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tALk2care: improving the experience and quality of life of patients with respiratory disease treated with oxygen therapy through relationship-centered care

tALk2care: mejorar la experiencia y la calidad de vida de los pacientes con enfermedades respiratorias tratados con oxigenoterapia a través de una atención centrada en las relaciones

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Abstract

Background: Communication is important for a good care experience. **Objective:** To know the benefits of 'tALk2care' training program for professionals on the patient's experience with oxygen therapy treatment at home. **Material and methods:** A prospective observational study was performed at the La Princesa University Hospital. 6 oxygen therapy technicians participated in the study. 3 technicians were trained by a psychologist with the 'tALk2care'. 169 respiratory patients with a new oxygen therapy prescription were treated by the technicians, being assigned to 2 groups: control or 'tALk2care'. The interventions related to oxygen therapy were the same in both groups, with communication training for technicians being the intervention of the 'tALk2care' group. The outcomes were experience, quality of life, motivation, perception of therapy, dyspnea and adherence. The outcomes for the technicians were self-efficacy in communication and the risk of burn-out. **Results:** After 6 months, 'tALk2care' group patients had a better relational experience ($F[1] = 10.572$; $p < 0.001$; $\eta^2 = 0.065$), global experience ($F[1] = 7.356$; $p < 0.007$; $\eta^2 = 0.046$), perception of therapy ($F[1] = 4.981$; $p = 0.027$; $\eta^2 = 0.032$), motivation ($F[1] = 158.942$; $p < 0.001$; $\eta^2 = 0.112$) and quality of life ($F[1] = 7.475$; $p = 0.007$; $\eta^2 = 0.046$). 'tALk2care' technicians presented greater self-perception in communication and personal-accomplishment and less emotional exhaustion and depersonalization. **Conclusion:** 'tALk2care' improves the communication skills of technicians and improves experience, and quality of life of patients who are cared for by these technicians.

Keywords: Oxygen therapy. Experience. Adherence. Quality of life. Outcomes. Communication.

Resumen

Antecedentes: La comunicación es importante para una buena experiencia de atención. **Objetivo:** Conocer los beneficios de la formación 'tALk2care' para profesionales en la experiencia del paciente con tratamiento de oxigenoterapia en casa. **Material y métodos:** Se realizó un estudio observacional prospectivo en el Hospital Universitario de La Princesa. En el estudio participaron 6 técnicos de oxigenoterapia. 3 técnicos fueron formados por un psicólogo con el 'tALk2care'. 169 pacientes respiratorios con nueva prescripción de oxigenoterapia fueron atendidos por los técnicos, siendo asignados a 2 grupos: control o 'tALk2care'. Las intervenciones relacionadas con la oxigenoterapia fueron las mismas en ambos grupos, siendo la capacitación

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en comunicación para técnicos la intervención del grupo 'tALk2care'. Los resultados fueron experiencia, calidad de vida, motivación, percepción de la terapia, disnea y adherencia. Los resultados para los técnicos fueron la autoeficacia en la comunicación y el riesgo de agotamiento. **Resultados:** Después de 6 meses, los pacientes del grupo 'tALk2care' tuvieron una mejor experiencia relacional ($F[1] = 10.572$; $p < 0.001$; $\eta^2 = 0.065$), experiencia global ($F[1] = 7.356$; $p < 0.007$; $\eta^2 = 0.046$), percepción de la terapia ($F[1] = 4.981$; $p = 0.027$; $\eta^2 = 0.032$), motivación ($F[1] = 158.942$; $p < 0.001$; $\eta^2 = 0.112$) y calidad de vida ($F[1] = 7.475$; $p = 0.007$; $\eta^2 = 0.046$). Los técnicos de 'tALk2care' presentaron mayor autopercepción en comunicación y realización personal y menor agotamiento emocional y despersonalización. **Conclusión:** 'tALk2care' mejora las habilidades comunicativas de los técnicos y mejora la experiencia y calidad de vida de los pacientes atendidos por estos técnicos.

Palabras clave: Oxigenoterapia. Experiencia. Adherencia. Calidad de vida. Resultados. Comunicación.

Introduction

Lung diseases have become one of the most important causes of illness and death worldwide¹. Chronic obstructive pulmonary disease (COPD) is one of the most common chronic lung diseases. Furthermore, according to the Global Chronic Obstructive Lung Disease Initiative (GOLD), this disease is the third most widespread cause of death worldwide². Interstitial lung disease (ILD) is a collective term representing a diverse group of pulmonary fibrotic and inflammatory conditions. Idiopathic Pulmonary Fibrosis (IPF) is one of these diseases. The prevalence of IPF is currently 1,650 per 100,000 people³.

These diseases are progressive, and weaken the affected person. This progression of lung dysfunction and worsening of respiratory symptoms causes patients to experience a gradual deterioration in their health over the years. Furthermore, their social life, physical performance, and daily activities are frequently disrupted and they may even begin to depend on others, especially their family members, to meet their own personal needs⁴. As a result, of all these problems, patients are deeply affected on a psychological level (fear, depression, isolation, anxiety...). Added to this is the loss of their social roles⁵. Oxygen therapy offers significant short- and long-term benefits in those with COPD. Immediate benefits include alleviation of hypoxemia and its sequelae, improvement in exercise capacity, reduction of dyspnea, and possibly sleep consolidation⁵. However, despite this benefit, there are still problems in achieving optimal adhesion⁶. Adherence to LTOT (long-term oxygen therapy) ranges from 45% to 70% and utilization for more than 15 hours per day is widely accepted as effective. Although several studies have addressed the level of patients' adherence to LTOT, few have suggested or evaluated interventions

that conducted to compliance enhancement⁷. The existence of erroneous beliefs about oxygen therapy seems to be one of the factors that directly influence adherence⁸. The perception of oxygen therapy by patients⁹ influences adherence. Some patients feel safe, the therapy allows them to have a better quality of life. However, a large number of patients express a negative attitude: fear, vision of oxygen versus self, restriction and shame. There appears to be uncertainty among patients regarding the purpose and benefits of oxygen therapy, although an underlying trust in health professionals and technicians attending the home is evident⁹. This trust appears to foster acceptance of a life-changing therapy, despite the impact, burden, and incomplete understanding of its benefits.

Patients see in all the professionals who care for them a source of information, and also a source of care to reduce their discomfort. From the doctor to the nurse, to the technician and even the person who assists from the contact center: they all have an important role because they are part of the patient's care plan.

Communication is the key. Research has shown that the empathy and communication skills of professionals who interact with patients, including when delivering bad news, can be improved through well-designed training programs^{10,11}. Conversations between caregivers and patients require a structured, relationship-centered approach, in which patients not only receive information with compassion and kindness, but also have their most important concerns addressed^{12,13-15}. Patient-reported outcome measures such as questionnaires that assess the impact of good communication and quality of life improve with good communication¹⁶. But improved communication must be implemented properly¹⁷. It has been shown that a simple help tool to address issues related to

Table 1. Contents of the ‘tALK2Car’ training program

Blocks	Contents	Duration
1 Know & understand our patients	Survival scheme: understand how patients process the diagnosis, what emotional expressions may occur and what coping strategies they can implement. Patient journey: know what the patient’s journey is from the moment of diagnosis to living with therapy in the long term. Perception of home respiratory therapies: know what beliefs (right and wrong) patients have about oxygen therapy.	1:45 minutes
2 Basic communication skills	Empathy: what is empathy and how to show it with tips. Strategies to show empathy when there is eye contact and when there is not (over the phone). Counseling: how to improve communication with the patient to better understand their needs. Non-verbal expressions: non-verbal language issues to take into account that can help us in healthcare activity.	1:45 minutes
3 Communication skills training	Simple communication techniques that help improve the quality of our interactions: Smile and greet Tell your name, role, and expectations Active listening and assist Rapport and relationship Say thanks...	1:30 minutes
4 Self-assessment	Survey to find out what is my way of communicating with patients	5 minutes

treatments produces an increase in the patient experience¹⁸. Tools have also been developed to measure patients’ perceptions of empathy during a consultation to provide physicians with feedback for self-assessment¹⁹. Some programs based on interventions such as motivational interviewing have been designed to improve adherence, but their scalability is complicated due to the resources required²⁰.

Relationship-centered care (RCC)²¹ is a framework for conceptualizing health care which recognizes that the nature and quality of relationships in health care influence the process and outcomes of health care. In this framework, it is where therapeutic communication becomes the most basic of tools to improve people’s quality of life. The principles of the RCC are:

- Relationships in healthcare activity must be based on authenticity: RCC emphasizes the importance of authenticity, in the sense that professionals must not simply act as if they have respect for the patient, but must show it²².
- Affection and emotion are important components of relationships in healthcare: We are not talking about psychological support, but rather about listening, interest and compassion²³.
- All care relationships occur in the context of reciprocal influence: allowing the patient to express themselves allows for improved identification of

needs and proactivity, reducing reactivity and corrective actions²⁴.

- RCC has a moral basis: as a human participant, the personal assistant behaves more genuinely than if he were playing a role. This type of honesty is morally desirable as an end in itself and allows the patient to express themselves and verbalize their needs²⁵.

RCC is related to the patient experience, understood as the sum of all the interactions that occur between the patient and the health system²⁶, and has as its components relational aspects (feeling understood, feeling listened to) and organizational aspects (being attended to when appropriate, being well organized.) There is evidence that reinforces that a good experience favors adherence to treatments, improves clinician-patient communication and improves the quality of life of patients. A Patient-Reported Experience Measures (PREM) and Patient-Reported Outcomes Measures (PROM) are measures of a patient’s perception of their personal experience of the healthcare they have received, and of their health status respectively²⁷.

On the other hand, there are studies that show that up to 47% of health professionals who care for patients with respiratory diseases present burn-out syndrome, and 30.8% express psychological distress²⁸. Some studies have shown that training in communication and

Table 2. Set of variables, measures and sources

Type of variable	Variables	Questionnaire	Measure
Socio-demographic	Age	Direct response	Number of years
Psychological	Perception about oxygen therapy	Ad hoc questionnaire with 4 response alternatives (bad perception to good perception of oxygen therapy)	Registration by categories
	Motivation	Ad hoc questionnaire with 5 response alternatives (very low motivation to very high motivation to use oxygen therapy). Based on the transtheoretical model of change ³²	Registration by categories
Clinics	Diagnosis	Direct response	Registration by categories
	Dyspnoea	mMRC (Modified Dyspnea Scale) ³³	Score from 0 to 4
Treatment	Adherence	Oxygen therapy concentrator	Number of hours of use
Quality of life	Quality of life	EuroQoL-5D-5L ³⁴	Score -1 to 1
	Experience	HowRwe ^{35,36}	Score from 1 to 10
Communication style (technicians)	Burnout	Maslach Burnout Inventssory (MBI) ³⁷	Score from 0 to 132
	Self-assessment	Communication Style Survey ad hoc (13 questions on different aspects of communication (greeting, smiling, summarizing explanations, attending...) with 5 response alternatives (from 'I do it little' to 'I always do it')	Score from 0 to 52

empathy skills is a preventive factor for Burnout syndrome in health professionals²⁹.

'tALk2care' is a communication skills training program specifically aimed at technicians and nurses who care for patients undergoing treatment with oxygen therapy or mechanical ventilation. The objective of this training is to improve professional-patient interactions, configuring a therapeutic relationship based on a good experience for the patient. The main objective of 'tALk2care' is to improve the experience of patients with respiratory disease with the prescription of oxygen therapy. The secondary objectives are to improve outcomes related to quality of life, in addition to improving the perception of oxygen therapy and motivation to use it.

This study aims to measure the impact of a communication program called 'tALk2care' in the context of home respiratory therapies on patients' quality of life and their experience. The 3 main hypotheses that were tested are: (1) 'tALk2care' training to oxygen therapy technicians improves the experience of the patient; (2) a better experience allows to improve the perception and knowledge about oxygen therapy; (3) improved perception of oxygen therapy experience implies an improvement in patient's quality of life.

Material and methods

Design

A prospective observational single-center study design was performed at La Princesa University Hospital in Madrid (Spain) between March 2023 and February 2024 in patients with respiratory diseases (COPD, Interstitial Lung Disease) with prescription for oxygen therapy (fixed concentrator with/without portable concentrator). Patients with new prescription of oxygen therapy treated at the Pulmonology Department were included. The study protocol was approved by the Clinical Research Ethics Committee of La Princesa University Hospital (Registration number: 5540).

For this study there were no changes in the care plan established by SEPAR for oxygen therapy (frequencies, interventions, procedures)³⁰. Only the evaluation of PROM and PREM questionnaires was included on the day of initiation of therapy and at 6 months.

Patients

The participants met the inclusion criteria: adult patients with prescription of oxygen therapy by the Pulmonology Department at La Princesa University Hospital. The exclusion criteria were: age under

Table 3. Baseline characteristics of the participants

Variable	Total sample (n = 169)		tALK2care group (n = 85)		Control group (n = 84)		Test	df	p
	Frequency (%)	Mean (SD)	Frequency (%)	Mean (SD)	Frequency (%)	Mean (SD)			
Sex (male)	95 (56.2)		51 (60)		44 (52.4)		0.996	1	0.318
Age	69.8 (5.18)			70.09 (5.42)		69.64 (4.95)	0.565	167	0.287
Diagnosis							0.051	1	0.821
COPD	132 (78.1)		67 (78.8)		65 (77.4)				
ILD	37 (21.9)		18 (21.2)		19 (22.6)				
Oxygen therapy prescription							0.323	1	0.570
16 hours	45 (26.6)		21 (24.7)		24 (28.6)				
24 hours	124 (73.4)		64 (75.3)		60 (71.4)				
Respiratory function									
FVC		80.34 (20.9)		81.72 (21.94)		79.07 (19.95)	0.773	148	0.220
FEV1		47.4 (21)		49.66 (20.59)		45.30 (21.46)	1.267	148	0.104
FVC/FEV1		48.63 (16.17)		51.11 (16.99)		46.33 (15.12)	1.822	148	0.070
Dyspnoea (mMRC)							3.603	4	0.462
Grade 0	1 (0.6)		0 (0)		1 (1.1)				
Grade 1	22 (13)		8 (9.4)		16 (18.4)				
Grade 2	61 (36.1)		31 (36.5)		30 (34.5)				
Grade 3	74 (43.8)		41 (48.2)		33 (37.9)				
Grade 4	11 (6.5)		5 (5.9)		7 (8)				
Quality of life (Euroqol-5D-5L)									
Mobility	2.01 (0.99)		1.95 (1.05)		2.07 (0.929)		6.490	4	0.165
Self-care	2.23 (0.73)		2.20 (0.737)		2.27 (0.734)		0.685	3	0.877
Usual activities	3 (0.89)		3.02 (0.913)		2.98 (0.878)		1.096	2	0.578
Pain/discomfort	2.02 (0.93)		1.99 (0.957)		2.06 (0.910)		2.051	4	0.726
Anxiety/depression	2.62 (1.07)		2.60 (1.08)		2.63 (1.07)		1.223	3	0.748
AVS		45.17 (18.33)		45.17 (18.99)		45.17 (17.75)	-0.001	167	0.500
EQ-5D-5L value		0.6042 (0.22)		0.6064 (0.23)		0.6019 (0.22)	0.129	167	0.449
Data from the technicians responsible for oxygen therapy									
Age		36.66 (6.02)		36.33 (4.16)		37 (8.54)	-0.121	4	0.455
Communication style		22.83 (7.08)		22 (7.93)		23.66 (7.76)	-0.260	4	0.404
Emotional exhaustion		26.50 (1.64)		25.67 (1.52)		27.33 (1.52)	-1.336	4	0.126
Depersonalization		9 (1.09)		8.67 (0.57)		9.33 (1.52)	0.507	4	0.319
Personal fulfillment		30.83 (2.22)		31.33 (3.05)		30.33 (1.52)	-0.707	4	0.259

18 years, cognitive impairment (diagnosis of dementia or severe mental disorder) to take questionnaires or refusal to answer them. Before participating in the study, all patients were informed of its objective, and signed an informed consent.

Patients included in this study correspond to the geographical area of La Princesa University Hospital (Madrid), which serves an approximate population of 323,000 people³¹, and with VitalAire (specific activity of home respiratory therapies of Air Liquide Healthcare

Table 4. Patients and technicians outcomes

Patients outcomes	'tAlk2care' group			Control group			p	Effect size
	n	Mean	SD	n	Mean	SD		
Relational experience	76	5.26	0.86	78	3.96	1.25	< 0.001	0.065
Organizational experience	76	4.28	1.46	78	4.03	1.23	0.184	0.012
Global experience	76	9.55	2.09	78	8	2.30	0.007	0.046
Perception about oxygen therapy	76	3.75	0.520	78	2.97	0.644	0.027	0.032
Motivation to use therapy	76	4.22	1.18	78	3.85	1.11	< 0.001	0.112
Quality of life – EQ-5D-5L score	76	0.76	0.21	78	0.66	0.27	0.007	0.047
Dyspnoea – mMRC	76	1.95	0.73	78	1.36	0.75	0.333	0.006
Adherence	76	20.3	5.31	78	20.4	5.12	0.838	0.033

Technicians outcomes	'tAlk2care' group					Control group				
	n	Pre		Post		n	Pre		Post	
		Mean	SD	Mean	SD		Mean	SD	Mean	SD
Self-efficacy	3	22	7.94	39.3	1.53	3	23.7	7.77	23.7	9.45
Emotional exhaustion	3	25.7	1.53	16.7	1.53	3	27.3	1.53	27.3	1.53
Depersonalization	3	8.67	0.57	5.67	0.57	3	9.33	1.53	9.33	1.15
Personal fulfilment	3	31.3	3.06	39.3	3.06	3	30.3	1.53	31.3	1.53

SD: standard deviation; AVS: analogical visual scale.

Spain) as a company providing oxygen therapy. Patients are cared for in their homes by specialized technical personnel who carry out a risk analysis (for example, smoking in at home, architectural barriers), train them in the management of the oxygen concentrator (fixed and/or portable), and review the therapy every six months. On the part of VitalAire, 6 technicians dedicated to oxygen therapy treatment agreed to participate.

Intervention

The 6 oxygen therapy specialists were randomly assigned to the two experimental groups. The 3 technicians assigned to the experimental group were trained in the "tAlk2care" program. This training took place for 6 hours, and was taught by a clinical psychologist with experience in respiratory patients. Table 1 shows the contents of this training. The contents were prepared based on scientific evidence from SEPAR, publications and studies, as well as communication techniques such as counseling and empathy training. The training consisted of theoretical explanations with

patient testimonials, and practical exercises to improve interactions with the patient. The goal was to improve qualitatively interactions between technicians and patients and caregivers.

The technicians responded to a Communication Self-efficacy survey (that measured different aspects of their way of communicating with the patient), and the Maslach Burnout Inventory (MBI), a questionnaire that measures the risk of suffering from burn-out. Patients responded to a form containing a series of questionnaires (see Table 2).

After the training, all the technicians ('tAlk2care' and control group) carried out their usual care activity. During the first home visit, they asked the patient to participate in the study, delivering the patient information sheet and informed consent. Once they agreed to participate by signing the consent form, the patients responded to the questionnaires without the technician participating in any way.

All patients were visited 6 months (day 180) from the start of therapy. At this visit, the technician carried out the corresponding activities (review of the equipment, replacement of material, consumables, identification of

patient needs, registration of adherence), and provided the form with the same questionnaires to measure the evolution of the outcomes. After this, the technicians answered the Communication Self-efficacy survey and the Maslach Burnout Inventory (MBI).

The main outcome was experience measured on day 1 and day 180. Secondary outcomes were regarding patients: quality of life, perception of therapy, motivation to use the therapy, dyspnea (day 1 and day 180), and adherence (average number of hours of use per day, Day 180). Regarding technicians: self-efficacy in communication and risk of burnout (day 1 and day 180).

Statistical analysis

G*Power³⁸ was used to calculate the necessary sample size. This is free-to-use software used to calculate statistical power in studies. Assuming an effect size (f^2) of 0.15, an alpha of 0.05 and with 2 predictors, the estimated sample size was 74 per group. Taking into account the profile of the patients, with the probability of dropouts and loss of participants, it was proposed a sample of 85 patients per group.

A descriptive analysis of the demographic and clinical characteristics of all patients included in the study was performed, as well as a baseline analysis. General linear repeated measures model were performed to measure the effect of the training program in the two measured times and compare it with the standard of care; Eta squared was calculated to know the effect size. The interpretation of eta squared suggests that around 0.01 is a small effect, that an eta squared around 0.06 indicates a medium effect, and that an eta squared greater than 0.14 is a large effect. For adherence, an independent samples t-test was performed. A value of $p < 0.05$ was considered statistically significant.

Results

Six home respiratory therapy technicians participated in the study. 3 of them were trained by a specialist psychologist. The average age of the technicians was 36.6 (SD 6.02).

Of the 173 patients who were prescribed oxygen therapy, 169 agreed to participate (mean age 69.8 + 5.18; 56.2% were male; 78.1% were COPD): 85 patients were randomly assigned to the 'tAlk2care' experimental group (mean age 70.09 + 4.42; 60% were male; 78.8% were COPD) and 84 to the control group (mean age 69.64 + 4.95; 52.4% were male; 77.4% were COPD) (see Table 3).

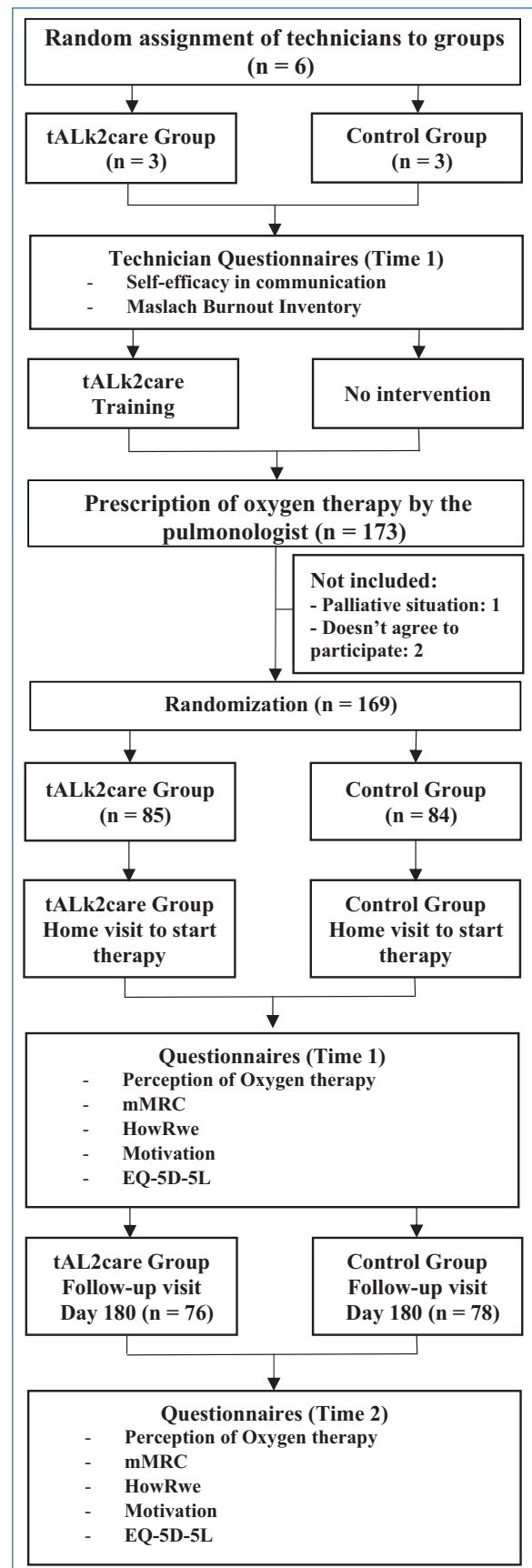


Figure 1. Study flowchart.

Of the 3 patients who were not included in the study, 2 did not agree to participate and 1 was in a palliative situation. From the 'tALk2care' group, 9 patients did not complete the study: 6 because they were not available, 2 did not want to respond to the questionnaires on day 180, and 1 showed significant physical deterioration. Of the control group, 6 patients did not complete the study: 5 because they were not available and 1 did not want to answer the questionnaires (see Fig. 1).

73.43% had a 24-hour oxygen therapy prescription. Regarding dyspnea, patients mostly presented grade 3 (43.8%) and 2 (36.1%) according to the mMRC scale. The mean FVC was 80.34 ± 20.9 , the mean FEV1 was 47.4 ± 21 , and the FEV1/FVC was 48.63 ± 16.17 . Regarding quality of life measured with Euroqol-5D-5L the average was 0.6042 ± 22 . No statistically significant differences were found between the 'tALk2care' and control groups (see Table 3). Regarding the technicians, their perception of self-efficacy in terms of communication in the 'tALk2care' group was 36.33 ± 4.16 and in the control group technicians it was 37 ± 8.54 . Regarding the Maslach Burnout Inventory, the two groups presented similar scores in the three factors (see Table 3).

At the 6-month visit, statistically significant better results were found in the 'tALk2care' group in overall experience ($p = 0.007$; with medium effect size [$\eta^2 = 0.046$]); in relational experience ($p < 0.001$; with a medium size effect [$\eta^2 = 0.046$]); in perception of oxygen therapy ($p = 0.027$; with a medium size effect [$\eta^2 = 0.032$]); in motivation ($p < 0.001$; with a large size of the effect [$\eta^2 = 0.112$]); in quality of life ($p = 0.007$; with a medium size effect [$\eta^2 = 0.047$]). No statistically significant differences were found in organizational experience ($p = 0.182$), nor in dyspnea ($p = 0.333$) nor in adherence ($p = 0.838$) (see Table 4).

The technicians who were trained with 'tALk2care' showed better self-perception in communication (mean = 39.3; SD = 1.53) than the technicians in the control group (mean = 23.7; SD = 9.45). Likewise, they presented less emotional exhaustion (mean = 16.7; SD = 1.53) than the control group (mean = 27.3; SD = 1.53) and less depersonalization (mean = 5.67; SD = 0.57 vs. mean = 9.33; SD = 1.15). Regarding personal accomplishment, the 'tALk2care' group obtained better scores (mean = 31.3; SD = 3.06) than the control group (mean = 31.3; SD = 1.53) (Table 4).

Discussion

The objective of this study was to know the impact of a communication-training program specific for patients

with respiratory pathology and aimed at home respiratory therapy technicians, and the benefit in the experience and quality of life of the patients. Our results find that care that includes relationship-centered communication, and not only focused on training in the use of the devices, improves not only the patient experience but also motivation, perception of the therapy and quality of life.

The 6-month experience shows that a quality relationship and communication, centered on empathy, is essential to promote a good experience with the care received and a better perception of the therapy. The follow-up of adherence at 6 months has shown that it remains within the acceptable range³⁹, above 15 hours/day. Motivation and good experience with care seem to have influenced these results. This seems to indicate that regardless of the importance of transmitting knowledge in the use of oxygen therapy, it is also important to ensure that the patient has the opportunity to ask questions, and this is only achieved by creating a therapeutic environment that the focus of 'tALk2care', framed in the RCC paradigm, promotes.

One of the key conclusions of the work on the optimization of oxygen therapy carried out by the American Thoracic Society was that oxygen users frequently experience significant and clinically unacceptable problems related to their oxygen equipment that decreases their quality of life. These include functional, mechanical, financial, and educational dimensions that affect their ability to work, exercise, travel, and interact with their families and community⁴⁰. Most patients are unaware of mechanisms by which to report and resolve their problems. Training is as important as allowing the patient to express their concerns, doubts or questions related to oxygen therapy.

In different studies, some patients noted barriers to optimal oxygen. Some of these barriers included confusion and lack of awareness of available oxygen delivery systems and devices, lack of a "patient-centric" approach to oxygen, something that the advocacy foundations that have received an increase in requests for assistance from their patients to resolve their oxygen problems^{6,41}. The few published patient-reported data regarding oxygen problems noted that the most frequent problems reported by patients were equipment malfunction, lack of portability (too heavy to manage), reduced availability of liquid oxygen systems (that provide high flow and portability), and the lack of portable oxygen concentrators (POCs) that deliver continuous, high-flow oxygen⁴⁰. They also encountered issues with access to oxygen, inadequate insurance coverage, and

equipment/information/service that did not meet their needs⁴¹. It is evident that knowledge is as important in therapy as the communication process with the patient in prescription, initiation and follow-up.

'tALK2care' promotes two-way communication: it helps the patient to express themselves, and the healthcare professional to identify the patients' needs, something important at the beginning of chronic treatment. All processes of illness, happen in relationships: relationships of an individual with himself/herself and with others. RCC is an important framework for conceptualizing healthcare, of great significance if we talk about treatments that impact daily life (as is the case of oxygen therapy), and where the explanation and early identification of needs. It will be key to good adherence. RCC can be defined as care in which all participants appreciate the importance of their mutual relationships, and this is what 'tALK2care' is based on⁴². Experiences based on the RCC paradigm have shown to improve the experience and quality of life of patients with chronic diseases: in COPD⁴³, diabetes⁴⁴, oncology⁴⁵ and even palliative care⁴⁶.

The study has some limitations that need to be highlighted. On the one hand, we cannot explore in detail the impact on the technicians, given that only 6 technicians participated in this study and this only allows us to compare means to see the benefit. Having more detailed information would help us know the best way to scale and implement 'tALK2care' throughout the oxygen therapy care process.

On the other hand, we do not have information on lung function at 6 months, which could give us information on the effect of the patient's clinical situation on their perception of oxygen therapy and their motivation to use it. Another limitation is that there have not been several contacts with the patient. It would be interesting to know the impact on patients who have already started therapy and sometimes patients first contact the call center, which refers the call to the technician. It would also be interesting to study what the impact is in terms of time spent with each patient in each intervention and also how many corrective actions have been avoided due to better communication. This would allow us to demonstrate the benefit of this program in terms of outcomes and also in terms of cost-benefit.

Conclusion

'tALK2care' is an intervention that improves the communication skills of the technicians responsible for oxygen therapy. This produces a better patient experience,

increases motivation to use oxygen therapy and quality of life. Patients with chronic respiratory diseases see their healthcare personnel as a source of support and information about their treatment. More effective communication, taking into account the individual needs and preferences of the patient, can help to lessen the impact that COPD or IPF has on the lives of patients and their loved ones.

Funding

None.

Conflicts of interest

The authors declare that does not exist an interest conflict.

Ethical considerations

Protection of humans and animals. The authors declare that the procedures followed complied with the ethical standards of the responsible human experimentation committee and adhered to the World Medical Association and the Declaration of Helsinki. The procedures were approved by the institutional Ethics Committee.

Confidentiality, informed consent, and ethical approval. The authors have followed their institution's confidentiality protocols, obtained informed consent from patients, and received approval from the Ethics Committee. The SAGER guidelines were followed according to the nature of the study.

Declaration on the use of artificial intelligence. The authors declare that no generative artificial intelligence was used in the writing of this manuscript.

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Home-based rehabilitation maintenance with or without an exercise-based pilates program in COPD patients

Mantenimiento con ejercicios domiciliarios con o sin un programa de pilates basado en ejercicio en pacientes con EPOC

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Abstract

Background: Pilates exercise may complement pulmonary rehabilitation of chronic obstructive pulmonary disease (COPD) to improve patient-reported outcome measures (PROMs). **Objective:** To investigate the effects on PROMs after incorporating a six-month Pilates exercise maintenance program compared to a home-only exercise program. **Material and methods:** A total of 32 participants with COPD (GOLD B) were assigned to either the intervention group ($n = 14$) or the control group ($n = 18$). Both groups participated in a three-month pulmonary rehabilitation program. For the next six months, the intervention group engaged in a Pilates exercise program and home-based exercises, while the control group continued with home-based exercises only. Primary outcomes were evaluated at baseline, three, six, and nine months. **Results:** At the nine-month follow-up, the intervention group exhibited significantly fewer activity-related impairments in the St George's Respiratory Questionnaire Activity domain ($p = 0.029$) and experienced less symptoms of anxiety ($p = 0.002$) and depression ($p = 0.014$) according to the Hospital Anxiety and Depression Scale scores. Other PROMs did not show statistically significant differences between the groups. **Conclusion:** Incorporating a six-month Pilates exercise maintenance program resulted in fewer activity-related impairments and reduced anxiety and depression symptoms compared to those who followed a home-based exercise maintenance program alone.

Keywords: COPD. Pulmonary rehabilitation. Community-based. Home-based. Pilates.

Resumen

Antecedentes: El ejercicio de Pilates puede complementar la rehabilitación pulmonar de la enfermedad pulmonar obstructiva crónica (EPOC) para mejorar las medidas de resultado reportadas por los pacientes (MRRP). **Objetivo:** Investigar los efectos en los MRRPs tras incorporar un programa de mantenimiento de ejercicios de pilates de seis meses en comparación con un programa domiciliario de ejercicios. **Material y métodos:** Un total de 32 participantes con EPOC (GOLD B) fueron asignados al grupo de intervención ($n = 14$) o al grupo de control ($n = 18$). Ambos grupos participaron en un programa de rehabilitación pulmonar de tres meses. Durante los siguientes seis meses, el grupo de intervención participó en un programa de ejercicios de Pilates y un programa domiciliario de ejercicios, mientras que el grupo de control continuó solo con un programa domiciliario de ejercicios. Los resultados primarios se evaluaron en la línea de base a los tres, seis y nueve meses.

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Resultados: En el seguimiento de nueve meses, el grupo de intervención presentó significativamente menos limitaciones relacionadas con la actividad en el dominio de Actividad del St George's Respiratory Questionnaire ($p = 0,029$) y experimentó menos síntomas de ansiedad ($p = 0,002$) y depresión ($p = 0,014$) según las puntuaciones del Hospital Anxiety and Depression Scale. Otras MRRPs no mostraron diferencias estadísticamente significativas entre los grupos. **Conclusión:** Incorporar un programa de mantenimiento de ejercicios de pilates de seis meses resultó en menos limitaciones relacionadas con la actividad y una reducción de los síntomas de ansiedad y depresión en comparación con aquellos que siguieron únicamente un programa domiciliario de ejercicios.

Palabras clave: Enfermedad pulmonar obstructiva crónica. Rehabilitación pulmonar. Comunitario. En casa. Pilates.

Introducción

Chronic obstructive pulmonary disease (COPD) is characterized as a disabling and long-term respiratory disease, with most notable symptoms including dyspnea and productive coughing. Pulmonary rehabilitation programs are fundamental to the effective management of COPD. Various settings for pulmonary rehabilitation and strengthening exercise programs have been shown to improve COPD-related symptoms such as dyspnea, as well as other patient-reported outcome measures (PROMs) including decreased activity-related impairments, improved health-related quality of life, and reduced psycho-emotional symptoms¹⁻⁴. Evaluating PROMs is crucial to assess the effectiveness of interventions as perceived by the patient.

Community-based pulmonary rehabilitation programs are cost-effective and may yield better outcomes compared to conventional care interventions⁵. However, it is vital to implement continued supervised maintenance exercise programs following pulmonary rehabilitation to reduce health care utilization and costs⁶. Home-based pulmonary rehabilitation can supplement traditional pulmonary rehabilitation conducted in primary care or community-based settings and has demonstrated positive effects on maintaining or improving PROMs related to COPD symptoms and health-related quality of life⁷⁻¹¹. In turn, a Pilates exercise program can complement traditional pulmonary rehabilitation by providing a controlled setting for deep breathing exercises that enhance the strength of inspiratory and expiratory muscles in COPD patients¹². The breathing techniques used in Pilates training can boost lung capacity, improve the functionality of the deeper abdominal muscles¹³ and enhance the strength of expiratory muscles¹². These improvements are expected to reduce dyspnea symptoms associated with COPD, thereby potentially alleviating

COPD-related anxiety and depression symptoms and enhancing health-related quality of life.

Therefore, our main goal was to compare the benefits of home-based pulmonary rehabilitation with or without a Pilates exercise program following an initial three months of traditional pulmonary rehabilitation in individuals with COPD. The primary outcomes were dyspnea symptoms, anxiety and depression symptoms, and health-related quality of life.

Materials and methods

Design and study population

This study followed a nonrandomized clinical trial design, where the participants were assigned to the groups by the researchers. Participant allocation did not follow specific patient or COPD-related criteria but was mostly based on their geographical location and availability to regularly attend the Pilates classes for the duration of the study.

Individuals with COPD were invited to participate in the study by a general practitioner or family medicine doctor working in the National Health Service (in the north of Portugal). Participant enrollment took place between July 2018 and May 2021. The inclusion criteria comprised male or female adults (≥ 18 years old) with COPD presenting stable symptoms for the previous 3 months. COPD was diagnosed as postbronchodilatation Forced Expiratory Volume in 1 second/Forced Vital Capacity (FEV_1/FVC) below 0.7¹⁴. Only those classified as grade B of the Chronic Obstructive Lung Disease (GOLD) 2024 system¹⁴ were included in this study to homogenize the symptoms of COPD among the participants. GOLD B was defined as 0 or 1 moderate exacerbation in the previous year (not leading to hospital admission), and symptoms scored by the modified Medical Research Council (mMRC) as ≥ 2

and COPD Assessment Test (CAT) as ≥ 10 . The exclusion criteria comprised older individuals (≥ 85 years old), those with a history of acute cardiac or respiratory events in the last month, or any medical condition that would limit their exercise tolerance (severe cardiac, musculoskeletal, neuromuscular conditions, or recent surgery). Individuals with psychiatric conditions or cognitive deficits, a history of neoplasia or immunologic diseases, or those unable or unwilling to attend the community-based exercise program were also excluded from the study.

Sample size

An *a priori* sample size calculation was conducted using the Software G*Power (Version 3.1.9.2, Kiel, Germany) based on the results of a previous randomized controlled trial¹⁵, using the between-group improvement in the St George Respiratory Questionnaire (SGRQ) total score as the reference outcome after 3 months of pulmonary rehabilitation. For the sample size calculation, a power (α) of 95% was considered to detect statistically significant differences ($p < 0.05$) using a two-tailed independent Mann-Whitney test. A total of 32 participants (16 per group) would be needed to achieve statistically significant power.

Ethics

The study was approved by the Institutional Ethical Committee (IRB number: ERS Norte, study ID T667, No.37/2017). All participants signed an informed consent form before enrolling in the study, and all procedures were conducted in accordance with the Declaration of Helsinki.

Procedures

Participants were divided into control and intervention groups using a nonrandomized method. For the first 3 months of the study, both groups followed the same traditional community-based pulmonary rehabilitation program. After the first 3 months, both groups were instructed to continue the recommended home-based exercises for an additional 6 months; however, the intervention group was also assigned to a 6 month Pilates exercise program in addition to the home-based exercises.

Both groups were evaluated at baseline and at the end of the 3 months of initial traditional community-based pulmonary rehabilitation. Additionally, both groups were followed over time and re-evaluated at the 6th and 9th month from baseline.

Intervention

TRADITIONAL COMMUNITY-BASED PULMONARY REHABILITATION

Both groups followed the same community-based pulmonary rehabilitation intervention (2 x/week, 60 minutes each session) for the first 3 months. The same physiotherapist conducted all intervention classes, with 4 to 12 participants per class. The traditional pulmonary rehabilitation program included warm-up, aerobic exercise, resistance strengthening, and cooldown (Table 1). For the aerobic exercise, participants were instructed to walk on a treadmill or in a closed circuit outside the clinic. They started with a walking speed perceived as level 5 on the modified Borg scale. Every 5 minutes, participants were asked about their perceived dyspnea/fatigue, and if above level 6, the walking speed was decreased until reaching level 5 on the modified Borg scale. For resistance strengthening, 2 sets of 10 to 12 repetitions were required for each major muscle group (Table 1). A load that evoked fatigue after 10 to 12 repetitions was selected. Exercise dosage followed the overload principle to promote improvements in muscular strength. Load was increased when the current workload allowed for 1 or 2 repetitions over the desired 12 repetitions after two consecutive training sessions.

In addition to the face-to-face sessions, participants in both groups were encouraged to follow a recommended home-based exercise program (Supplementary data 1) throughout the study to complement the in-person classes. Participants recorded their adherence to the home-based program in an exercise diary. At the end of each week, participants were contacted (via telephone) to report their compliance with the recommended home-based exercises. If compliance was under 80% (controlled every 3 months), the participant was excluded from the follow-up.

EXERCISE PROGRAM BASED ON PILATES PRINCIPLES

The intervention group received an exercise program based on Pilates principles, implemented twice a week (45 minutes per session) and led by the same physiotherapist trained in Pilates exercise. The first session started with demonstrating a set of five basic and less physically demanding exercises (Fig. 1A–E). Each exercise was completed for 2 sets of 10 repetitions (or 8 times for each side for unilateral exercises) with 60 seconds of rest between sets and exercises. The duration of each exercise (2 sets) was around 4 to 8 minutes (including rest time and depending on the difficulty of exercises and if it was a unilateral exercise).

Table 1. Initial 3 month community-based pulmonary rehabilitation intervention program

Activity	Duration	Intensity	Exercise
Warm-up	10 minutes	Low	Joint mobility and light stretching exercises (spine, upper and lower limbs) Breathing exercises
Aerobic exercise	20 minutes	Low (4-6 on perceived dyspnea/fatigue on the modified Borg scale)	Walking
Resistance exercise	20 minutes	Moderate (70-75% RM)	Strengthening exercises of the major upper muscle groups and lower limb muscle groups as well as trunk and low back muscles using free weights and ankle weights
Cooldown	10 minutes	Low	Static and dynamic exercises at the upright position in addition to the same exercises used in the warm-up

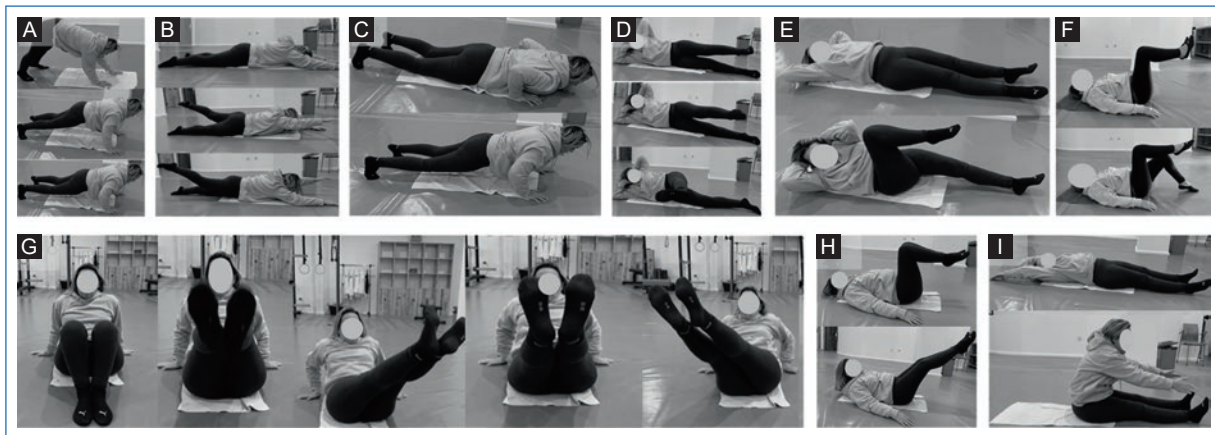


Figure 1. Summary of exercises included in the first 3 months of exercise program based on pilates principles. the first five initial exercises included: **A:** imprinting; **B:** one leg stretch; **C:** shoulder bridge; **D:** clam; **E:** saw. Then, there were four progression exercises: **F:** mermaid; **G:** spine stretch forward; **H:** bird dog; **I:** cat stretch.

Difficulty progression of exercises was made on a case-by-case basis. There were four additional and more challenging exercises available for progression (Fig. 1F–I). Participants progressed in difficulty when demonstrating that one of the five previous exercises was not challenging enough (< 4 points on the modified Borg scale). To progress in difficulty, the participant had to demonstrate that the exercise was performed with proper technique and without any compensatory movements. If the participant was unable to perform the new exercise (after progression), the exercise was regressed to the previous, less challenging one. The following week, the technique with the new exercise was retested, and progression/regression was decided as appropriate.

After the first 3 months of the Pilates exercise program, the physiotherapist introduced a set of five new

exercises (Fig. 2A–E) to provide variability and include more physically demanding exercises. There were four additional exercises available for progression (Fig. 2F–I). During this stage, each exercise was required to be completed for 2 sets of 12 repetitions, using the same progression and regression rules. However, if the participant could not tolerate the difficulty of the new set of five exercises, they regressed to the exercises of the previous stage and then progressed as tolerated. The full Pilates exercise protocol is described in the supplementary data 2.

PATIENT-REPORTED OUTCOME MEASURES

At baseline, we collected the sociodemographic and biometric data of all participants. The PROMs were collected at baseline and at 3, 6, and 9 months follow-up.

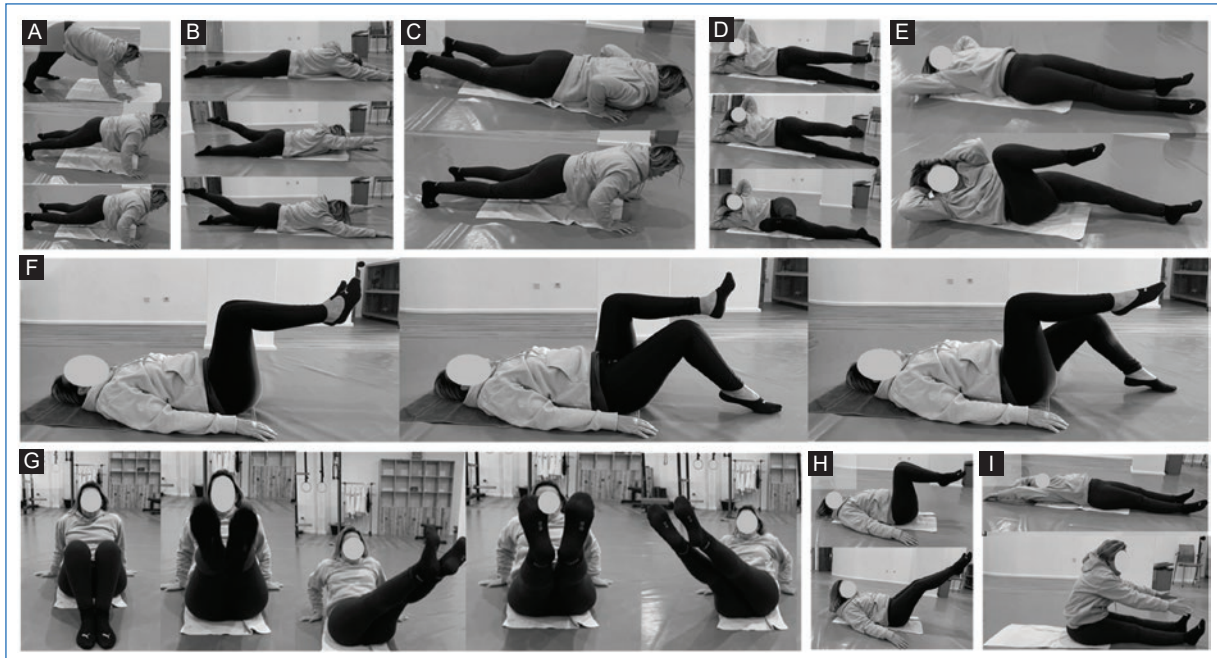


Figure 2. Summary of exercises included in the second 3 months of exercise program based on Pilates principles. The first five initial exercises included: **A:** push up; **B:** swimming; **C:** leg pull prone; **D:** side kick; **E:** criss cross. Then, there were four progression exercises: **F:** scissors; **G:** hip twist; **H:** hundred; **I:** roll up.

The PROMS included those related to dyspnea symptoms evaluated by the mMRC dyspnea scale, COPD-related quality of life evaluated by the CAT and the SGRQ, and psychological status evaluated by the Hospital Anxiety and Depression Scale (HADS). Further information on these PROMs is available in supplementary data 3.

STATISTICAL ANALYSIS

The software IBM SPSS version 29.0 was used for all statistical analyses. Due to the nonGaussian distribution (Kolmogorov–Smirnov test) and the small sample size, nonparametric statistics were conducted. Continuous variables are described using median and interquartile range, and categorical variables as count and frequency (%). A p value of 0.05 was used to determine statistical significance.

Between-group statistical differences were compared using the Mann-Whitney U tests. The χ^2 test was used to compare categorical variables between groups, with the Fisher exact test applied when appropriate. Within-group statistical differences were compared using the one-way repeated measures Friedman test with posthoc

Wilcoxon signed-rank tests for pairwise comparisons between follow-up endpoints.

Results

Characteristics of included participants

Of the 200 participants screened for inclusion in this study, 140 were initially excluded due to not fulfilling the eligibility criteria. A further 25 participants were excluded as they presented other COPD grades besides GOLD B, and an additional 3 patients had a GOLD stage III or IV denoting severe airflow impairments. A total of 32 participants were eligible and enrolled in the study, with 14 allocated to the intervention group and 19 to the control group. One patient in the control group was lost to follow-up at 3 months (Fig. 3).

The baseline sociodemographic and biometric characteristics were homogeneous between the groups (Table 2). The baseline GOLD characteristics (stage and symptoms) were not significantly different between the groups. Only the rate of COPD-related exacerbations was statistically different, with the intervention group showing a significantly higher rate of two or more exacerbations in the past year (78.6% vs. 0.0%).

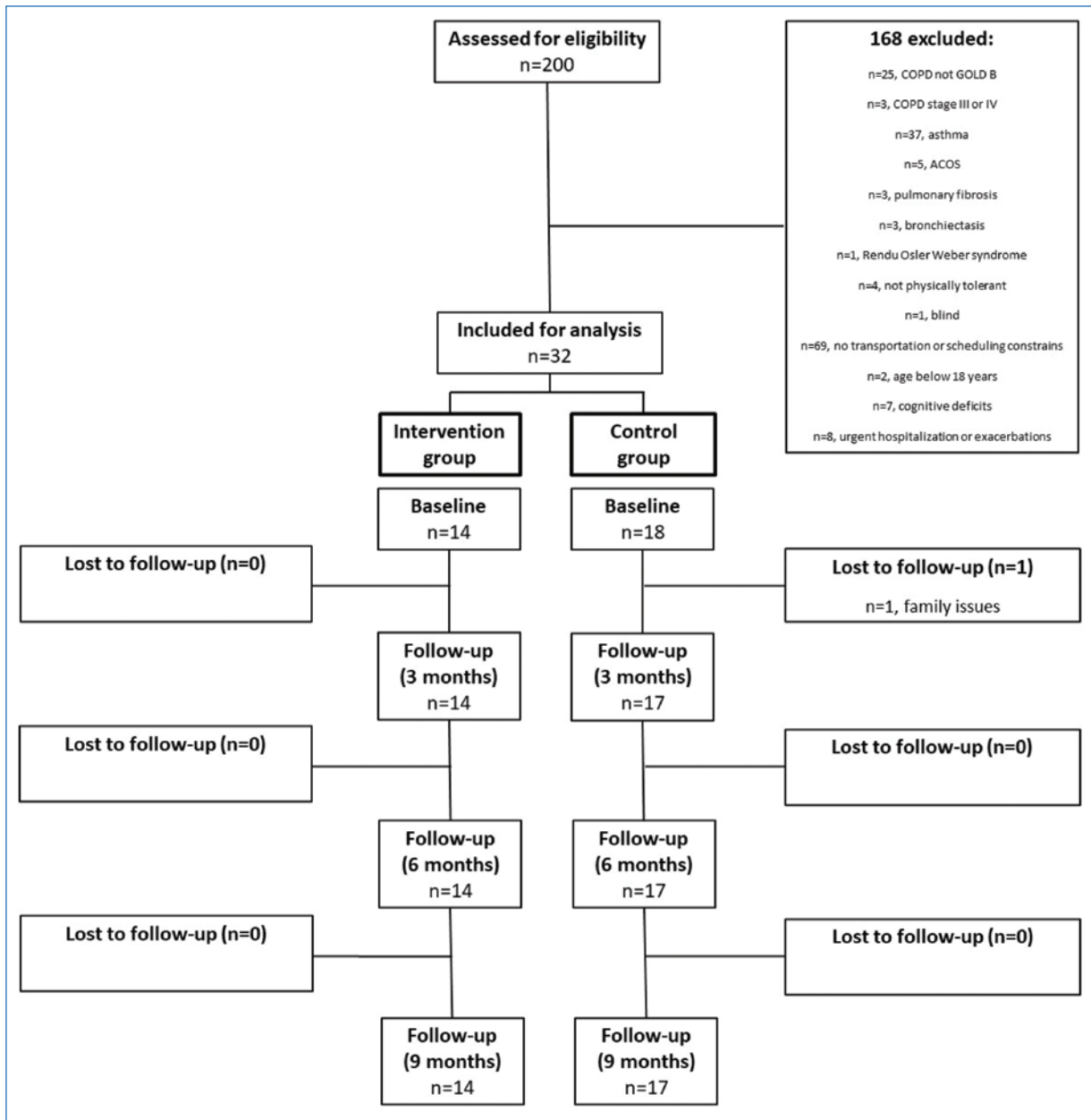


Figure 3. Flow diagram of the study.

Patient-reported outcome measures

The mMRC significantly improved after 3 months of traditional pulmonary rehabilitation in both groups and was not significantly different between groups at baseline and at the 3 month endpoint. Both groups showed a statistically significant improvement over time, but not when comparing the 9 months to 3 months, and without significant differences between groups at each follow-up endpoint (Table 3).

The CAT score was not significantly different between groups at baseline and after 3 months of traditional pulmonary rehabilitation. However, the control group showed a statistically significant improvement of 8 points at 3 months ($p = 0.009$). After 6 months of pilates (9 month follow-up), the intervention group showed a statistically significant decrease of 7 points ($p = 0.033$), while the control group displayed a deterioration of the CAT score from 8 points at 3 months to 14 points at 6 months ($p = 0.023$), which maintained at 9 months.

Table 2. Baseline sociodemographic, biometric and COPD-related characteristics of included participants

Variables	Intervention group (n = 14)	Control group (n = 18)	p
Age (years)	65.5 (11.8)	66.5 (10.5)	0.561
Sex			
Male	11 (78.6%)	12 (66.7%)	0.694*
Female	3 (21.4%)	6 (33.3%)	
Heigh (m)	1.65 (0.16)	1.65 (0.13)	0.955
Weight (Kg)	74.0 (19.5)	72.0 (22.8)	0.925
% of fat mass	26.1 (15.4)	25.3 (18.6)	0.357
BMI (kg.m ⁻²)	25.7 (6.9)	25.3 (7.0)	0.837
GOLD stage (%)			
GOLD I	2 (14.3%)	9 (50.0%)	0.061*
GOLD II	12 (85.7%)	9 (50.0%)	
GOLD symptoms			
CAT < 10	3 (21.4%)	0 (0%)	0.073*
CAT ≥ 10	11 (78.6%)	18 (100%)	
GOLD exacerbations			
AECOPD < 2	3 (21.4%)	18 (100%)	< 0.001*
AECOPD ≥ 2	11 (78.6%)	0 (0%)	

*Exact Fisher test.

AECOPD: acute exacerbations of COPD; BMI: Body Mass Index; CAT: COPD Assessment Test; GOLD: Global Initiative for Chronic Obstructive Lung Disease system.

However, the CAT score was not statistically significant between groups at the 6 and, 9 months follow-up (Table 3).

The SGRQ total score was not significantly different between groups at baseline and after 3 months of traditional pulmonary rehabilitation. However, the control group showed a statistically significant improvement of 17.3 points at 3 months ($p = 0.004$). During the subsequent 6 months of intervention (9 month follow-up), the intervention group showed a statistically significant improvement ($p = 0.023$), whereas the control group did not show any change ($p = 0.326$). There were no significant differences between the groups at any of the follow-up timepoints (Table 3).

When comparing the SGRQ domains, there were no significant differences between groups at baseline and after 3 months of traditional pulmonary rehabilitation. Statistically significant improvements were observed from baseline to 3 months in the SGRQ Activity and Impact domains for the control group, with a median decrease of 12.5 points ($p = 0.022$) and 24.2 points ($p = 0.026$), respectively. Despite these improvements in the control group at 3 months, there were no significant

between-group differences (Table 3). For the SGRQ Activity domain, the control group further improved by 11.2 points at 6 months ($p = 0.030$) but reverted back to 47.7 points at 9 months without any differences compared to 3 months ($p = 0.925$). For the SGRQ Impact domain, the control group showed a deterioration of the score at 6 months with an increase of 14.2 points ($p = 0.044$), which remained at 9 months ($p = 0.152$ compared to 6 months and $p = 0.156$ compared to 3 months). The intervention group did not show any statistically significant improvement between follow-up timepoints (Table 3). However, the SGRQ Activity domain was significantly lower in the intervention group at 9 months (29.8 vs. 47.7 points, $p = 0.029$), indicating less impairment in daily-living activities. No significant changes were observed for the SGRQ Symptoms domain between the two groups or at any timepoints during the follow-up within each group (Table 3).

The HADS score was not significantly different between groups at baseline and after 3 months of traditional pulmonary rehabilitation. The HADS Anxiety domain showed a significant improvement over time in the intervention group, improving by 1 point at 3 months ($p = 0.027$) and

Table 3. Comparison within and between groups at baseline and follow-up of patient-reported outcome measures (PROMs)

Variables	Groups	Baseline	3 months	6 months	9 months	Overall
mMRC (0-4)	Intervention	2.0 (1.0)	2.0 (1.0)*	1.5 (2.0)	0.5 (2.0)	0.002
	Control	2.0 (1.0)	1.0 (0.0)*	1.0 (1.0)	1.0 (1.0)	0.004
	Between groups	0.071	0.161	0.769	0.297	
CAT (0-40)	Intervention	19.5 (14.0)	16.0 (11.0)	11.5 (16.0)	9.0 (17.0) [†]	0.019
	Control	16.0 (9.0)	8.0 (12.0)*	14.0 (8.0) [†]	14.0 (8.0)	0.002
	Between groups	0.866	0.053	0.570	0.215	
SGRQ - Symptoms (0-100)	Intervention	42.4 (36.6)	42.4 (25.6)	38.0 (25.0)	35.8 (51.1)	0.041
	Control	43.3 (30.8)	32.1 (43.4)	42.9 (37.6)	28.5 (33.3)	0.095
	Between groups	1.000	0.493	0.597	0.468	
SGRQ - Activity (0-100)	Intervention	55.3 (46.2)	45.6 (49.1)	34.1 (50.1)	29.8 (32.8)	0.031
	Control	59.5 (15.8)	47.0 (32.6)*	35.8 (39.3) [†]	47.7 (23.4)	0.005
	Between groups	0.357	0.739	0.922	0.029	
SGRQ - Impact (0-100)	Intervention	26.0 (46.9)	25.3 (31.9)	19.3 (32.5)	17.3 (40.7)	0.321
	Control	38.5 (26.8)	14.3 (35.8)*	28.5 (36.6) [†]	26.6 (33.3)	0.014
	Between groups	0.561	0.131	0.262	0.681	
SGRQ - Total (0-100)	Intervention	34.1 (37.8)	35.7 (32.4)	34.4 (33.6)	35.1 (34.8) [†]	0.066
	Control	43.6 (20.8)	26.3 (38.1)*	33.4 (20.1)	29.9 (34.8)	0.005
	Between groups	0.206	0.423	0.681	0.444	
HADS - Anxiety (0-21)	Intervention	7.0 (9.0)	6.0 (6.0)*	4.5 (6.0)	5.5 (7.0) [†]	0.001
	Control	8.0 (8.0)	7.0 (5.0)	9.0 (5.0) [†]	10.0 (5.0) [†]	0.101
	Between groups	0.985	0.681	0.010	0.002	
HADS - Depression (0-21)	Intervention	3.5 (8.0)	4.5 (5.0)	4.0 (4.0)	4.0 (6.0)	0.427
	Control	8.0 (4.0)	6.0 (5.0)*	8.0 (5.0) [†]	6.0 (3.0)	< 0.001
	Between groups	0.054	0.653	0.002	0.015	

Data presented as median and IQR. Lower values denote less dyspnea (mMRC), less impairment (CAT and SGRQ) and less anxiety and depression symptoms (HADS).

*p < 0.05 vs baseline.

[†]p < 0.05 vs 3 months.

CAT: COPD Assessment Test; HADS: Hospital Anxiety and Depression Scale; SGRQ: St George Respiratory Questionnaire; mMRC: modified Medical Research Council.

by another 0.5 points at 9 months ($p = 0.041$ compared to 3 months). The control group deteriorated at 6 months by 2 points ($p = 0.027$) and by another 1 point at 9 months ($p = 0.041$ compared to 3 months). The HADS Depression domain did not show any significant changes in the intervention group. In the control group, although it improved by 2 points after 3 months of traditional pulmonary rehabilitation ($p = 0.016$), it reverted by 2 points at 6 months ($p < 0.001$) without significant differences at 9 months compared to 3 months ($p = 0.303$). The intervention group

showed significantly less anxiety and depression symptoms than the control group at both 6 and, 9 months follow-up (Table 3).

Discussion

The results of the study demonstrate significant improvement in decreasing the impairment in daily activities and reducing the anxiety and depression symptoms following a 6 months combined intervention of

Pilates with home-based exercises as compared to only home-based exercises in individuals with COPD. Participants in the intervention group experienced a significantly higher rate of exacerbations at baseline and it is thus advised some caution when analyzing the results at follow-up as exacerbations may influence treatment indications and prognosis.

In the evaluation of dyspnea carried out through mMRC, both intervention and control groups exhibited a significant improvement in mMRC scores after 3 months of traditional pulmonary rehabilitation. There were no significant differences in mMRC scores between the groups at baseline and the 3 month endpoint, highlighting the effectiveness of both interventions in enhancing breathlessness perceptions. Although there were not statistically significant differences in dyspnea symptoms (as measured by the mMRC) in both groups at 9 months follow-up, there was a significant change over-time within each group ending with a median mMRC of grade 1 in both the intervention and control groups. The median improvement surpassed the minimal clinically important difference (MCID) of -0.5 and -1 units^{16,17} and changed from breathless at a normal walking pace (grade 2) to breathless only when walking at a fast pace or up a slight hill (grade 1). These results are superior to those reported for community-based pulmonary rehabilitation that show no significant improvement in the mMRC as compared to usual care⁵. A recent Cochrane review¹⁸ found significant improvement in dyspnea assessed with the mMRC when comparing inspiratory muscle training against control or sham groups. Likewise, a recent trial¹⁹ found a similar improvement in the mMRC after 3 months of traditional pulmonary rehabilitation in patients with mild COPD. These findings may help explain the improvements in the mMRC after 3 months of traditional pulmonary rehabilitation. Other studies^{20,21} found improvement in mMRC after 3 months home-based pulmonary rehabilitation in mild COPD patients which highlight the role of home-based exercises and why our control group with only home-based exercises also maintained the improvement (from the initial traditional pulmonary rehabilitation) throughout the remaining 6 months of the study.

The CAT scores did not significantly differ between groups initially and after 3 months of traditional pulmonary rehabilitation. Whereas the control group did not show any within-group significant improvement, the Pilates intervention demonstrated a notable improvement at 9 months (median improvement of 7 points), indicating its potential benefit in reducing COPD symptoms captured by the CAT questionnaire. This large improvement is more than 4-fold the established MCID

of -1.6 units for the CAT score¹⁸, ending at 9 months with a low COPD-related symptom burden impact. The improvement in the CAT score by the intervention group is consistent (and event superior) to those reported in a recent study²¹ showing a decrease of 6 points after 3 months of a home-based pulmonary rehabilitation program. Likewise, the median 7 points improvement is also superior to the pooled change of -3 points in CAT score reported in a recent Cochrane review¹⁸ comparing inspiratory muscle training against control or sham groups. Conversely, the CAT of the control group at 6 months reverted back to baseline values, highlighting that home-based exercises may not be enough to maintain the improvement in COPD-related symptom burden obtained after the traditional pulmonary rehabilitation.

In the evaluation carried out by SGRQ, the total scores did not significantly differ between groups at baseline and after 3 months of traditional pulmonary rehabilitation. The control group showed a significant improvement after the initial 3 months traditional pulmonary rehabilitation but deteriorated at follow-up with no significant changes comparing to the baseline values. The improvements in the SGRQ Activity and Impact domains in the control group at 3 months indicate the effectiveness of traditional pulmonary rehabilitation. The intervention group did not show such significant improvement at 3 months, which may be related to the higher rate of exacerbations at baseline. The subsequent improvements in the SGRQ Activity domain in the control group at 6 months suggest that the maintenance with home-based exercises was effective in reducing the COPD impact on daily activities. However, the SGRQ Activity domain deteriorated in the control group at 9 months, with the intervention group showing significantly less breathlessness-related impairment in daily-living activities. The between-group difference at 9 months was almost 18 points which is clinically relevant and surpassing by 450% the -4 units established as MCID^{22,23}. The isolated improvement in the SPGQ Activity domain is also consistent with the findings of a recent meta-analysis⁵ which showed that improvement in the SPGQ score was only significant for the SPGQ Activity domain. While traditional pulmonary rehabilitation programs significantly improve COPD-related quality of life as measured by the SPGQ^{19,24}, further long-term improvements with maintenance pulmonary rehabilitation programs are not expected^{18,25}.

The HADS Anxiety and Depression domains depicted varying patterns of improvement and deterioration over time in both groups. Notably, the intervention group showed significantly reduced anxiety and depression

symptoms relative to the control group at the 6- and 9 month follow-ups highlighting the potential influence of Pilates on these PROMs. While the control group deteriorated or maintained the anxiety and depressive symptoms at 9 months, the intervention group showed a significant decrease in anxiety symptoms after the 6 months of Pilates program. However, the median improvement did not surpass the MCID of -1.8 to -1.3 units²⁶. These findings denote a potential role of Pilates in the psychological status of COPD patients. Indeed, the anxiety symptoms in the intervention group at 9 months were almost half of those for the control group. The control group had a median HADS anxiety score of 10 points at 9 months, which is borderline value and close to the 11 points determined for clinical anxiety²⁷. Although the significant differences in the depression domain at 9 months, it is important to highlight that the intervention group already had fewer depressive symptoms at baseline (albeit not statistically significant), which may partially explain the differences at 9 months follow-up. The improvement in anxiety and depressive symptoms is consistent with those reported in randomized control trials that implement community-based pulmonary rehabilitation programs^{28,29} as well as for clinical trials implementing Pilates exercise in other clinical conditions³⁰⁻³³. However, maintenance programs seem to not improve the anxiety and depression symptoms as compared to usual care³⁴, which may explain why the control group with only home-based exercises experienced deterioration of the HADS score. It may be speculated that improvement in anxiety symptoms in the intervention group may be from a better control of respiratory function and relaxation from the Pilates exercise. Likewise, the lower anxiety and depression symptoms in the intervention group throughout the 6 months after the traditional pulmonary rehabilitation may be related to the group interaction and sociability with other COPD patients during the Pilates sessions. This is an important finding because improving anxiety and depression symptoms is a challenging clinical task in patients with COPD, even when tailored psychological interventions based on a cognitive behavioral approach are implemented³⁵.

Clinical implications

The study findings underscore the importance of traditional pulmonary rehabilitation and Pilates as effective interventions in improving COPD symptoms and patient well-being. The observed improvements in some PROMs suggest potential benefits of these interventions in reducing COPD-related impairments and in enhancing

quality of life and mental health in individuals with COPD. The superior improvement in the SGRQ Activity domain indicate that Pilates exercise may be a useful tool to control the dyspnea symptoms and reduce the COPD-related daily living impairments that are often compromised by respiratory problems such as cough and shortness of breath³⁶. Moreover, the lower anxiety symptoms in the intervention group underpin that face-to-face group rehabilitation should be prioritized when available to promote peer-to-peer interaction and thus decrease the potential risk of worsening of psychological status of COPD patients when undertaking home-based rehabilitation isolated and by themselves. Indeed, it is known that social isolation and loneliness are common issues among patients with COPD³⁷ and that these patients are at risk of for mental disorders as anxiety and depressive states are diagnosed in more than half of COPD patients³⁸. It is important that clinicians, policymakers, and health systems take steps to identify and address social isolation and loneliness in COPD individuals³⁹ and promote group-based sessions where possible. In cases where face-to-face pulmonary rehabilitation is not possible, it is important to identify barriers in the home environment and facilitators to implement home-based strategies for effective self-management interventions.

Future research directions

Future research could focus on exploring the sustainability of intervention effects beyond 9 months and examining the long-term benefits of augmenting the traditional pulmonary rehabilitation with a Pilates exercise program.

Further investigations could also explore the individual variability in response to these interventions (with a higher sample size) and identify optimal approaches for personalized COPD management.

Limitations

This study has some limitations that need to be acknowledged. The small size is small and unbalanced, with the intervention group having a lower sample due to the exclusion of some patients who were not classified as GOLD B. Although 28 participants with grades other than GOLD B or GOLD stage III/IV completed the entire study, it was decided to exclude these from analyses to prevent selection bias. The small sample resulted in low statistical power in analyses and may have led to type II error in not finding statistical differences when the between-groups differences were clinically relevant

(e.g., for mMRC or CAT). Another limitation was the lack of randomization that may have introduced selection bias at the start of the study; although no significant differences were found between groups for PROMs at baseline, the participants in the intervention group experienced a significantly higher rate of exacerbations which may have influenced the follow-up outcomes. Future studies should implement randomization procedures to mitigate risk of selection bias. All patients were from the same region of the country which may have introduce selection and geographical bias and limits the generalizability (external validity) of our findings to individuals with COPD from other regions of the country and other countries. Outcome evaluations were made by the same health professional that delivered the interventions (not blinded) which may have introduced a risk of detection bias and influenced the way that outcome measures were assessed. Moreover, the intervention group received for 6 months a Pilates exercise program (in addition to the home-based exercises) as compared to the control group that only engaged in the home-based exercises which may have had an effect on PROMs due to added exposure to intervention and influence the adherence to a longer-lasting intervention (6 months). The control group receive minor intervention during the follow-up period (home-based exercises), as it would have been unethical to leave these patients without any guidance. To minimize the risk of performance bias, participants in the intervention group were instructed to also follow the same home-based program, with the adherence of both groups being monitored to ensure that it met at least 80% of the recommended frequency.

Conclusion

This study highlights the positive impact of augmenting the traditional pulmonary rehabilitation with a Pilates exercise program in decreasing the impairment in daily activities and reducing the anxiety and depression symptoms in individuals with COPD. These findings contribute to the growing body of evidence supporting the efficacy of comprehensive rehabilitation programs in enhancing COPD management and patient well-being.

Author contributions

All authors were involved in the conception of this study. M. Barbosa performed the interventions, collected all data and conducted the statistical analysis. C. Argel-de Melo and R. Torres guided and provide

advice during all steps, as well as critical revision of the content. All authors contributed to the drafting of the manuscript and approved the final submitted work.

Supplementary data

Supplementary data are available at DOI: 10.24875/RPR.24000039. These data are provided by the corresponding author and published online for the benefit of the reader. The contents of supplementary data are the sole responsibility of the authors.

Funding

None.

Conflicts of interest

None.

Ethical considerations

Protection of humans and animals. The authors declare that the procedures followed complied with the ethical standards of the responsible human experimentation committee and adhered to the World Medical Association and the Declaration of Helsinki. The procedures were approved by the institutional Ethics Committee.

Confidentiality, informed consent, and ethical approval. The authors have followed their institution's confidentiality protocols, obtained informed consent from patients, and received approval from the Ethics Committee. The SAGER guidelines were followed according to the nature of the study.

Declaration on the use of artificial intelligence. The authors declare that no generative artificial intelligence was used in the writing of this manuscript.

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Neumotórax bilateral como presentación de linfangioleiomiomatosis en paciente con esclerosis tuberosa

Bilateral pneumothorax as a presentation of lymphangioleiomyomatosis in a patient with tuberous sclerosis

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Resumen

Se aborda el caso clínico de una paciente de 21 años con antecedentes de esclerosis tuberosa diagnosticada en la infancia, quien desarrolló neumotórax bilateral como presentación inicial de una linfangioleiomiomatosis (LAM). El caso se complica con la recurrencia del neumotórax y la necesidad de hasta dos bullectomías. El objetivo es reseñar el interés de este caso por su forma de presentación y la particularidad del tratamiento, destacando la relevancia de la detección temprana de LAM, sus posibles complicaciones y la disponibilidad de tratamientos plausibles.

Palabras clave: Linfangioleiomiomatosis. Esclerosis tuberosa. Neumotórax.

Abstract

This clinical case is about a 21-year-old woman with a history of tuberous sclerosis diagnosed in childhood, who developed bilateral pneumothorax as the initial presentation of lymphangioleiomyomatosis (LAM). The case is complicated by the recurrence of pneumothorax and the need for up to two bullectomies. The aim is to review the interest of this case due to its presentation and the particularity of the treatment, highlighting the relevance of early detection of LAM, its potential complications, and the availability of plausible treatments.

Keywords: Lymphangioleiomyomatosis. Tuberous sclerosis. Pneumothorax.

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Introducción

La linfangioleiomiomatosis (LAM) es una enfermedad pulmonar intersticial rara, caracterizada por el crecimiento anormal y no neoplásico de células musculares atípicas en los vasos sanguíneos, linfáticos y alveolos. Ello se traduce en numerosas formaciones quísticas que afectan de manera bilateral a los pulmones¹.

A menudo se asocia con la esclerosi tuberosa (ET), una enfermedad caracterizada por múltiples tumores benignos: hamartomas en cerebro, corazón, pulmones e hígado, así como angiomiolipomas renales². El 30% de las mujeres con ET desarrollan cambios quísticos compatibles con LAM.

Un tercio de los casos se deben a mutaciones autosómicas dominantes del complejo ET (TSC1 o TSC2) que codifican las proteínas hamartina y tuberina, lo que se traduce en la activación de la vía del objetivo de la rapamicina (mTOR), produciendo una proliferación desregulada celular y de vasos sanguíneos, culminando en una remodelación pulmonar alterada^{1,3,4}.

Observación clínica

Presentamos el caso de una mujer de 21 años, de padres filipinos pero nacida en España. Exfumadora hasta hace cuatro años, actualmente vapeadora.

Diagnóstico prenatal de ET con la siguiente forma de presentación: angiofibromas faciales, manchas hipopigmentadas y collagenomas en tronco, así como angiomiolipomas hepáticos y renales, y *nevus* de Becker en miembro inferior derecho. Hasta el momento asintomática. Embarazo normal hace tres años, con hija nacida sana.

Acude a urgencias por dolor torácico y disnea de dos días de evolución. Se realiza una radiografía de tórax en la que se objetiva un neumotórax bilateral espontáneo, requiriendo por ello colocación de dos tubos torácicos (Fig. 1).

La ampliación del estudio en una tomografía computarizada de tórax (Fig. 2) identifica múltiples lesiones quísticas aéreas redondeadas de escasos milímetros, con afectación difusa y bilateral, así como varios nódulos pulmonares sólidos menores de 4-5 mm afectando a todos los lóbulos pulmonares, que dado el contexto clínico sugieren corresponder a una hiperplasia micronodular neumocitaria multifocal. Además, en los cortes del abdomen superior incluidos se observan múltiples lesiones sugerentes de angiomiolipomas hepáticos y renales, así como otras sugestiva de mielolipoma adrenal.

Tras la colocación de los drenajes, se resolvió el neumotórax derecho, pero en el hemitórax contralateral desarrolló fuga aérea persistente, por lo que requirió de bullectomía izquierda y abrasión pleural. Se confirma

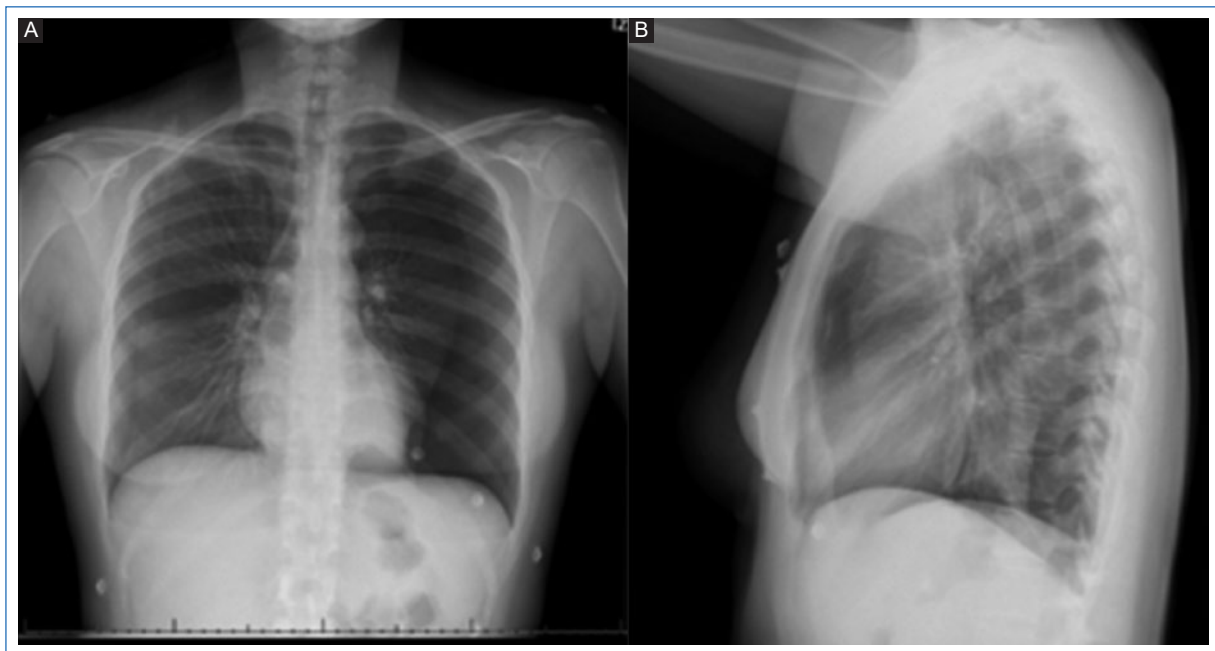


Figura 1. A: radiografía de tórax en proyección postero-anterior a su llegada a urgencias con hallazgo de neumotórax bilateral, más acusado en el lado izquierdo, llegando a ser completo. **B:** radiografía de tórax en proyección lateral a su llegada a urgencias con hallazgo de neumotórax bilateral, observándose hiperclaridad radiológica a nivel anterior y postero-inferior.

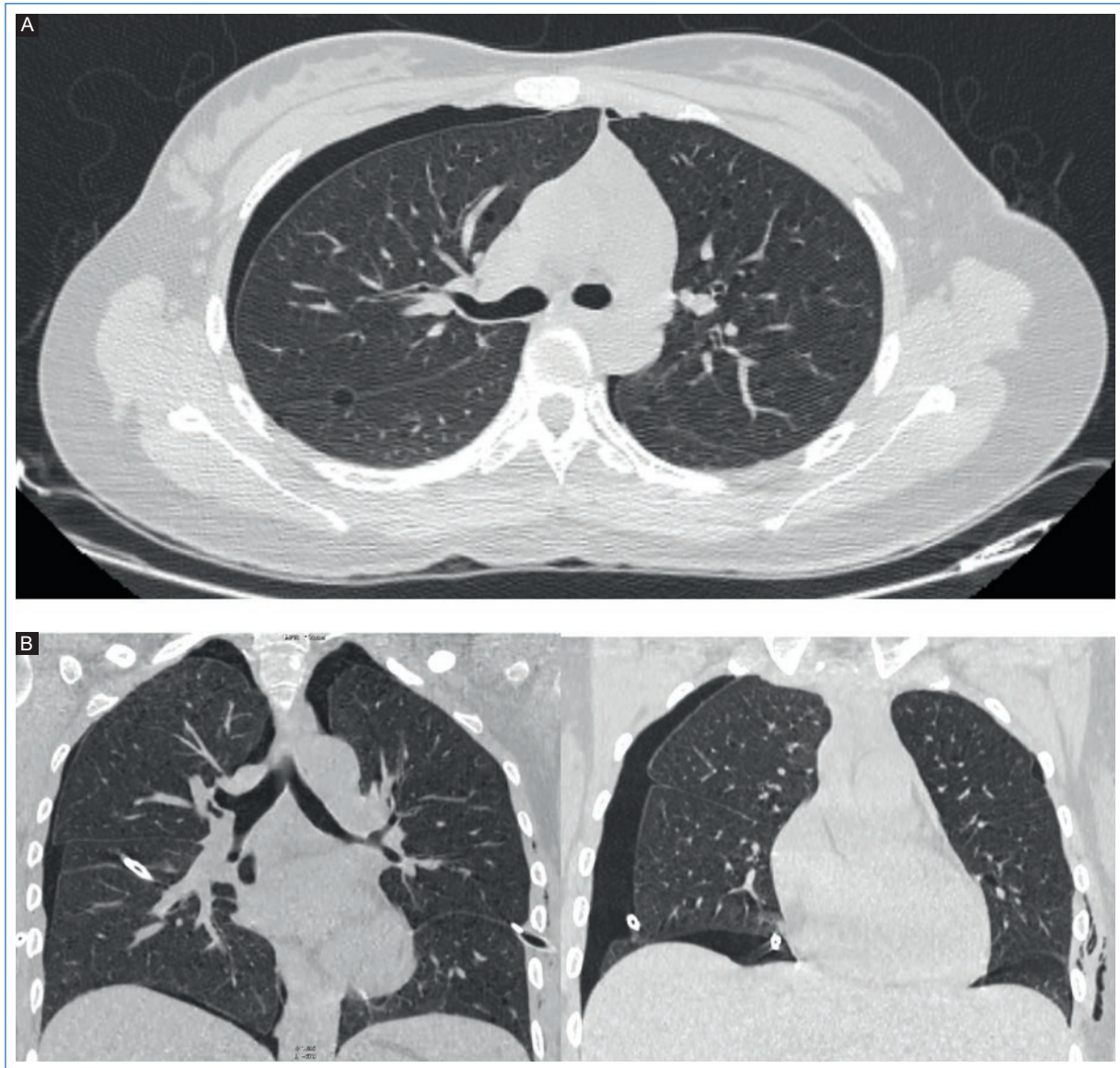


Figura 2. A: tomografía computarizada de tórax con contraste intravenoso. Corte axial a nivel subcarinal. Se observan múltiples quistes de paredes delgadas y dispersos de forma difusa por el parénquima pulmonar, así como neumotórax bilateral. **B:** corte sagital de misma tomografía a nivel de carina traqueal y de tronco pulmonar.

entonces el proceso clínico, con hallazgos histológicos compatibles con el diagnóstico clínico de linfangioleiomiomatosis (HMB-45 positivo, actina músculo liso positivo/CD1 a negativo, S100 negativo).

Se completa estudio con un perfil analítico de autoinmunidad que resulta negativo, así como con ecocardiograma transtorácico que resulta normal.

La paciente es dada de alta con evolución posterior favorable. Es revisada en consulta dos meses más tarde, donde refiere clínica de 24 horas de evolución sugerente de nuevo neumotórax derecho que finalmente se confirma radiológicamente el mismo día de la consulta. En este segundo episodio, la paciente

vuelve a requerir de una nueva bullectomía por desarrollar de nuevo fuga aérea persistente.

Tras el alta, se inicia tratamiento con sirolimús con buena tolerancia, con estabilidad clínica a los seis meses de la segunda intervención, realizando actualmente una vida normal sin limitación por su enfermedad, sin nuevos episodios de neumotórax.

Discusión

La LAM se trata de una enfermedad poco frecuente y compleja, que se asocia a la ET hasta en un 26% de los casos^{5,6}. Su presentación clínica

inicial puede ser como disnea progresiva, neumotórax o quilotórax⁷. En el caso de ser como neumotórax, los pacientes suelen tener mejor pronóstico⁵. En nuestro caso queremos destacar la necesidad de hasta dos bullectomías por desarrollo de fuga aérea persistente tras la retirada de los drenajes pleurales, condición que se da de manera más frecuente en pacientes con este tipo de patología⁸.

Su severidad y progresión pueden testarse con ayuda de pruebas de cuantificación en la tomografía, en las pruebas de función pulmonar (que suelen mostrar flujos y difusión reducidos), el test de la marcha, pruebas de esfuerzo cardiopulmonar y con la medición del factor de crecimiento endotelial⁹. En nuestro caso, por mayor disponibilidad se optó como pruebas complementarias para la evaluar su severidad por las pruebas de función pulmonar y el test de la marcha.

Como opciones médicas de tratamiento, dada la patogénesis explicada previamente, son útiles el sirolimús y el everolimús, dos inhibidores de mTOR. Estos consiguen estabilizar la función pulmonar y reducir el tamaño de los quistes pleurales y angiomiolipomas^{3,7,10,11}.

Financiamiento

La presente investigación no ha recibido ninguna beca específica de agencias de los sectores públicos, comercial o con ánimo de lucro.

Conflicto de intereses

Los autores declaran no tener ningún conflicto de intereses.

Consideraciones éticas

Protección de personas y animales. Los autores declaran que para esta investigación no se han realizado experimentos en seres humanos ni en animales.

Confidencialidad, consentimiento informado y aprobación ética. El estudio no involucra datos personales de pacientes ni requiere aprobación ética. No se aplican las guías SAGER.

Declaración sobre el uso de inteligencia artificial. Los autores declaran que no utilizaron ningún tipo de inteligencia artificial generativa para la redacción de este manuscrito.

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¿Es útil la ecografía en los pacientes con patología pulmonar intersticial?

Is ultrasound useful in patients with interstitial lung pathology?

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Resumen

Las enfermedades pulmonares intersticiales difusas son un grupo heterogéneo. El objetivo del trabajo es valorar la utilidad de la ecografía con las escalas existentes en la literatura para el cribado o el seguimiento de estos pacientes. Se analizó si existía asociación estadística entre las diferentes escalas radiológicas y las pruebas funcionales en una cohorte de 40 pacientes. Utilizando el índice Warrick se clasificó a los pacientes en 4 leves, 20 moderados y 16 graves, mientras que con la escala Buda para ecografía se clasificaron en 16 leves, 10 moderados y 14 graves, y por el score modificado en 34 leves y 6 moderados. Se observa una asociación estadísticamente significativa entre DLCO e índice Warrick ($p = 0,03$) y entre índice Warrick y score ecográfico ($p = 0.006$). Se coincide con la literatura en la relación entre DLCO, scores radiológicos y la asociación con la puntuación ecográfica; aun así, se necesitan más estudios para estandarizar criterios.

Palabras clave: Ecografía. Enfermedades pulmonar intersticial difusa. Escalas.

Abstract

Diffuse interstitial lung diseases are heterogeneous group. The objective is to assess the usefulness of ultrasound with the existing scores in the literature for the screening or monitoring of these patients. We analyzed whether there was a statistical association between the different radiological scores with the functional tests in a cohort of 40 patients. Using the Warrick scores, patients were classified as 4 mild, 20 moderate and 16 severe, while with the Buda ultrasound score they were classified as 16 mild, 10 moderate and 14 severe, while using the modified score as 34 mild and 6 moderate. A statistically significant association is observed between DLCO and Warrick index ($p = 0.03$) and between Warrick index and ultrasound score ($p = 0.0006$). The relationship between DLCO and radiological scores and the association with ultrasound scoring is consistent with the literature; however, more studies are needed to standardize ultrasound criteria.

Keywords: Ultrasound. Diffuse interstitial lung disease. Scores.

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Introducción

Las enfermedades pulmonares intersticiales difusas (EPID) son un grupo heterogéneo de enfermedades resultantes del daño parenquimatoso pulmonar por patrones variables de inflamación y fibrosis. La histopatología incluye principalmente una alteración extensa de la arquitectura alveolar y de las vías respiratorias, con cambios en el compartimento pulmonar intersticial. Las EPID se pueden dividir en las de causas o asociaciones conocidas, como la neumoconiosis, las relacionadas con conectivopatías y la neumonitis por hipersensibilidad; la sarcoidosis y las neumonías intersticiales idiopáticas representan las de etiología desconocida, entre otras¹.

El diagnóstico se basa en la combinación de datos clínicos, funcionales, radiológicos e histológicos. La tomografía computarizada de alta resolución (TCAR) de tórax se considera el método de referencia no invasivo para la evaluación, la cuantificación y el seguimiento. Sin embargo, la TCAR se ve limitada por su coste, su disponibilidad y los riesgos de la radiación ionizante. Por ello, la realización periódica de TCAR debe considerarse prudentemente y de manera individualizada².

Durante la última década, la ecografía pulmonar ha surgido como un método complementario a la TCAR para evaluar las EPID. La presencia de múltiples líneas B bilaterales difusas representa el sello ecográfico del síndrome intersticial pulmonar. Este describe un grupo diverso de condiciones clínicas, desde el edema pulmonar hasta la fibrosis pulmonar, entre otras.

A pesar de la utilidad de la ecografía pulmonar en esta patología, todavía no existen criterios estandarizados. Además, solo en el 21,22% de los casos se emplea la ecografía para su estudio³.

El objetivo es analizar las diferentes escalas existentes en la literatura y su aplicabilidad en el cribado de pacientes con sospecha de patología intersticial.

Método

Estudio de una cohorte de 40 pacientes derivados a la consulta especializada de enfermedades intersticiales ante la sospecha de dicha patología. El reclutamiento fue de enero a diciembre de 2023.

A todos los pacientes se les realizaron pruebas funcionales completas, tomografía computarizada y ecografía pulmonar. La ecografía se realizó de manera sistematizada, por un mismo neumólogo con experiencia, dividiendo el tórax en seis cuadrantes (dos anteriores, dos laterales y dos posteriores) y analizando el número de líneas B y las características de la línea pleural (Fig. 1).

Los casos se clasificaron como leves, moderados y graves según la escala Warrick para tomografía computarizada de tórax y según la escala Buda y score modificado para la ecografía (Tabla 1).

Resultados

El 57,5% de la cohorte eran varones, con una edad media de $71,6 \pm 9,78$ años. Todos presentaban sintomatología respiratoria, siendo la más frecuente la disnea. La patología intersticial más frecuente fue la fibrosis pulmonar idiopática, presente en el 55% de los pacientes, seguida de una miscelánea (27,5%) con neumonitis por hipersensibilidad, el 12,5% con EPID asociada a artritis reumatoide y el 5% con sarcoidosis.

Las pruebas funcionales están recogidas en la tabla 2. Radiológicamente, el 67,5% presentaban un patrón compatible con neumonía intersticial usual y el resto un patrón de neumonía intersticial no específica. Al aplicar el índice radiológico Warrick se clasificó a los pacientes en 4 leves, 20 moderados y 16 graves. Según la escala ecográfica Buda hubo 16 leves, 10 moderados y 14 graves, y por el score modificado fueron 34 leves y 6 moderados.

En el análisis de los datos se observó que existía una asociación estadísticamente significativa entre la prueba de difusión de monóxido de carbono (DLCO, *diffusing capacity of the lungs for carbon monoxide*) y el índice Warrick ($p = 0,003$), mientras que no existía correlación estadísticamente significativa entre la capacidad vital forzada (CVF) y el índice ($p = 0,56$). Al comparar los mismos datos (DLCO y CVF) con el score modificado tampoco se encontraron diferencias ($p = 0,14$ y $p = 0,12$, respectivamente). Además, tampoco existe una asociación estadísticamente significativa entre tener una DLCO o una CVF menores del 80% y una puntuación patológica mediante ecografía empleando el score modificado (prueba χ^2 , $p = 0,44$ y $p = 0,39$, respectivamente), aunque sí existe una correlación significativa entre la puntuación Warrick y la puntuación ecográfica mediante el score modificado ($p = 0,0006$). No hubo asociación estadísticamente significativa entre las diferentes variables con la escala Buda ($p > 0,005$).

Discusión

La patología intersticial pulmonar es un grupo muy heterogéneo de enfermedades⁴. El curso de muchas de ellas es hacia un daño pulmonar irreversible provocando fibrosis y disnea incapacitante, con deterioro de

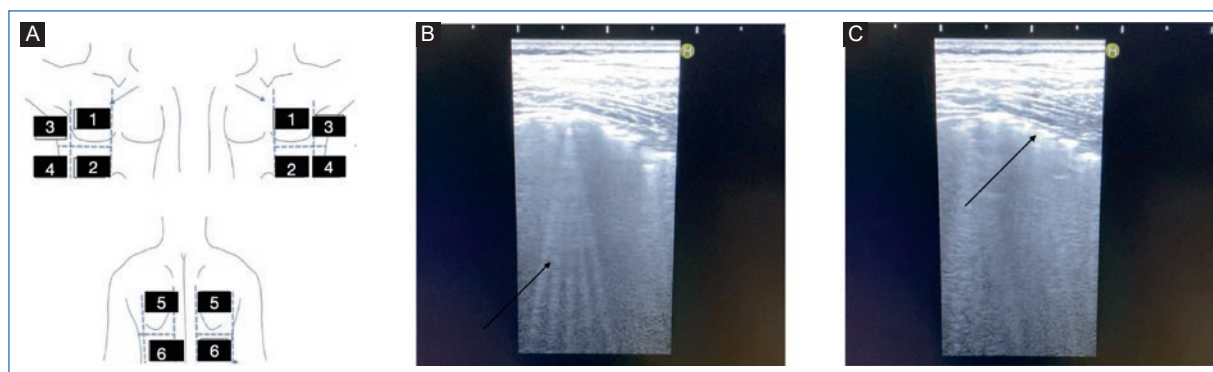


Figura 1. **A:** representación de los cuadrantes ecográficos estudiados. **B:** imagen ecográfica en la que se observa un aumento de líneas B en un campo (flecha). **C:** imagen ecográfica en la que se objetiva una línea pleural fragmentada (flecha).

Tabla 1. Variables de las diferentes escalas

Buda score	Score modificado ecografía	Índice Warrick
1 punto: pulmón blanco 2 puntos: línea pleural irregular 3 puntos: < 3 líneas B 4 puntos: < 4 líneas B 5 puntos: línea pleural fragmentada 6 puntos: signo de cometa	Líneas B: 0: 0 puntos < 4: 1 punto 4-6: 2 puntos > 6: 3 puntos	1 punto: vidrio deslustrado 2 puntos: pleural irregular 3 puntos: línea subpleural 4 puntos: panalización 5 puntos: quistes subpleurales
1 área: 1 punto 2 áreas: 2 puntos 3 áreas: 3 puntos	Línea pleural: Normal: 0 puntos Degradada: 1 punto Irregular: 2 puntos Fragmentada: 3 puntos	1-3 segmentos: 1 punto 4-9 segmentos: 2 puntos > 9 segmentos: 3 puntos
Interpretación: 3-14 puntos: leve fibrosis 15-20 puntos: moderada fibrosis 21-35: grave fibrosis	Interpretación: 0-34 puntos: leve 34,5-64 puntos: moderado > 64: grave	Interpretación: 0: normal < 8 puntos: leve 8-15 puntos: moderado > 15 puntos: grave

Tabla 2. Valores medios de las pruebas funcionales respiratorias

	Cohorte global	Pacientes FPI	Pacientes EPID no FPI
CVF	2346,5 ± 952,6 ml	2679 ± 209,93 ml	2045 ± 203,02 ml
DLCO	3,33 ± 1,73 ml/min/mmHg	3,59 ± 0,447 ml/min/kg	3,03 ± 0,32 ml/min/mmHg

CVF: capacidad vital forzada; DLCO: prueba de difusión de monóxido de carbono; EPID: enfermedad pulmonar intersticial difusa; FPI: fibrosis pulmonar idiopática.

la calidad de vida y mortalidad precoz. Por ello, es importante la búsqueda de nuevas herramientas que permitan un diagnóstico temprano y un seguimiento con una monitorización sencilla⁵.

En la literatura, los estudios sobre ecografía y EPID comenzaron con Picano y Gargani⁶, quienes proponen > 5 líneas B como punto de corte para el diagnóstico

de EPID, y con Gargani et al.⁷, que proponen el corte en > 10 líneas B.

Vicente-Rabaneda et al.⁸ realizaron un metaanálisis en 2021 y observaron una variabilidad entre los diferentes estudios en las áreas consideradas, pero encontraron asociación entre diversas variables, como la relación entre ecografía e índice Warrick ($p < 0.001$)

o el número de líneas B con DLCO y CVF ($p = 0.0001$ en ambos).

Nosotros hemos observado correlación entre el score modificado y el índice Warrick, en concordancia con el estudio realizado por Zhang et al.⁹, en el que analizaron 60 pacientes con las mismas escalas (Warrick, Buda y score modificado) y llegaron a la conclusión de que el score modificado para la ecografía era más útil, junto con el índice Warrick. Sin embargo, no se encontró asociación entre diferentes datos funcionales (DLCO, CVF) y los datos ecográficos, pero sí con el índice Warrick ($p = 0,003$), a diferencia de otros estudios, como el de Huang et al.¹⁰, en el que se incluyeron 88 pacientes y se objetivó una asociación significativa entre los datos ecográficos y la alteración de todos los parámetros funcionales (CVF, volumen espiratorio forzado el primer segundo y DLCO).

En conclusión, el uso de la ecografía pulmonar en las EPID puede ayudar al cribado de estas enfermedades en etapas precoces o ser útil durante su seguimiento, por su fácil disponibilidad y rapidez de realización; aun así, se necesitan estudios más robustos, con mayor número de pacientes, para obtener unos criterios estandarizados y valorar la posibilidad de incorporar la ecografía en los algoritmos diagnósticos.

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Consideraciones éticas

Protección de personas y animales. Los autores declaran que para esta investigación no se han realizado experimentos en seres humanos ni en animales.

Confidencialidad, consentimiento informado y aprobación ética. El estudio no involucra datos personales de pacientes ni requiere aprobación ética. No se aplican las guías SAGER.

Declaración sobre el uso de inteligencia artificial. Los autores declaran que no utilizaron ningún tipo de inteligencia artificial generativa para la redacción de este manuscrito.

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Endobronchial metastasis as a diagnosis of melanoma: an exceptional case

Metástasis endobronquial como diagnóstico de melanoma: un caso excepcional

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We present the case of a 42-year-old male, former smoker, referred due to an episode of hemoptysis. The computed tomography (CT) scan revealed a pulmonary mass in the left lower lobe. During bronchoscopy, an endobronchial lesion was identified in the anterior segment of the left lower lobar bronchus (Fig. 1), which led to the diagnosis of a malignant melanoma (MM) as the only known manifestation of the tumor. An extension study with PET-CT showed metastatic foci in the abdomen, an inguinal subcutaneous nodule, and bone. Ocular and cutaneous melanoma were excluded. Treatment with dabrafenib + trametinib was initiated and continued for a year until he finally died of cerebral hemorrhage secondary to brain metastasis of melanoma.

MM can arise in the skin, mucous membranes, or other pigmented areas, comprising less than 0.01% of malignant lung tumors¹. Primary bronchial metastases are rare, usually derived from an occult MM or another primary site. Diagnosis often occurs following symptoms of central airway obstruction or incidental findings, without evident cutaneous lesions. Palliative endoscopic treatment is considered for significant airway obstruction. The prognosis of MM is generally unfavorable, with short survival. BRAF gene inhibitors are a therapeutic option in advanced melanomas. The median survival post-detection of pulmonary metastases is short, although surgical resection may improve survival².

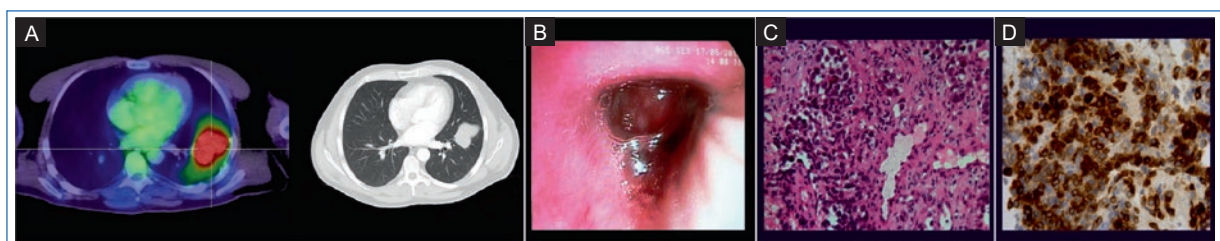


Figure 1. **A:** PET-CT and CT: lung mass in left lower lobe of 5.6 cm (SUV max 10.8). **B:** endobronchial mass completely occluding the anterior segment of the left lower lobar bronchus. **C:** hematoxylin-eosin: discohesive nidus of large and atypical cells in bronchial stroma. **D:** immunohistochemistry shows intense positivity with Melan A in tumor cells.

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None.

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Protection of humans and animals. The authors declare that no experiments involving humans or animals were conducted for this research.

Confidentiality, informed consent, and ethical approval. The study does not involve patient personal

data nor requires ethical approval. The SAGER guidelines do not apply.

Declaration on the use of artificial intelligence. The authors declare that no generative artificial intelligence was used in the writing of this manuscript.

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Birt-Hogg-Dubé syndrome (BHDS) detected incidentally

Síndrome de Birt-Hogg-Dubé (SBHD) detectado incidentalmente

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A 60-year-old woman with no past history of interest had an incidental finding of cystic lung lesions and renal lesion on computed tomography (Fig. 1). The patient was asymptomatic, and the physical examination was unremarkable. Laboratory and pulmonary function tests were normal. Biopsy of the renal lesion was attempted, but due to its small size, it was not possible to obtain an adequate sample. In the genetic study, the germ line variant c.1579_1580insA in the *FLCN* gene was detected, finally diagnosed as a Birt-Hogg-Dubé syndrome (BHDS). A genetic study was recommended for first-degree relatives. A clinical-radiological follow-up was performed, with no symptoms or progression of the lesions.

In a patient with pulmonary cystic lesions, the main differential diagnosis should include: pulmonary Langerhans cell histiocytosis (LCH), lymphangioleiomyomatosis (LAM), Birt-Hogg-Dubé syndrome (BHD), interstitial lymphoid pneumonia (ILP) and pneumonia due to *Pneumocystis jirovecii*. In LCH and *Pneumocystis jirovecii* pneumonia, cysts predominate in upper fields, and in the latter the most characteristic features are ground-glass opacities. Cysts in ILP are distributed in basal areas together with a ground-glass or reticulonodular pattern¹⁻². In BHD and LAM, the cysts have a bibasal location, and there may be cutaneous and renal lesions².

The main differential diagnosis in this case, is with lymphangioleiomyomatosis (LAM). However, in LAM the cysts are usually multiple, rounded, 2-10 mm in size, symmetrically distributed and without any lobar

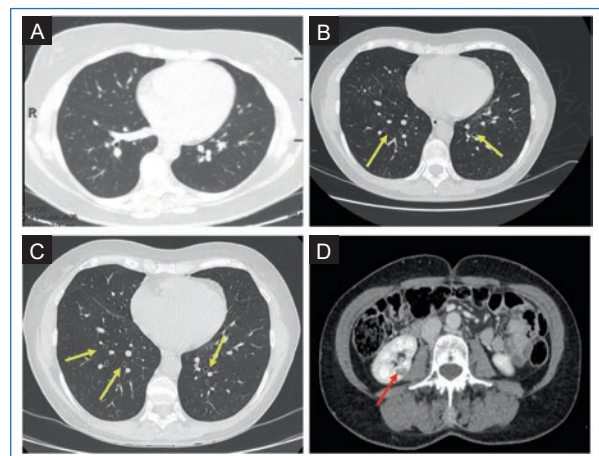


Figura 1. A: computed tomography (CT) scan performed 1 year before. It shows normal pleuropulmonary and airway structures. **B-C:** chest CT scan performed 2 years later. Several cystic lesions are observed within the lung parenchyma, a few millimeters in size, predominantly bilobar (yellow arrow). **D:** hypodense lesion in the lower pole of the right kidney (red arrow).

predominance¹⁻². Whereas most patients with BHDS present with < 20 cysts, < 10 mm in diameter, irregular and bibasal/paramediastinal in location²⁻³. And although both pathologies typically affect young adults, the radiological lesions of BHDS typically present between 40-50 years of age¹. This allows us to conclude that sometimes radiological patterns can be indistinguishable, so imaging tests are not sufficient and diagnostic confirmation is required.

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from patients, and received approval from the Ethics Committee. The SAGER guidelines were followed according to the nature of the study.

Declaration on the use of artificial intelligence.

The authors declare that no generative artificial intelligence was used in the writing of this manuscript.

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Bulla pulmonar creciente en un paciente con enfermedad pulmonar intersticial inflamatoria: a propósito de un caso

Growing pulmonary bulla in a patient with inflammatory interstitial lung disease: a case report

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Presentamos el caso de una mujer de 70 años, originaria de Ecuador y sin antecedentes médicos relevantes. Durante su ingreso hospitalario por una afección no relacionada, se detectó un patrón reticulonodular en una radiografía de tórax. Se completó el estudio con tomografía computarizada de tórax, el cual reveló la presencia de una enfermedad pulmonar intersticial crónica con signos incipientes de fibrosis, además de una gran bulla en el lóbulo inferior derecho (Figs. 1 y 2).

Se ampliaron pruebas complementarias, incluyendo una espirometría que mostró una capacidad de difusión del 58%, con los demás parámetros dentro de los límites normales. Los análisis microbiológicos e inmunológicos no revelaron hallazgos de interés. La criobiopsia pulmonar fue compatible con neumonía intersticial no específica, sin evidencia de enfermedad autoinmunitaria sistémica.

El tratamiento inicial consistió en la administración de rituximab y corticosteroides, seguido de micofenolato como terapia de mantenimiento. Sin embargo, meses después, la paciente reingresó debido a un neumotórax secundario a la rotura de la bulla, lo que requirió la colocación de un drenaje endotorácico. Tras la retirada del drenaje, la paciente presentó una recidiva del neumotórax, por lo que se realizó una pleurodesis con talco. A pesar de este procedimiento, la paciente

sufró una nueva recidiva y se le dio el alta con una válvula de Heimlich, la cual fue retirada de forma ambulatoria, persistiendo una bulla residual.

Durante los 2 años siguientes, la paciente experimentó un empeoramiento funcional y radiológico significativo, lo que condujo al diagnóstico de fibrosis pulmonar progresiva. Se decidió entonces añadir nintedanib como tratamiento antifibrótico¹⁻³.

La bulla residual en el lóbulo inferior derecho continuó creciendo, aumentando la sintomatología de la paciente y provocando reingresos recurrentes. La resección quirúrgica de la bulla fue descartada debido al alto riesgo quirúrgico. Una broncoscopia reveló una significativa malacia del lóbulo inferior derecho, con la pared vencida hacia la luz y segmentos cerrados, sugiriendo un probable efecto válvula. En consecuencia, se concluyó que la paciente no era candidata para un tratamiento endobronquial.

La neumonía intersticial no específica es una condición clínica dentro del espectro de las enfermedades pulmonares intersticiales difusas (EPID). La coexistencia de bullas pulmonares en pacientes con EPID es una complicación infrecuente⁴, pero significativa, ya que puede predisponer a neumotórax espontáneos recurrentes, como se observó en este caso.

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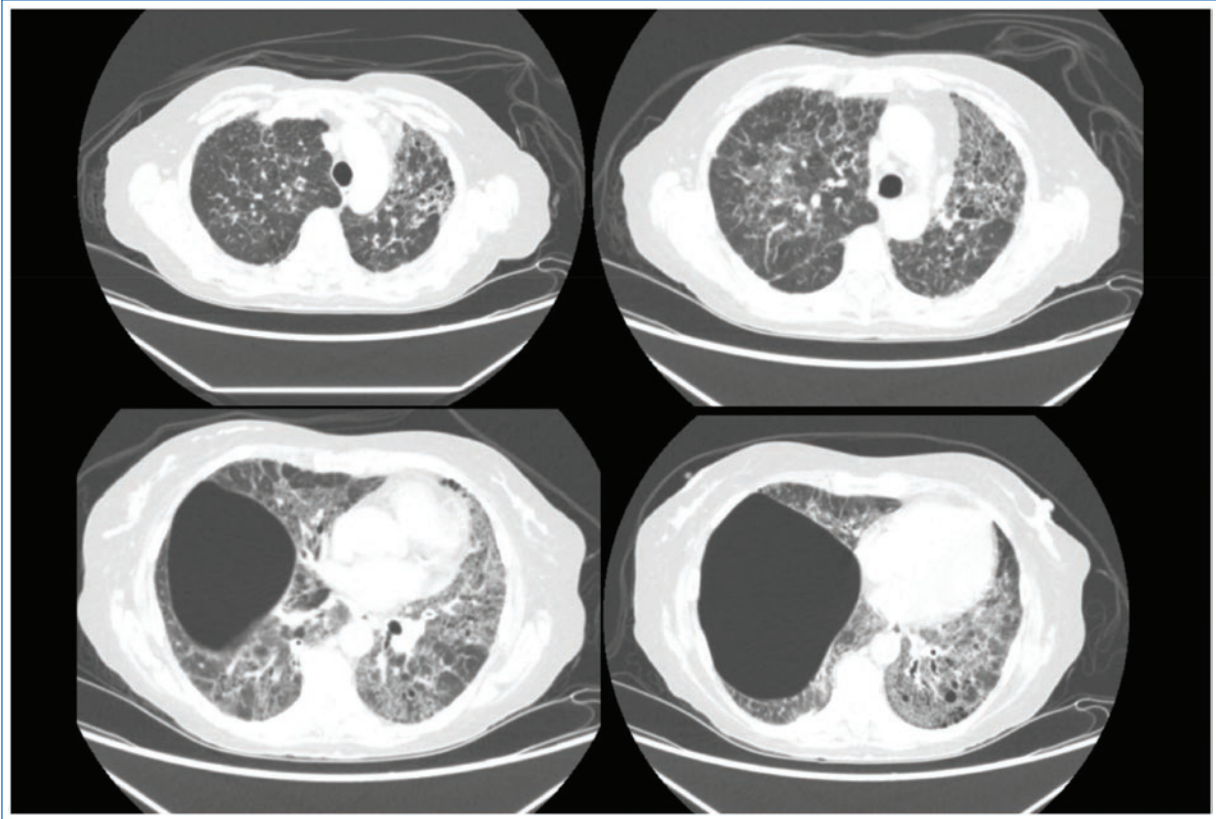


Figura 1. Tomografía computarizada en con cortes axiales a lo largo del pulmón que muestra opacidades en vidrio esmerilado, pequeñas bronquiectasias y un incipiente panal periférico. Estos hallazgos son compatibles con neumonía intersticial no específica³ fibrótica temprana, coincidiendo con una bulla de 8.4 cm en el lóbulo inferior derecho.

Las bullas pueden actuar como reservorios para patógenos oportunistas⁵, tales como *Aspergillus*, llevando a la formación de aspergilomas, que requieren manejo específico. También existe la posibilidad, aunque rara, de transformación neoplásica dentro de las bullas, lo cual debe ser considerado en pacientes con EPID crónica.

En este caso, el manejo de las complicaciones bullosas resultó desafiante. A pesar de la intervención con pleurodesis y la utilización de una válvula de Heimlich, la paciente continuó presentando recidivas del neumotórax. La opción de resección quirúrgica fue descartada debido al alto riesgo, y el tratamiento endobronquial no fue viable por la malacia significativa del lóbulo afectado.

La progresión de la fibrosis pulmonar y el incremento de la sintomatología llevaron a la adición de nintedanib, un agente antifibrótico que ha demostrado eficacia en

la ralentización de la progresión de la fibrosis en la EPID.

Este caso destaca la importancia de un enfoque integral y multidisciplinario en el manejo de los pacientes con EPID y complicaciones bullosas. A pesar de las intervenciones médicas y quirúrgicas, el pronóstico permanece reservado debido a la progresión de la fibrosis y las complicaciones asociadas.

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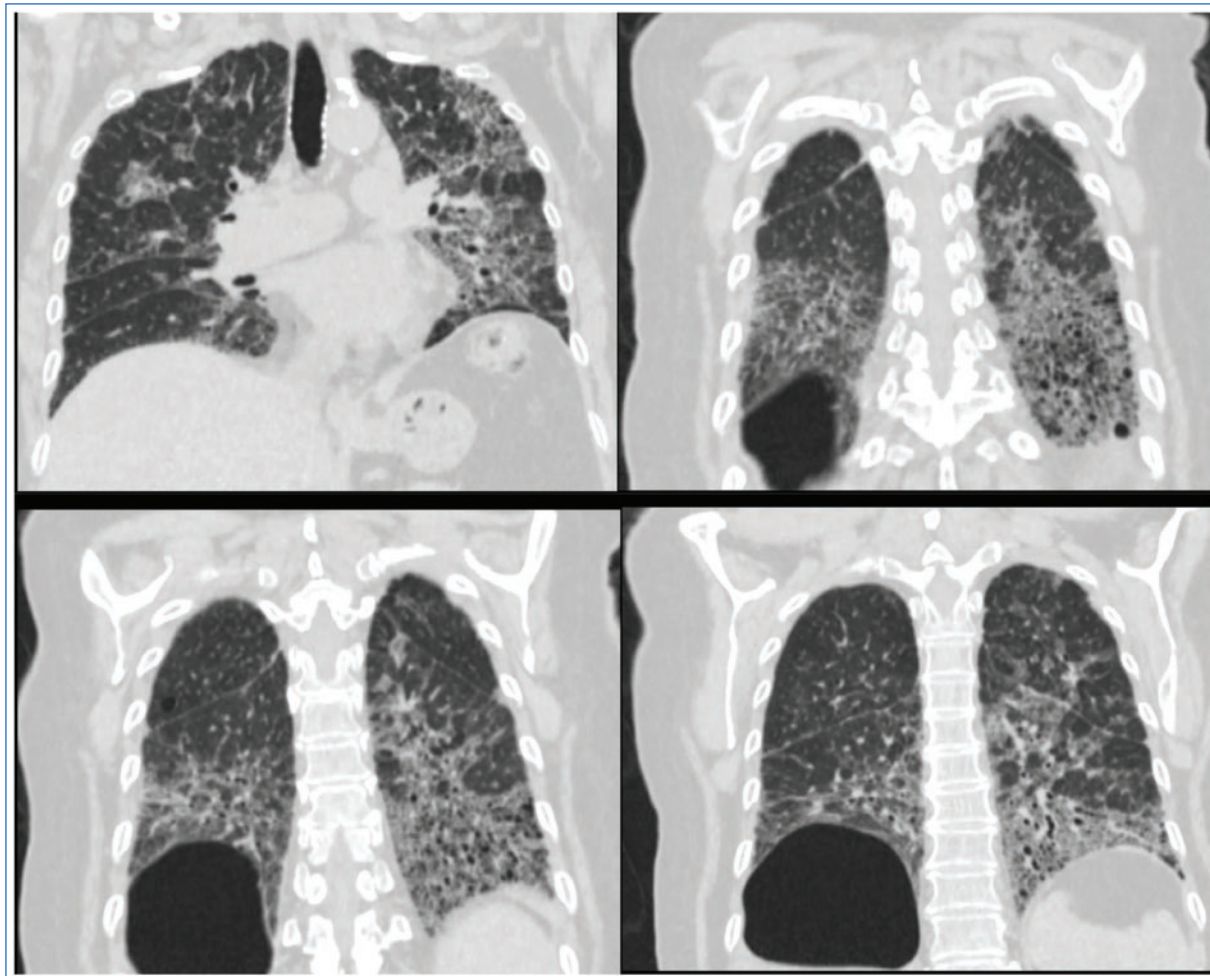


Figura 2. Cortes coronales de la misma tomografía computarizada de la figura 1.

Consideraciones éticas

Protección de personas y animales. Los autores declaran que para esta investigación no se han realizado experimentos en seres humanos ni en animales.

Confidencialidad, consentimiento informado y aprobación ética. El estudio no involucra datos personales de pacientes ni requiere aprobación ética. No se aplican las guías SAGER.

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