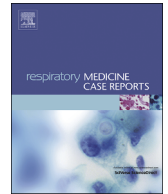




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Case Report

Inspiratory muscle training for diaphragmatic dysfunction: A case series

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ABSTRACT

Inspiratory muscle training (IMT) is used across various pathology domains to improve respiratory function. Limited literature exists which demonstrates IMT benefit among patients with Diaphragmatic dysfunction. 7 individuals with a mean age of 59.6yrs had unilateral diaphragmatic dysfunction (UDD) post cardiac surgery and were referred to a cardiac rehab program where an IMT strength based protocol was prescribed. IMT implementation over an average of 13 weeks yielded an average improvement in maximum inspiratory pressure (MIP) of 48 % (p value 0.018), peak inspiratory flow rate (PIFR) of 45 % (p value 0.018), forced expired volume in 1 sec (FEV1) of 15 % (p value 0.028) and forced vital capacity (FVC) of 15 % (p value 0.018). This case series of data adds to the limited evidence that exists currently and outlines the benefits of IMT application within unilateral diaphragmatic dysfunction.

1. Introduction

The diaphragm is a critical component of the respiratory pump and contributes to 30–60 % of the total minute ventilation (TMV) [1]. However, clinical presentations of diaphragm dysfunction are often non-specific and can be mistaken for other more common causes of dyspnoea [2]. This unexplained dyspnoea from diaphragmatic dysfunction can be assumed in review of a restrictive respiratory pattern on pulmonary function test (PFT), reduction in FVC when PFT performed in supine position, abnormal diaphragm position on plain chest imaging, reduced exercise capacity and reduced MIP in reference to predicted value [3,2]. Clare Laroche et al. concluded that 9 of the 11 patients in their study had a recent onset of hemidiaphragm paralysis or dysfunction that was associated with a reduction in maximal transdiaphragmatic pressure, leading to reduced inspiratory capacity, increase in breathlessness and decrease in exercise tolerance associated with the condition and not explained by a deterioration in other pathological conditions [3].

With unilateral diaphragmatic paralysis, vital capacity (VC) and maximum voluntary ventilation (MVV) decreases by 20–30 %, with 20 % reduction in oxygen uptake on the affected side [1]. The severity of diaphragm dysfunction can vary from partial loss of force generation (weakness) to complete loss of function (paralysis) and can be either unilateral or bilateral (involving both hemidiaphragms) [2]. Unilateral diaphragm dysfunction is usually tolerated because of compensatory mechanisms, such as increased motor output to accessory breathing muscles and normal hemidiaphragm [2]. The paralysis group presented in Mayra Pereira et al. (2021)

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study outlines an increased inspiratory muscle effort combined with reduced tidal volume, increased respiratory rate (RR) at a given work rate or ventilation compared to the controls, and reduced exercise performance and higher dyspnoea [4]. EMG data obtained within this study showed a greater percentage of accessory inspiratory muscle activation of scalenes and sternocleidomastoid and at peak exercise, paralysis group reached twice the percentage of accessory inspiratory muscle activation than that of the controls [4]. Mayra Pereira's 2021 findings align with early publication of single case study where phrenic nerve conduction tests did not show evidence of improvements in any of recorded measures, suggesting that improvements in inspiratory muscle function were in non-diaphragmatic inspiratory muscles, than related to improvements in diaphragm contractility [5]. A combination of the hypertrophy of contralateral hemidiaphragm, increased stiffness of paralysed side, and increased accessory inspiratory muscle activation are thought to be contributors to the improvements seen from IMT [1,3–5]. Michele Schaeffer 2023 study also confirm that meaningful reductions in dyspnoea and improvement in exercise tolerance in people with unilateral diaphragm dysfunction is likely via improvements in strength, coordination and/or oxygenation of the extra – diaphragmatic respiratory muscles [6].

Our cases series demonstrates the outcomes of inspiratory muscle training (IMT) within a series of patients with unilateral diaphragm dysfunction, utilising a pressure threshold device. This case series contributes to the literature regarding IMT's effect on expiratory and inspiratory parameters.

2. Methods

All client testing and outcome measures were collected at single site, Robina health Precinct by the same clinician. The data from seven clients was collated to form the case series. A respiratory, cardiothoracic, or cardiology specialist provided a referral for the client to complete inspiratory muscle training. Screening and clearance for treatment came from the treating medical team. As this treatment is usual care for clients identified with unilateral diaphragmatic dysfunction/phrenic nerve palsy, assessment and treatment with IMT was offered. Inform consent was obtained to participate in the training program, and regular follow up over 12 weeks.

Relative and absolute contra-indications for spirometry and inspiratory muscle testing were assessed prior initial assessment. Worsening of inspiratory/expiratory measures with training or increased shortness of breath with training indicated to withhold or cease IMT, none of the patients undergoing testing or training obtained any adverse symptoms. Oxygen saturation and heart rate (HR) was assessed to determine adverse response to testing and training. The risk of diaphragmatic cramp was mitigated by explanation of IMT protocol and conservative exercise prescription initially. The first two weeks of training was performed at 15 % MIP to familiarise client to training and condition diaphragm for later higher loads (30–50%MIP). The client had to purchase the inspiratory pressure threshold device (Powerbreathe Classic), though this didn't present an issue for any of the client's partaking in IMT.

Supervising clinician used his professional judgement in review of absolute and relative contra-indications and experience to guide the IMT course of treatment.

No control within this study existed as all clients referred for IMT was offered it as usual care. The pre-post change in the outcome measures was calculated using the preferred Wilcoxon Signed Ranks Test (since the sample size was small).

Given the paucity of literature in this clinical population, the research team are keen to provide preliminary evidence of the potential efficacy of inspiratory muscle training in patients undergoing rehabilitation following a cardiac-related procedure requiring sternotomy. Thus, this project is intended as a pilot study to provide “proof-of-concept” data that will subsequently inform a larger, controlled program of research.

Patients were referred to a cardiac rehabilitation program and received usual care cardiac rehab, inclusive of IMT, IMT specifically addressing diaphragmatic dysfunction. These patients reflect the entire cohort who have completed the IMT protocol, to date, and do not reflect a selective inclusion based on positive outcomes.

The Powerbreathe K5 breathelink device and software were utilized for testing of MIP and PIFR and used for familiarizing client to training method. Training functionality of software provided average power, inspiratory flow rate and inspiratory pressures. The powerbreathe K5 device and software was approved on Australian register of therapeutic goods (ARTG) with the identifier 294782 and was used for testing MIP and PIFR. The ‘EasyOne Pro – Pulmonary Function analyser, adult’ was used for testing FEV1, FVC and FEV1/FVC ratio. Interpretation of inspiratory and expiratory outcome provided evidence of whether respiratory pattern was restrictive and/or obstructive. The EasyOne was approved by ARTG with the identifier of 131960. All clients used the *powerbreathe classic* pressure threshold device to complete training protocol. The prescribed resistance was graduated as per [Table 1](#) protocol. For patients post median sternotomy IMT was not commenced until after 6 weeks post procedure and/or when patient was painfree (see [Table 2](#)).

Research was approved by the Gold Coast Hospital Health Service Ethics committee.

3. Results

Client's age ranged from 39 to 71 years of age with a mean of 59.6 years and median of 64years. Data represented 3 females and 4 males. Median time of follow up ranged from 7 weeks to 22weeks, with a median of 12 weeks and mean of 13wks, longer follow up period was associated with a longer timeline to achieve inspiratory measure threshold and shorter period associated with training ceasing due to one client receiving a pulmonary hypertension diagnosis with respiratory specialist recommendation to cease IMT. Clients had an average FEV1 % predicted of 62.7 % at baseline which increased to 70.3 % predicted with training. Clients had an average FVC % predicted of 63.3 % at baseline which increased to 70.3 % predicted with training.

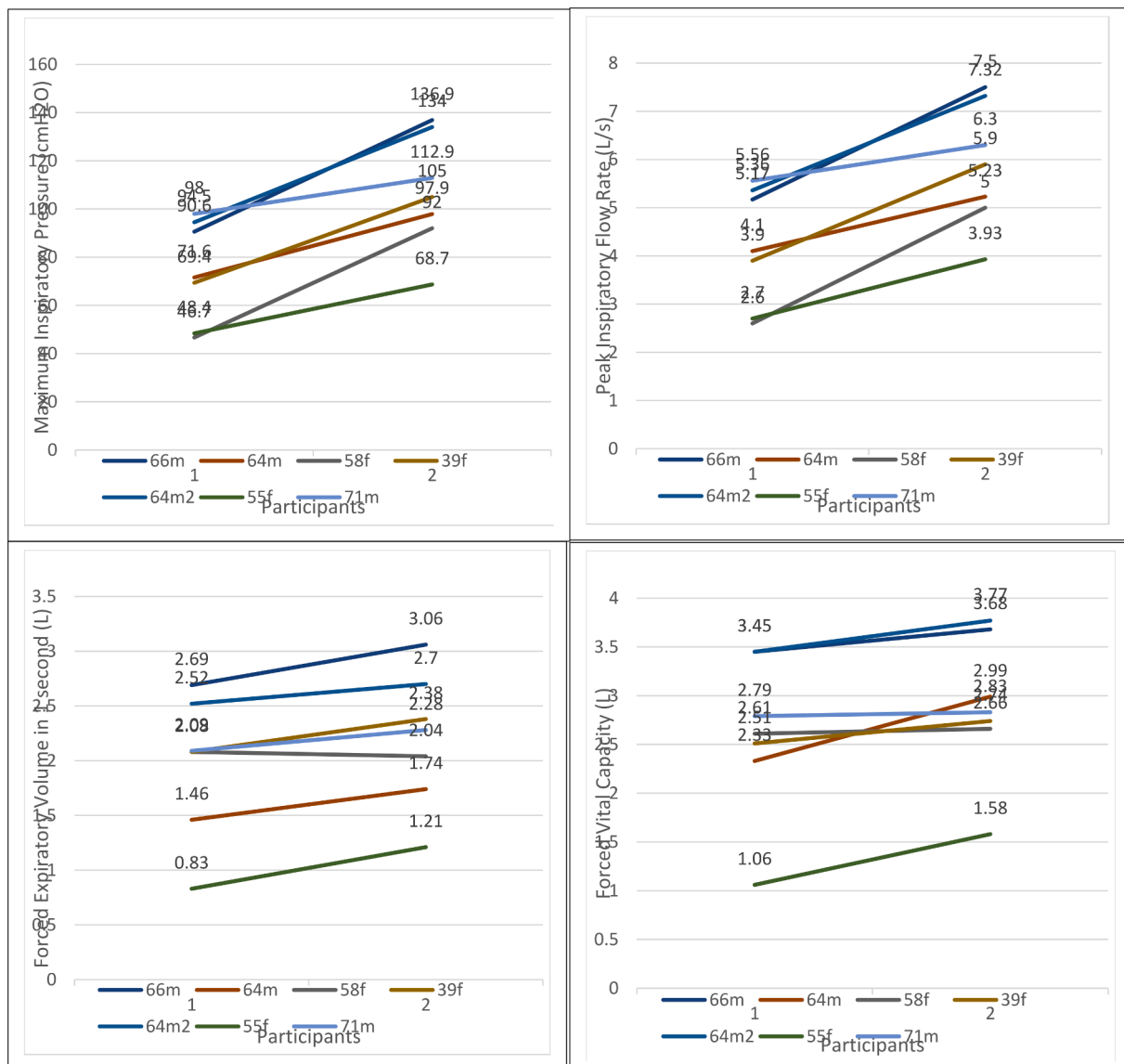
IMT implementation over an average of 13 weeks yielded an average improvement in MIP of 48 % (p value 0.018), PIFR of 45 % (p value 0.018), FEV1 of 15 % (p value 0.028) and FVC of 15 % (p value 0.018).

Table 1
IMT protocol.

Consult No.	Description of Consult
1	<ul style="list-style-type: none"> • Test expiratory and inspiratory measures Habituation = first two weeks Prescribe load at 15 % MIP first week and if client tolerating 30 breathes with ease progress load by 5 % as tolerated between 15 and 30%MIP, 30 breaths, twice a day, 5 days/wk
2	<ul style="list-style-type: none"> • 2 weeks after training commenced -retest inspiratory measures Next 4 weeks Strength building (Aim ↑ MIP by >20 %) = 2–6 wks Aiming for graduated progression 30–50%MIP, progressing load only if client completing 30 breathes continuous, 30 breaths, twice a day, 5 days/wk
3	<ul style="list-style-type: none"> • 6 weeks after training commenced – retest inspiratory measures and if significant improvement in values observed consider re-ax expiratory values with spirometry <ul style="list-style-type: none"> • Alter Load between 30 and 50%MIP, 30 breaths, twice a day, 5 days/wk
4	<ul style="list-style-type: none"> • 10–12weeks after training commenced Assess for Inspiratory and expiratory improvement plateau • If plateau in improvement observed transition to Maintenance phase = Indefinitely or until asymptomatic or return to usual exercise tolerance • If MIP and PIFR still improving maintain training up to max of 50 % MIP until plateau in values achieved
Maintenance	<ul style="list-style-type: none"> • Maintenance phase Load = 30–50%MIP, 30 breaths, twice a day, 2 days/wk

Table 2
Outcome measure analysis.

Variable	Pre-Value mean	Post -Value mean	% Change	P - value
MIP (cmH20)	74.17	106.77	48	0.018
PIFR (L/s)	4.2	5.88	45	0.018
FEV1 (L)	1.96	2.2	15	0.028
FVC	2.6	2.89	15	0.018



4. Discussion

We showed that IMT in people with UDD improves inspiratory and expiratory measures. Our cohort at baseline showed an average FVC of 63.3 % predicted, which aligns with other literature observing a 35 % reduction in FVC [4], FVC of 60%predicted [5] and FVC 72 ± 7 % predicted in IMT group and FVC 65 ± 9 % predicted in control group [6]. The baseline FEV1 of 62.7 % predicted within our cohort also aligns to observations in another study of 66 ± 5 % predicted in IMT group and 58 ± 8 in control group [6]. Our baseline characteristics although small sample size represents the effect of UDD on expiratory measures outlined within other studies.

IMT implementation over an average of 13 weeks yielded an average improvement in MIP of 48 % (p value 0.018), PIFR of 45 % (p value 0.018), FEV1 of 15 % (p value 0.028) and FVC of 15 % (p value 0.018). The absence of a control group is a limitation of our data, although the improvement observed within our participants is supported by Kodric et al. [1]. Kodric et al. demonstrated that 41.7 % of IMT group (36 people) experienced a partial improvement and 36.1 % experienced a complete improvement compared to 87.5 % of control group (16 people) having no improvement and 12.5 % a partial improvement. MIP significantly improved from baseline in the active treated group (P < 0.001, but it failed in the controls (p = 0.120), showing that IMT with a threshold device increases diaphragm mobility and improves inspiratory muscle strength [1]. Shaeffer, M et al. also demonstrating that MIP was higher post – training in the IMT group (p = 0.01), but not the control group, and the pre to post change in MIP was also different between groups (p = 0.001) [6].

Our study participants all obtained ventilatory muscle weakness and UDD secondary to phrenic nerve injury post surgery. In the absence of Inspiratory muscle strength improvement and continued significant breathlessness burden, surgical plication is an option the treating medical team can consider. Surgical plication places the paralysed diaphragm in a position of maximum inspiration with underlying lung parenchyma expansion. Patient selection is critical given reported post-operative complication rates as high as 25 % [2] Metka Kodric et al. [1] estimate that the need for surgical diaphragm plication could be decreased to no more than 5 % of the total cases with IMT. None of our study participants required surgical diaphragm plication post IMT intervention.

5. Conclusion

A strength based protocol of IMT demonstrates improvements in inspiratory outcomes measures and expiratory volumes. The findings within our study need to be interpreted cautiously due to small sample size, and lack of control however the positive outcomes of IMT support IMT as a treatment modality for patients with diaphragmatic dysfunction post sternotomy and this is supported by existing literature.

CRedit authorship contribution statement

Grant Turner: Conceptualization, Project administration, Writing – original draft, Writing – review & editing. **Surendran Sabapathy:** Writing – review & editing. **Myles Moore:** Writing – review & editing. **Sylvio Provenzano:** Writing – review & editing. **Andrie Stroebel:** Writing – review & editing. **Lawrence P. Cahalin:** Conceptualization, Formal analysis, Writing – review & editing.

Declaration of competing interest

No conflicts of interest.

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