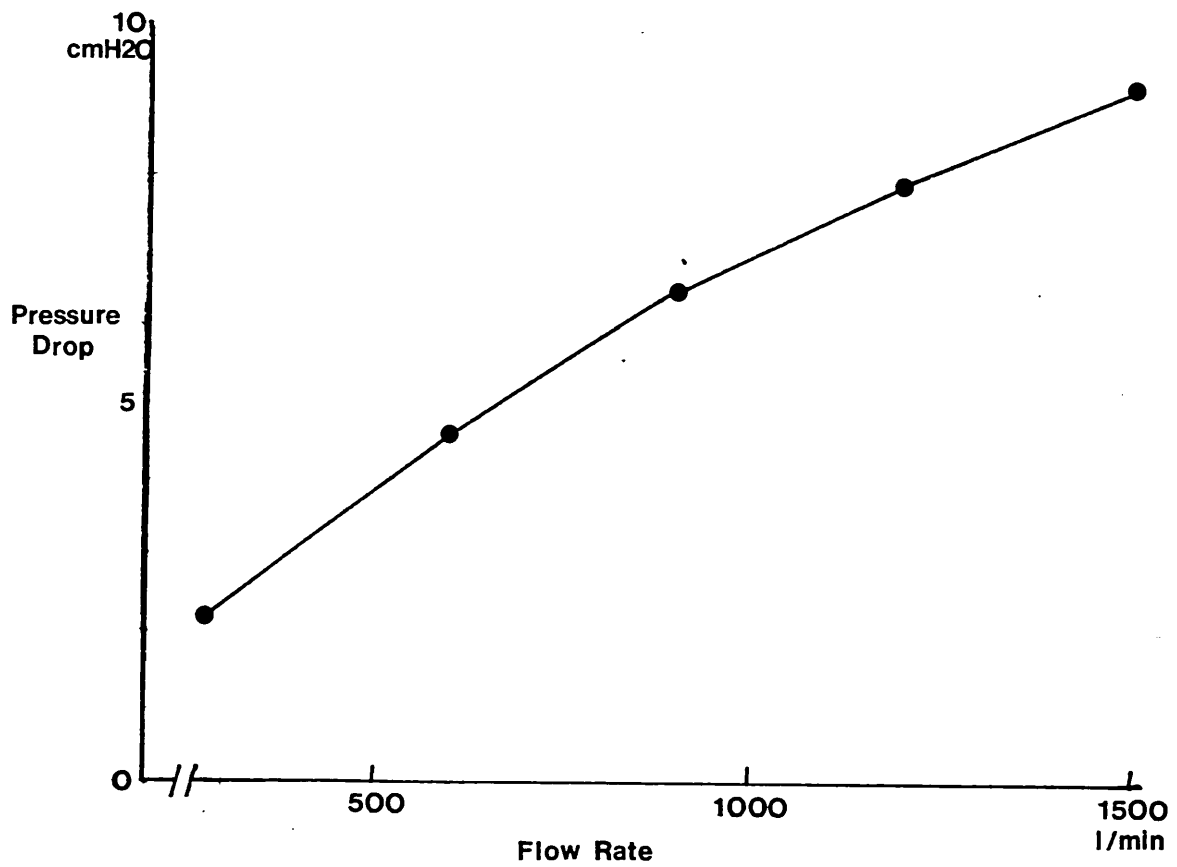
The pressure and flow waveforms obtained with the cuirass model and a leak flow rate of 1300 l per minute are shown in Figures 7g and 7h respectively. These are similar to those obtained in the studies described in section 11:5:1, using a square waveform for cuirass-assisted ventilation (Figures 7a and 7b). The pressure and flow waveforms obtained at a leak flow rate of 300 l/min are shown in Figure 7e and 7f respectively. Decreasing the leak flow below 300 l/min caused the servo mechanism to hunt, resulting in oscillation of the pressure plateau. As the leak flow rate was decreased from 1300 to 300 l/min, the pressure waveform became similar to the template waveform fed into the pump, with a servo loop overshoot at the start.

11:5:3 External negative pressure and the connecting tubing

Figure 9 shows the relationship between flow and pressure drop down the connecting tubing. Changing the pressure used in the servo mechanism from the pump inlet pressure to the pressure inside the cuirass shell resulted in an increase in peak external negative pressure from 30 to 37 cmH₂O.

FIGURE 9

The Relationship between Flow Rate and
Pressure Drop down the Connecting Tubing



11:5:4 Servo control of peak external negative pressure

The pressure and flow waveforms obtained with pressure inside the cuirass used in the servo feedback loop are shown in Figure 7c and 7d.

Figure 10 shows the ability of the Newmarket and Cape pumps to compensate for increasing leakage flow rate. There is little difference between the two pumps, but the Newmarket maintains a slightly higher pressure than the Cape. Figure 11 shows the peak suction pressure produced with decreasing leakage flow rate. The Cape pump produces a higher pressure than the Newmarket pump, generating peak suction pressures of -100cm of water.

The distribution of peak suction pressures overnight with the two pumps are given in Figures 12 and 13.

FIGURE 10

The Effect of Increasing Leak on Peak External Negative Pressure (Peak ENP) for the Cape and Newmarket Cuirass Pumps

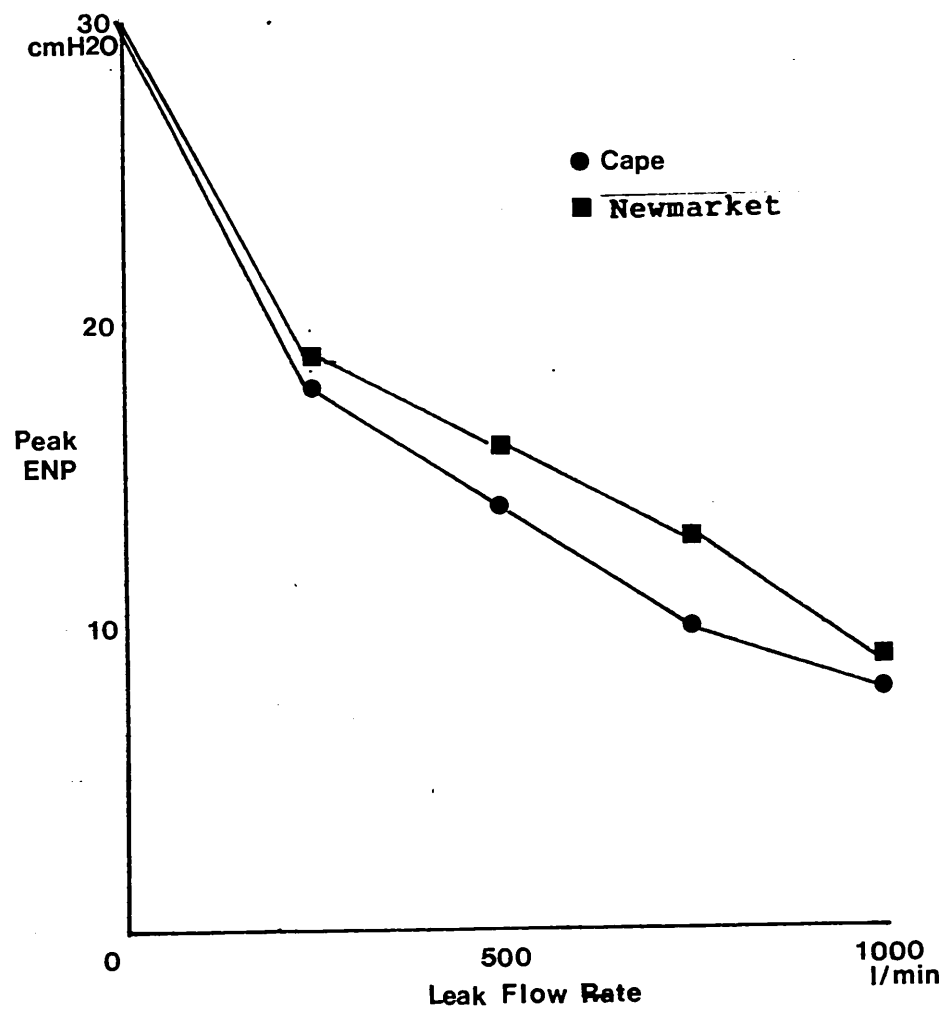


FIGURE 11

The Effect of Decreasing Leak on Peak External Negative Pressure (Peak ENP) for the Cape and Newmarket Cuirass Pumps

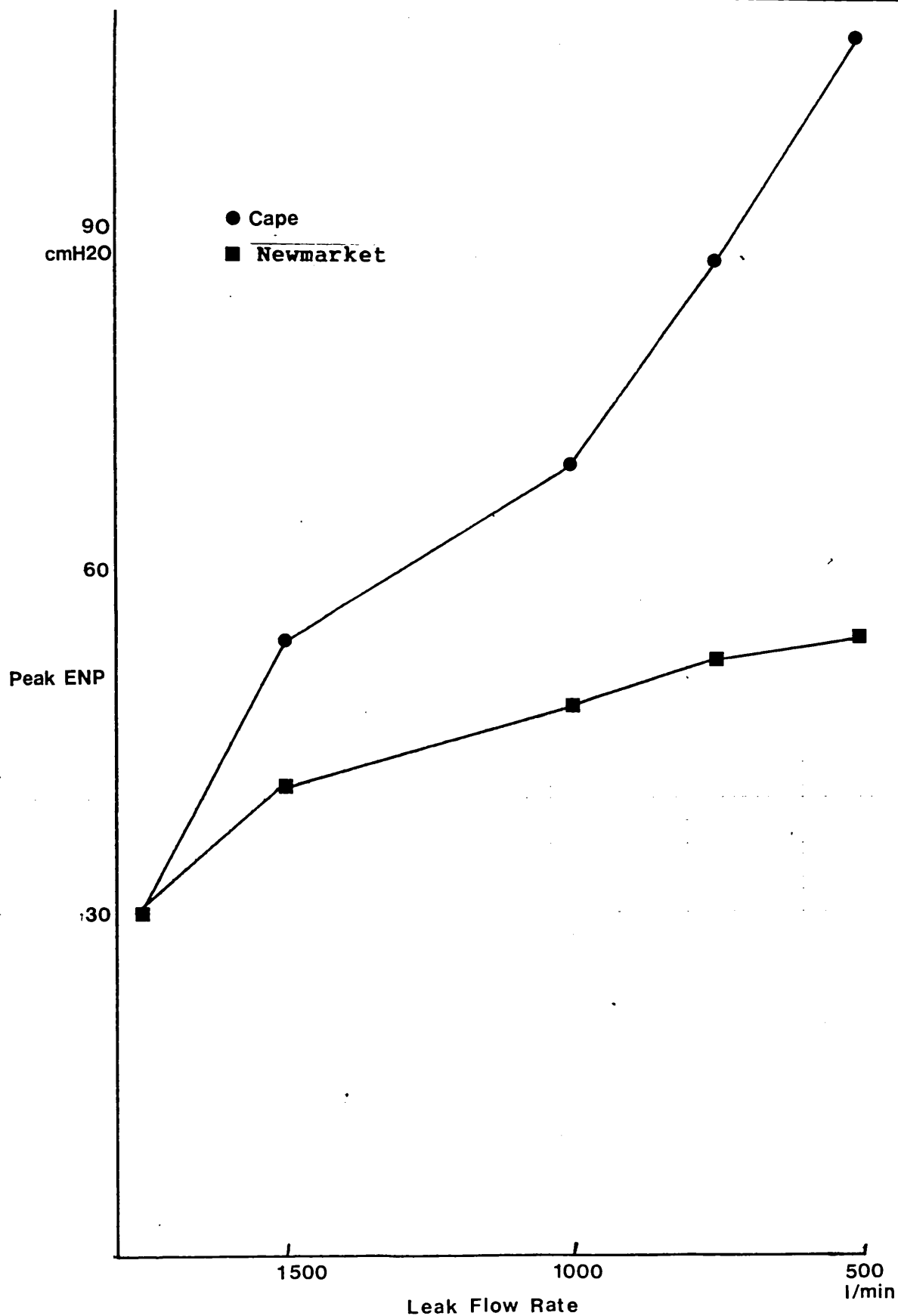


FIGURE 12

Percentage of Pump Cycles Reaching Different Peak External
Negative Pressures (Peak ENP) with the Cape Cuirass Pump

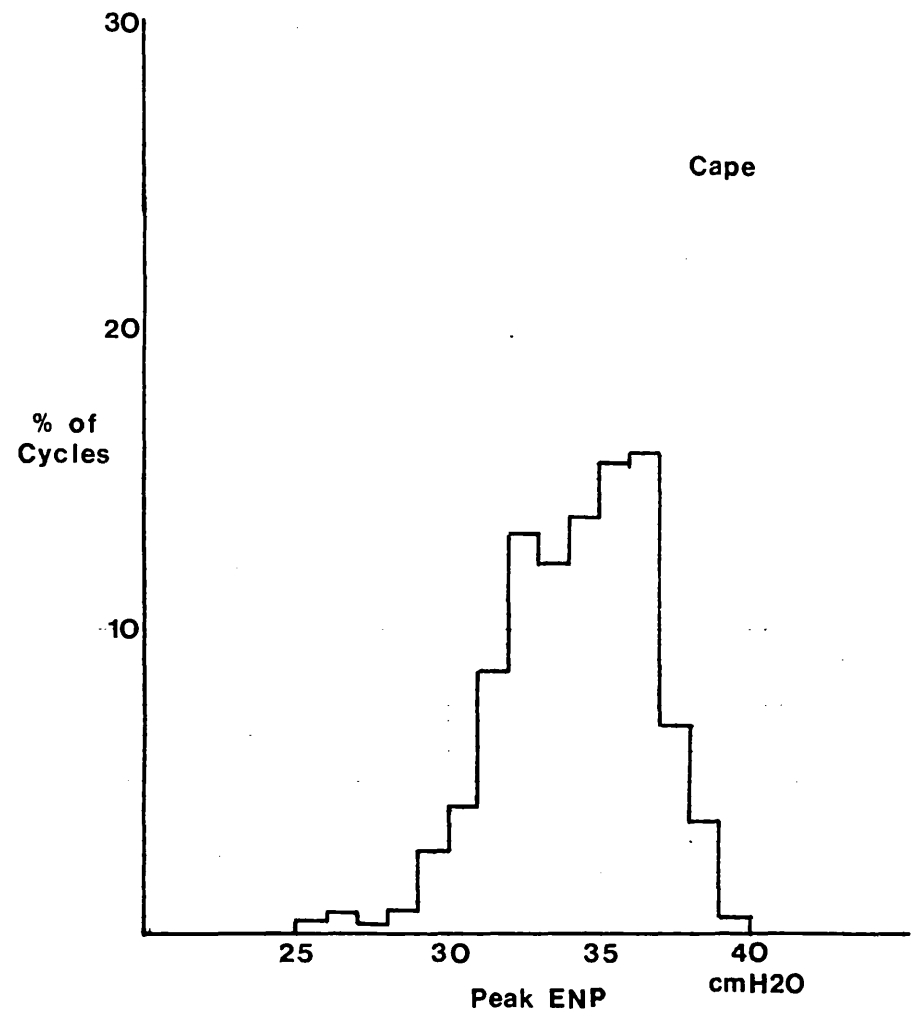
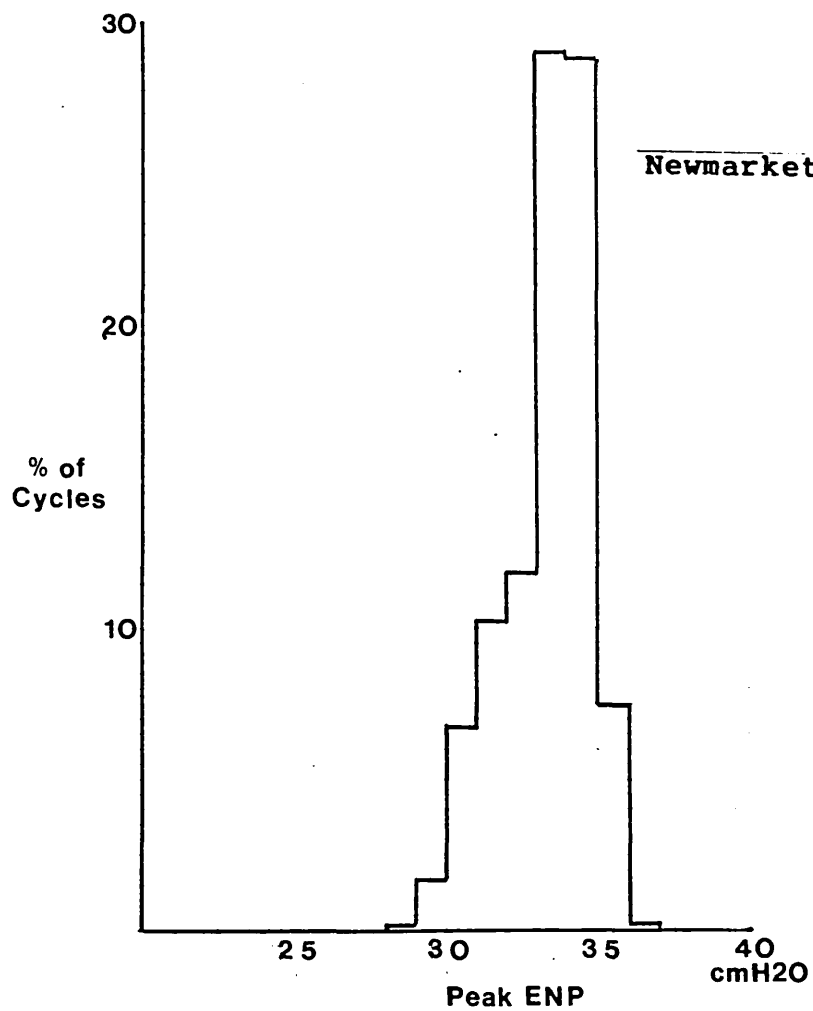


FIGURE 13

Percentage of Pump Cycles Reaching Different Peak External
Negative Pressures (Peak ENP) with the Newmarket Cuirass Pump



11:5:5 Patient-triggered cuirass-assisted ventilation

Triggering of the pump external negative pressure cycle was easily achieved with the Respitrace and with nasal thermistors to detect the onset of spontaneous inspiration. Contamination of surface diaphragmatic electromyograms by electrocardiographic artifact made this signal unsuitable as a trigger for the pump. In two of eight patients studied, accessory muscle activity persisted during cuirass-assisted ventilation so that integrated sternomastoid electromyograms could be used as a signal to trigger the pump. On account of the damping demonstrated in the earlier sections of this chapter, the delay from the time the trigger threshold was reached until the peak of the external negative pressure cycle was approximately one second, even when the half sinewave was replaced by a square wave. The peak of spontaneous inspiratory muscle activity only coincided with the peak of the external negative pressure cycle when sternomastoid electromyograms were used as the trigger signal. Chest wall motion and nasal airflow could be detected shortly after the onset of inspiratory muscle activity, but this delay was sufficient to prevent complete synchronisation of the spontaneous and external negative pressure cycles.

In the two subjects in whom electromyographic triggering was successful (WB and DH), ear oxygen saturation was 15% and 5% higher respectively during

triggered cuirass-assisted ventilation compared to untriggered cuirass-assisted ventilation. When spontaneous respiratory muscle activity ceased and the pump reverted to the backup rate of twelve cycles per minute, ear oxygen saturation fell to the same extent as when the pump was run without the triggering programme.

11:6 DISCUSSION

The Newmarket pump servo mechanism is capable of generating a square pressure waveform when there is little air leak from the cuirass model. When the leakage of air from the model is in the range recorded in the patients during cuirass-assisted ventilation, the upstroke of the pressure waveform is slurred. The high leakage flow rates during cuirass-assisted ventilation therefore limit the extent to which the pressure waveform can be modified.

These high flow rates are associated with a small drop in pressure down the connecting tubing. This pressure drop can be minimised by using as short a length of connecting tubing as possible, with an internal diameter of at least 5 cm as previously recommended by the Council on Physical Medicine (1947) and Scales et al (1953). Sensing pressure inside the cuirass shell corrects for this pressure drop, but is probably unnecessary in the routine clinical use of cuirass-assisted ventilation unless close control of peak external negative pressure is required.

The servo mechanism of the Newmarket pump is effective in preventing suction pressures greatly in excess of the initial setting being generated. This is seen in the experimental model and confirmed in Figure 13 by the small percentage of peak external negative pressures greater than 35 cmH₂O. The Cape pump produces greater peak suction pressures with closure of leaks in the experimental model and in clinical use more time is spent at peak external negative pressures greater than 35 cmH₂O.

The difference in ability to cope with increasing leakage is less marked between the two pumps in the experimental model. Comparison of Figures 13 and 14 reveals that the Newmarket pump maintains the peak external negative pressure slightly closer to 35 cmH₂O. The better compensation for decreased leakage compared to increased leakage is reflected in the asymmetry of the distribution of peak external negative pressure in Figure 41. With both pumps 99% of peak external negative pressures were within the range 25 to 40cm of water, and the clinical importance of a narrower distribution of peak suction pressure within this range is likely to be slight.

Stephenson et al (1961) reported the use of inspiratory muscle electromyograms to trigger cuirass-assisted ventilation in patients with poliomyelitis, using the intercostal, sternomastoid,

platysma, pectoralis major or temporalis muscles. The system was used successfully in seventeen subjects for a total of 900 patient hours, principally in the subjects' own homes.

The system used in the studies described in this chapter was less successful than Stephenson's in that constant supervision was required to adjust the electromyograph electrodes because of intermittent excessive electrical interference or loss of skin contact. Marks et al (1963) also found that supervision was necessary when using nasal airway pressure to trigger external negative pressure. A more robust triggering system could be of value in overcoming problems of incoordination between the patient's own respiratory efforts and the external negative pressure cycle. Chihara et al (1988) have recently reported the use of nasal thermistors to trigger cuirass-assisted ventilation, but the ability of this system to improve coordination during sleep has not yet been described.

CHAPTER 12

CONCLUSIONS

Respiratory failure is a well recognised complication of diseases affecting the chest wall. The pathophysiological changes which lead to respiratory failure in these patients are increasingly well documented, particularly with recent interest in gas exchange during sleep. Much is still unknown about the effects of assisted ventilation on patients with chest wall disease. Previous studies have often included different methods of ventilatory assistance, the numbers of subjects has sometimes been small or the duration of the study short, and frequently there is a lack of physiological data.

In this thesis, daytime studies using individually moulded cuirass respirators showed that cuirass-assisted ventilation could increase ventilation and improve arterial blood gases in patients with chest wall disease. Tidal volume was proportional to the peak external negative pressure used, but in some patients increasing the peak external negative pressure above 20 cmH₂O reduced tidal volume. Although there was little difference between the two respirators in most patients, jacket respirators produced larger tidal volumes than cuirass respirators at the same peak external negative pressure in some patients. Both of these observations suggest that cuirass-assisted ventilation may restrict

expansion, and in each individual patient the peak external negative pressure which is used in the long term should be assessed by measurement of the tidal volumes achieved with different peak external negative pressures. Asynchronous movement of the ribcage and abdomen was corrected by cuirass-assisted ventilation in five out of six patients, but paradoxical inward inspiratory motion of the ribcage was induced in two patients. Increasing the respiratory rate resulted in a larger minute volume except in patients with airflow limitation, but external positive pressure proved to be of little clinical value.

The studies during sleep did not involve sleep staging, but transcutaneous carbon dioxide tensions were reduced by cuirass-assisted ventilation, suggesting an improvement in ventilation. Some patients continued to have episodes of hypoxia during sleep despite cuirass-assisted ventilation. Where these hypoxic episodes reflect inability of cuirass-assisted ventilation to maintain ventilation without some synchronous inspiratory muscle contraction, it may be appropriate to try a respirator which applies external negative pressure to a larger area of the chest wall, such as a tank respirator or one of the larger jacket respirators. Where the hypoxic episodes persisting during cuirass-assisted ventilation are the result of upper airway obstruction, more effective assisted

ventilation may be achieved by the use of intermittent positive airway pressure, for instance through a nasal mask.

Twenty one of the twenty five patients with chest wall disease commenced on nocturnal cuirass-assisted ventilation at home were alive after one year. Daytime arterial blood gases had improved, but the mechanism of this improvement remained unclear. Although daytime studies showed that cuirass-assisted ventilation was able to rest the respiratory muscles, the reduction in PaCO_2 after one year could not be explained by an increase in respiratory muscle strength. Lung volumes increased after one year of nocturnal cuirass-assisted ventilation, suggesting that the mechanical properties of the lungs and chest wall had improved. The reduction in PaCO_2 may also have been the result of greater respiratory centre sensitivity to carbon dioxide. Although no change in respiratory drive was seen in the present study, this may have been the result of the small number of subjects in whom it was measured and the insensitivity of the technique. Cardiac output was not affected by cuirass-assisted ventilation in short term studies, but after one year of nocturnal cuirass-assisted ventilation, clinical scores of right heart had improved. This could be a consequence of the improvement in daytime arterial blood gases or have been the result of correction of sleep hypoxia.

The one year survival is comparable with previous studies of nocturnal assisted ventilation in similar patients. Two of the four deaths occurred in subjects with progressive neuromuscular disease. The death of one other subject could probably have been avoided if a different method of assisted ventilation had been used, but she had been unwilling to consider this. Little impact was made on breathlessness in the surviving patients, despite improved arterial blood gases and lung volumes.

Although one year of cuirass-assisted ventilation was not expensive, the major contribution to the expense was the cost of nursing care on the Assisted Ventilation Unit. Some of the routine assessment admissions did not lead to any change in therapy, adjustment of the cuirass or alteration of the pump setting. A reduction in the cost per patient could have been achieved by substituting a home visit for some of these hospital admissions.

Studies of a new fan pump showed that further improvements of the servo system are needed to allow the pump to compensate for increasing air leakage around the rim of the cuirass shell. Although patient-triggered cuirass-assisted ventilation is possible with this pump, an effective triggering system for use at home has yet to be designed. Further research in the design of the cuirass pump should probably be directed towards reduction in the cost and weight of the present unit.

A number of factors are likely to lead to an increase in the number of patients requiring nocturnal assisted ventilation over the next few years. It is becoming apparent that patients who had had a thoracoplasty performed in the 1950's are now presenting with respiratory failure as a late complication. The numbers of such patients is likely to increase over the next decade or so. Increasing numbers of patients who had poliomyelitis in the middle of this century are also likely to present with late respiratory complications. Awareness amongst physicians of the possibility of disordered gas exchange during sleep and of the benefits of nocturnal assisted ventilation will result in referral of many of these patients, and increasing numbers of those with other diseases of the chest wall, to centres offering facilities for assisted ventilation. The effects of cuirass-assisted ventilation described in this thesis should help identify which of these patients will benefit from cuirass-assisted ventilation, and suggest that the facility for setting up cuirass-assisted ventilation at home may need to be more widely available over the next few years.

APPENDIX 1Individual Case Summaries

- 1 SS Congenital idiopathic scoliosis. Respiratory symptoms for four years prior to commencing cuirass-assisted ventilation. Non-smoker.
- 2 HH Congenital idiopathic scoliosis. Respiratory symptoms for two years prior to commencing cuirass-assisted ventilation. Ex-smoker.
- 3 AP Congenital idiopathic scoliosis. Presented with acute respiratory failure. Smoker.
- 4 MB Idiopathic scoliosis, first detected at age of twelve years. Presented with acute respiratory failure. Four previous episodes of acute respiratory failure, one necessitating intermittent positive pressure ventilation in 1980. Non-smoker.
- 5 SG Congenital scoliosis associated with neurofibromatosis. Bilateral foot amputations for club foot. Spina bifida occulta. Presented in acute respiratory failure. Five previous episodes of acute respiratory failure, but never required assisted ventilation. Ex-smoker.
- 6 BC Congenital idiopathic scoliosis. Respiratory symptoms for five to ten years. Ex-smoker.
- 7 WW Thoracic kyphosis following childhood spinal tuberculosis. Respiratory symptoms for three years prior to commencing cuirass-assisted ventilation. Domiciliary oxygen for one year. Presented in acute respiratory failure. Ex-smoker.
- 8 BS Poliomyelitis aged 3 years, but without respiratory problems during the initial illness. Residual limb muscle weakness, but ambulant and fully independent. One year history of respiratory symptoms prior to referral. Non-smoker.
- 9 AV Poliomyelitis aged 3 years, but without respiratory problems during the initial illness. Residual limb muscle weakness limiting walking to short distances around the house with the aid of a brace. Needed intermittent positive pressure ventilation following hysterectomy in 1979. One year history of respiratory symptoms. Non-smoker.
- 10 YH Poliomyelitis in infancy, but without respiratory problems during the initial illness. Confined to a wheelchair on account of limb muscle weakness. Presented with acute respiratory failure. Non-smoker.

- 11 MS Scoliosis detected first in adolescence, thought initially to be idiopathic. Spinal fusion performed at age of 14 years. Presented with one year history of respiratory symptoms. Proximal limb muscle weakness developed during year of cuirass-assisted ventilation. Muscle biopsy evidence of unclassified myopathy. Non smoker.
- 12 TC Scoliosis first detected at age of 13 years. Harrington rod insertion at age of 15 years. Right heart failure and pulmonary hypertension developed in adolescence. Noted to have proximal muscle weakness at the time of referral to the Assisted Ventilation Unit. Muscle biopsy showed a mitochondrial myopathy. Occasional smoker.
- 13 GP Duchenne type muscular dystrophy, with scoliosis and limb muscle weakness. Acute respiratory failure in 1982, with progressive respiratory symptoms since then. Non-smoker.
- 14 RR Unclassified myopathy first detected in infancy. Severe limb muscle weakness confining him to a wheelchair. Spinal surgery aged 9 years. Oesophageal stricture following prolonged nasogastric intubation. Intermittent oesophageal dilatation performed for dysphagia. Acute respiratory failure one year prior to commencing cuirass-assisted ventilation. Non-smoker.
- 15 JG Familial limb girdle type dystrophy, with first onset of weakness in fourth decade. Able to walk only a few steps unaided. Non-smoker.
- 16 RB Unclassified proximal myopathy first noticed at age of thirty. Presented with acute respiratory failure necessitating intermittent positive pressure ventilation, from which he could not subsequently be weaned. Non-smoker.
- 17 JL Presented with confusion. Subsequently found to be hypoxic and hypercapnic and required urgent intermittent positive pressure ventilation. Weaned to nocturnal intermittent positive pressure ventilation and referred for external negative pressure ventilation. Limb muscle fasciculation noted. Electromyography and muscle biopsy evidence of denervation. Ex-smoker.
- 18 WB Left sided 7 rib thoracoplasty performed in 1951, preceded by phrenic nerve crush and artificial pneumothorax. Onset of respiratory symptoms in 1967, since when he had used domiciliary oxygen. Acute respiratory failure in 1975. Ex-smoker.
- 19 MH Left sided 6 rib thoracoplasty performed in 1950's. Presented with six month history of respiratory symptoms. Non-smoker.

- 20 DH Left sided 8 rib thoracoplasty performed in 1952. Non-smoker. Presented with acute respiratory failure.
- 21 IJ Right sided 9 rib thoracoplasty performed in 1950's, preceded by phrenic nerve crush and artificial pneumothorax. Non-smoker.
- 22 NW Left sided 8 rib thoracoplasty performed in 1949, preceded by phrenic nerve crush. Two year history of respiratory symptoms. Non-smoker.
- 23 HC Right sided 6 rib thoracoplasty performed in 1950's. Presented in acute respiratory failure. Non-smoker.
- 24 KN Bileteral thoracoplasty performed in 1950's, with total of seven ribs removed. Presented in acute respiratory failure. Non-smoker.
- 25 DS Right sided 8 rib thoracoplasty performed in 1958. respiratory symptoms for two years prior to commencing cuirass-assisted ventilation.

APPENDIX 2Depression Questionnaire

- 0 I do not feel sad
 - 1 I feel sad
 - 2 I am sad all the time and I can't snap out of it
 - 3 I am so sad or unhappy that I can't stand it
-
- 0 I am not particularly pessimistic or discouraged about the future
 - 1 I feel discouraged about the future
 - 2 I feel I have nothing to look forward to
 - 3 I feel that the future is hopeless and that things cannot improve
-
- 0 I do not feel like a failure
 - 1 I feel I have failed more than the average person
 - 2 As I look back on my life, all I can see is a lot of failures
 - 3 I feel I am a complete failure as a person
-
- 0 I am not particularly dissatisfied
 - 1 I don't enjoy things the way I used to
 - 2 I don't get satisfaction out of anything anymore
 - 3 I am dissatisfied with everything
-
- 0 I don't feel particularly guilty
 - 1 I feel bad or unworthy a good part of the time
 - 2 I feel guilty
 - 3 I feel as though I am very bad or worthless
-
- 0 I don't feel disappointed in myself
 - 1 I am disappointed in myself
 - 2 I am disgusted with myself
 - 3 I hate myself
-
- 0 I don't have any thoughts of harming myself
 - 1 I feel I would be better off dead
 - 2 I have definite plans about committing suicide
 - 3 I would kill myself if I had the chance
-
- 0 I have not lost my interest in other people
 - 1 I am less interested in other people than I used to be
 - 2 I have lost most of my interest in other people and have little feeling for them
 - 3 I have lost all my interest in other people and don't care about them at all
-
- 0 I make decisions about as well as ever
 - 1 I try to put off making decisions
 - 2 I have great difficulty in making decisions
 - 3 I can't make any decisions at all anymore

- 0 I don't feel I look any worse than I used to
 - 1 I am worried that I am looking old or unattractive
 - 2 I feel that there are permanent changes in my appearance and that they make me look unattractive
 - 3 I feel that I am ugly or repulsive looking
-
- 0 I can work about as well as I used to
 - 1 It takes extra effort to get started at doing something
 - 2 I have to push myself very hard to do anything
 - 3 I can't do any work at all
-
- 0 I don't get any more tired than usual
 - 1 I get tired more easily than I used to
 - 2 I get tired from doing anything
 - 3 I get too tired to do anything
-
- 0 My appetite is no worse than usual
 - 1 My appetitie is not so good as it used to be
 - 2 My appetite is musch worse now
 - 3 I have no appetite at all any more

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