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The impact of new bronchial drainage device in a complex form of respiratory disease

Rinedy Lisumbu Mondonga[1,*], Florin Mihălțan[1,2], Aida Andreea Fănică[1]

[1]Secția Pneumologie III, Institutul Național de Pneumoftiziologie Marius Nasta, Bucharest, Romania
[2]Universitatea de Medicină și Farmacie Carol Davila, Bucharest, Romania

Abstract

English:

Drainage of secretions in patients with severe lung damage is a challenge. In many cases, due to the significant reduction in muscle mass, difficulties of this nature arise. We present the case of an 82-year-old patient, vaccinated against Severe acute respiratory syndrome coronavirus 2 (SARS COV2), anti-influenza, anti-pneumococcal and anti-haemophilus, former smoker, with no significant exposure to known professional respiratory noxes, with severe comorbidities (cardiac, ophthalmological, rheumatological, neurological, and diabetological) and a congenital pulmonary malformation – right lung hypoplasia (diagnosed in 2008), extensive cystic and varicose bronchiectasis, probably also in a congenital context, with progressive collapse of the right parenchyma and displacement of the mediastinum, complicated with recurrent respiratory infectious episodes with pathogens – Gram-negative germs, anaerobic germs, and repetitive low-amount haemoptysis for 3 years, as well as severe restrictive respiratory dysfunction. Currently, the patient is under bronchodilator treatment, and home oxygen therapy. For about 3 years, regular modern drainage sessions using the studied device have been introduced. Patient adherence was good. The benefits brought by this device on the clinical development of the patient were: efficient drainage of secretions, improvement of the quality of life, and prolongation of survival in a patient with complex pulmonary pathology and multiple comorbidities in development for 13 years. This new therapeutic drainage option has been competing with pre-existing ways (Cough Assist) for several years, demonstrating its multiple benefits in patients with chronic respiratory failure and severe comorbidities.

Keywords

bronchiectasis • Swyer–James–MacLeod Syndrome • new bronchial drainage device (SIMEOX)

Impactul unui nou tip de aparat de drenaj bronșic într-un caz de patologie complexă respiratorie

Rezumat

Romanian:

Drenajul secrețiilor la pacienții cu afectare pulmonară severă reprezintă o provocare. Reducerea semnificativă a masei musculare poate induce dificultăți de drenaj. Prezentăm cazul unei paciente de 82 de ani, vaccinată antiSARS COV2, antigripal, antipneumococic si antihaemophillus, fosta fumătoare, fără expunere semnificativă la noxe respiratorii profesionale, cunoscută cu comorbidități severe (cardiace, oftalmologice, reumatologice, neurologice și diabetologice) și o malformație congenitală pulmonară - hipoplazie pulmonară dreaptă (diagnosticată în 2008), bronșiectazii chistice și varicoase extensive, probabil tot în context congenital, cu colabarea progresivă a parenchimului drept și deplasarea mediastinului, ceea ce a condus la episoade infecțioase respiratorii recidivante cu agenți patogeni – germeni Gram negativi, germeni anaerobi, hemoptizii de cantitate mică repetitive de 3 ani, precum și o disfuncție respiratorie restrictivă severă. Pacienta se află sub tratament bronhodilatator și oxigenoterapie la domiciliu. De circa 3 ani au fost introduse ședințe regulate de drenaj modern cu dispozitivul Simeox. Aderența pacientei a fost una bună. Beneficiile aduse de acest aparat asupra evoluției clinice a pacientei au fost: drenajul eficient al secrețiilor, îmbunătățirea calității vieții și prelungirea supraviețuirii la o pacientă cu patologie pulmonară complexă și multiple comorbidități în evoluție de 13 ani. Noua opțiune terapeutică de drenaj a intrat în competiție de câțiva ani cu modalitățile preexistente (Cough Assist), demonstrându-și beneficiile multiple la pacienții cu insuficiență respiratorie cronică și comorbidități severe.

Cuvinte-cheie

bronșiectazie • sindromul Swyer–James–MacLeod • aparat de drenaj bronsic (SIMEOX)

*Corresponding author: Rinedy Lisumbu Mondonga

E-mail: rilis1991@gmail.com

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Introduction

Swyer–James–MacLeod syndrome (SJMS), also known as Swyer–James syndrome or hyperlucid lung syndrome, is an uncommon syndrome of unilateral functional hypoplasia of the pulmonary vasculature together with emphysema sometimes associated with bronchiectasis (1,2). The condition was first described in the 1950s in different continents, by the pulmonologist William Mathieson Macleod in England (1954) and the doctors Paul Robert Swyer and George James in Canada (1953) (3,4).

This rare lung condition (with a prevalence of 0.01% in a series of 17.450 reviewed chest radiologic images (5)) is characterised by the radiographic hyperlucent appearance of a single lobe or an entire lung. It is thought to occur as a complication of post-infectious bronchiolitis obliterans in childhood.

Bronchiolitis obliterans describes a clinical syndrome of small airway obstruction that is irreversible with the use of bronchodilators. It is triggered by damage to the bronchiolar epithelium and possibly the adjacent alveoli. This damage, which occurs during early childhood, prevents the normal development of the alveolar ducts.

The damage can be caused by a number of factors: respiratory infections playing a major role, as well as reduced pulmonary blood flow in SJMS. Although full recovery is possible, the repair process can lead to excessive proliferation of granulation tissue leading to narrowing or complete obliteration of the small airways. Recurrent unilateral small airways obstruction involving terminal bronchi and bronchioles that occurs early in life triggers an inflammatory response, leading to the development of submucosal fibrosis (6). The resulting luminal irregularity further contributes to airway occlusion, becoming more permanent. This, in turn, causes alveolar air trapping, eventually leading to emphysema, which is defined as a permanent overdistension of the distal air spaces to the terminal bronchioles (1). Destruction of the alveolar wall results in loss of elastic recoil, causing the airway to collapse during expiration and air capture. Therefore, the typical appearance of unilateral hyperlucency can be found on chest radiographs.

The other, perhaps most important factor responsible for radiographic hyperlucency of a single hemithorax is reduced pulmonary blood flow. Fibrosis of the interalveolar septa leads to obstruction of the pulmonary capillary bed and reduces blood flow to the main branches of the pulmonary artery. In addition, hyperinflation of the alveoli, as seen in emphysema, creates additional mechanical resistance to blood flow through the capillary bed. Since this process occurs early in life, arterial development is thwarted, resulting in a hypoplastic pulmonary vasculature (7).

In addition, reduced ventilation of the lung or diseased segment also results in compensatory vasoconstriction of the pulmonary vessels, leading to functional restriction of arterial flow, which further contributes to radiographic hyperlucency (1,4,8). Functional hypoplasia of the bronchial vasculature can affect the growth of the affected lung, resulting in a small or normal-sized lung (3). In some patients with SJMS, bronchiectasis may develop adjacent to bronchiolitis obliterans and is likely a sequela of chronic infections.

Causative agents implicated in recurrent lung infections include viruses (measles, respiratory syncytial virus, influenza A, Paramyxovirus *Morbillivirus*, adenovirus types 3, 7, and 21) and bacteria (*Bordetella pertussis, Mycobacterium tuberculosis, Mycoplasma pneumoniae*) (4,5,9–11).

Some patients are diagnosed with SJMS in childhood, and others by chance in adulthood when they undergo investigations for another reason (e.g. asthma symptoms or recurrent chest infections) (12). This rare condition presents a diagnosis challenge because its presentation can be nonspecific and mimic other respiratory conditions.

Patients with this condition can display symptoms, or can be asymptomatic (6). Symptomatic patients often have recurrent lung infections and exertional dyspnoea (1,13,14). The most frequent symptoms are: wheezing, dyspnoea on exertion, reduced tolerance to exertion, and cough (often productive) sometimes with haemoptysis and pleuritic pain radiating to the heart (3).

During the clinical examination, a visible reduction in chest expansion, hyperresonance to percussion, and a decrease in respiratory sounds on the affected side are found (3). In cases where the mediastinum is displaced to the affected side, the beating of the apex may also be displaced (6,15).

The disease can be confused with both congenital and acquired conditions, for example: congenital lobar emphysema, localised interstitial pulmonary emphysema, pulmonary development abnormalities, Poland syndrome, and pneumothorax in severe cases.

Computed tomography (CT) of the chest is usually the modality of choice used to assess the extent and distribution of disease. It can present with emphysema, bullae, bronchiectasis, atelectasis, and scarring of fibrotic nature. Bronchiectasis, present in approximately 30% of patients with SJMS, can be cylindrical or saccular (6).

CT pulmonary angiography is not essential for diagnosis but would demonstrate hypoplasia of the pulmonary vasculature in the affected lung or segment (6).

Ventilation/perfusion scintigraphy may reveal a unilateral ventilation and perfusion defect corresponding to the diseased area. Some advocate it as a first-line investigation (after a

chest radiograph) in the evaluation of patients with suspected SJMS (16).

SJMS usually presents with a restrictive pattern on pulmonary function testing. Patients with bronchiectasis may have a mixed restrictive–obstructive or even predominantly obstructive defect, in which airflow obstruction is irreversible with bronchodilators (5,17,18). Total lung capacity (TLC) may be slightly reduced (18).

Tests such as alpha-1 antitrypsin deficiency are useful to rule out rare inherited causes of bronchiectasis/emphysema (6).

Diagnosis criteria for this syndrome require one of the following (19):

- Unilateral loss of lung volume with hyperlucency demonstrated by chest X-ray;
- Unilateral reduction of vasculature on chest tomography;
- Unilateral perfusion loss on Tc-99m technetium chest scintigraphy.

Management of SJMS is patient-centred and delivered using the interprofessional team approach. Conservative management is the main treatment.

Prevention and prompt treatment of recurrent respiratory infections is necessary as primary prevention. Thus, it is recommended that patients be vaccinated against pneumococcus and influenza (6). Mucolytics may be useful in patients with bronchiectasis. Chest physiotherapy using techniques such as percussion and postural drainage is important to ensure effective sputum clearance (18). Corticosteroids and inhaled bronchodilators are also used (6). In cases of respiratory failure, long-term oxygen therapy may be indicated. Patients should also be referred for pulmonary rehabilitation to maximise respiratory function. Surgery is reserved for (1) patients with recurrent lung infections, (2) unresponsive patients, or (3) patients whose symptoms are not adequately controlled with optimal medical management.

The evolution of the disease is related to the presence of bronchiectasis. Patients with no bronchiectasis or with cylindrical bronchiectasis often have mild respiratory symptoms and tend to improve spontaneously (2). Those with saccular bronchiectasis, on the other hand, suffer from recurrent bouts of pneumonia and are most at risk for surgery.

Presentation of the case

We present the case of an 82-year-old patient, a former smoker, with no significant exposure to occupational respiratory noxes, with multiple associated conditions, in our clinic's care since 2008 when she presented to a pulmonology consultation complaining of cough with mucopurulent sputum and dyspnoea at small efforts.

In Table 1 the associated conditions of the patient are listed.

Table 1. Associated conditions of the patient.

Respiratory conditions	Chronic respiratory failure under oxygen therapy at home, right lung hypoplasia, extensive cystic and varicose bronchiectasis, episodes of recurrent respiratory infections, recurrent small haemoptysis, nonstaged COPD
Cardiac conditions	Grade II hypertension, ischaemic heart disease, paroxysmal atrial fibrillation converted to sinus rhythm under anticoagulant, beta-blocker and antiarrhythmic treatment, mild tricuspid insufficiency, mild aortic insufficiency, medium PH
Endocrinological conditions	Polynodular goitre, macronodular tumour formation located at the level of the right thyroid lobe
Metabolic conditions	Type II diabetes in treatment with oral antidiabetics
Ophthalmological conditions	Right eye cataract surgery
Neurological conditions	Parkinson's disease
Rheumatologic conditions	Bilateral coxarthrosis and lumbosacral spondylodysatrosis

COPD, chronic obstructive pulmonary disease.

At the time of the presentation, the patient had performed a native chest CT in another hospital unit, mentioning: right lung hypoplasia, cystic and varicose bronchiectasis, and mediastinal deviation to the right with compensatory hyperinflation of the left lung.

Additional paraclinical investigations were performed. Blood tests showed leukocytosis with neutrophilia, and increased inflammatory syndrome (PCR = 50 mg/dL), suggesting a bacterial infection. There was no renal failure, and serum glucose was within normal limits.

The chest X-ray in antero-posterior incidence and left profile (Figure 1) showed inhomogeneous opacities distributed in the right lung, apico-caudally disseminated hypertransparency areas with a tendency to converge in the lower half of the left hemithorax, traction to the right of mediastinum, trachea and aortic button, and compensatory left lung hyperinflation. On the left profile increased retrosternal hyperlucency was observed.

Complex lung function measurement (pletismography and measurement of diffusion capacity) was attempted, but the patient only managed to perform a simple spirometry, showing severe restriction.

Fiberbronchoscopy was performed, with the aim to aspirate the bronchial secretions, and to sample for bacteria, fungi, and possibly acid-fast bacilli, aiming for a targeted antibiotic treatment. Fiberbronchoscopy provided additional arguments for an acute respiratory infectious episode: tractions and deviations of bronchial statics, with deviation of the tracheo-bronchial axis to the right. Bilateral bronchiectatic territories are possible, but there is a patent suppurative aspect only on the right side predominantly in the lower lobe territory. Samples showed: Gram-negative germs sensitive to carbapenems, fluoroquinolones, and extended-spectrum penicillins, as well as fungi.

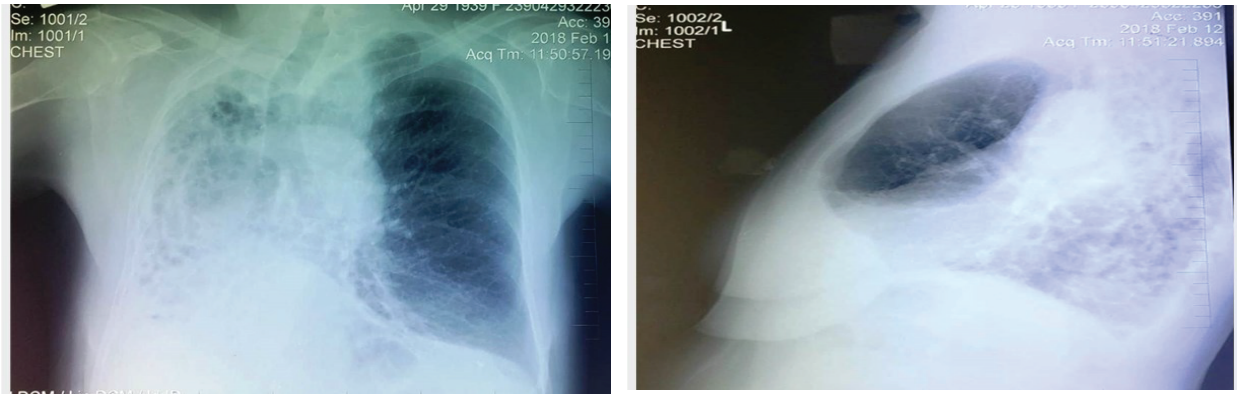


Figure 1. Antero-posterior incident radiological images and left profile.

During the hospitalisation, the patient was initially treated with empirical antibiotic treatment consisting of meropenem 2 g/day, 14 days; later fluconazol was added, loading dose 150 mg/day, for 3 days with monitoring of renal function, and later dose reduction to 50 mg/day for the next 7 days; expectorants, rehydration solutions, and vitamin therapy, together with cardiological, diabetes, and neurological medication and oxygen therapy 3–4 L/min through nasal cannula. Respiratory kinesitherapy sessions were also performed. The respiratory physiotherapist instructed the patient to be able to perform some breathing exercises at home. The evolution of the patient was favourable, with slow clinical improvement, with no adverse reactions.

The patient was discharged after 15 days of hospitalisation, with improved symptoms (dyspnoea on moderate efforts only), blood count within normal limits, no inflammatory syndrome, and two tests each negative for nonspecific flora and fungi. The diagnosis was SJMS, based on chest X-ray – the unilateral loss of lung volume with hyperlucency was demonstrated not only by the chest X-ray and the restrictive functional test but also by symptoms.

The patient's management at home was conservative, aiming to prevent and promptly treat recurrent respiratory infections, and thus incorporating respiratory physiotherapy to ensure the most efficient elimination of sputum and increase lung capacity. Inhaled bronchodilators were added (ipratropium bromide inhaler 20 µg, 2 puffs/day), and oxygen therapy 5 L/min through a facial mask.

Since the patient lived outside the city and encountered difficulties in travelling, contact was maintained by phone, or she was admitted in the local hospital during acute exacerbations. She performs respiratory physiotherapy sessions at home and exercises using an electric bicycle for hands and feet 1/month, and has been undergoing foot massage, Vitalzen type, since 2021.

During the long-term monitoring of our patient, for about 15 years, she also cooperated in a series of investigations, which were conducted in clinics/hospitals in her county (Table 2 and Figure 2), and underwent several therapeutic interventions in the local hospital.

Over time, the patient experienced recurrent respiratory infections. The incriminating germs were Gram-negative, anaerobes sensitive to carbapenems, fluoroquinolones, and broad-spectrum penicillins, an antibiogram similar to that in our hospital. No XDR infections were identified. Intravenous antibiotic treatment was often required. The patient is vaccinated against SARS COV2 (three doses), anti-influenza vaccine every season, anti-pneumococcal, anti-haemophilus, and the family takes all required protective measures when visiting the patient.

Considering the patient's age and reduced mobility, and knowing the large number of infectious exacerbations despite preventive treatment, we decided to add in the patient's management a medical device for drainage of pulmonary secretions, the new bronchial drainage device 'SIMEOX', with a twice daily use for 15 min.

The evolution was marked by an improvement of the symptoms with an increase in the clearance of the secretions, and the drainage was easy, which favoured the adherence to the new bronchial drainage device. The number of exacerbations has been reduced to 1/year currently versus approximately 5/year in previous years.

The new bronchial drainage device (SIMEOX) is a medical device developed by the French company PhysioAssist, about 5 years ago, inspired by sludge transport technologies (a typical example of techno-mimesis (20) for patients with respiratory conditions). It is intended to be used by adults and children aged >8 years who suffer from a lung disease and who have difficulties in clearing bronchial secretions (cystic fibrosis, chronic obstructive pulmonary disease (COPD),

Table 2. Imaging description over time.

2019	2020	2021
Native chest CT	**Chest CT with contrast**	**ANGIO CT (Figure 2)**
Marked thoracic asymmetry with traction of the mediastinum and the heart at the level of the right hemithorax, shows in the lower half of the right hemithorax multiple bronchiectasis with no fluid – pneumothorax; *Important*: Free main bronchi, left hemithorax with normal appearance, absence of mediastinal masses, no detectable lymph node of pathological importance at the mediastinal or axillary level.	Most likely agenesis of the right lung, with repeated respiratory infections that resulted in the formation of bronchiectasis. There are no suspicious nodular lesions or areas of alveolar filling in the left lung, which is hyperinflated. Some mediastinal adenopathies up to approx. 15 mm short axis at the precarinal level. No pleural or pericardial collections.	The right lung parenchyma collapsed, dysmorphic, distorted by the presence of fibrotic changes and extensive cystic bronchiectasis. Emphysema bubble located at the level of the collapsed lung parenchyma of 28/62 mm, which pulls on the ipsilateral mediastinal structures. Left pulmonary compensatory hyperventilation. With no visible systematised alveolar condensation areas, the pulmonary pattern with a mosaic appearance is most likely ventilation turbulence. Bands of fibrosis located at the level of the lingular segment. Vertebral haemangioma located at the level of the T12 vertebral body. Thyroid gland with normal dimensions, inhomogeneous – to be correlated with thyroid ultrasound. No CT signs suggestive of bilateral PE.

CT, computed tomography.

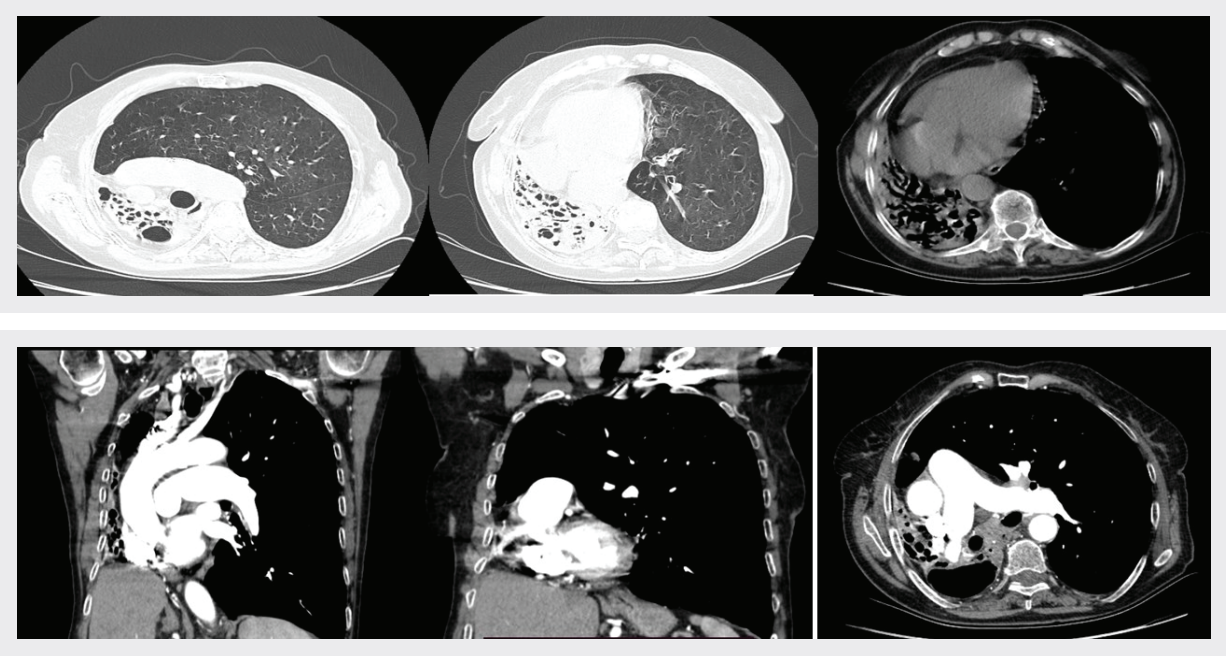


Figure 2. Imaging aspects – Angio CT 2021. CT, computed tomography.

bronchiectasis, primary ciliary dyskinesia) and can be used at home (21).

The device works by sending a low-frequency vibrating pneumatic signal throughout the patient's bronchial tree, liquefying mucus and transporting it from the most distal parts of the peripheral airways to the central bronchi to promote its removal by the patient when they expectorate at the end of the session or after the sessions. Between each very short negative pressure pulse generated by the vibrating pneumatic signal, the patient is reconnected to atmospheric pressure (22).

The patient is in an upright position (sitting) and holds the mouthpiece between the teeth with one hand, and the remote control with the other. The patient breathes in slowly through the nose, and then holds the breath briefly to bring the air behind the secretions. During relaxed expiration, the patient activates the bronchial drainage device signal by pressing the GO button on the remote control: small volumes of lung air are collected at a frequency of 12 Hz (6 Hz at the end of the cycle). This very rapid trapping of air generates a negative pressure pulse in the bronchial tree. As the unit continues to draw air during expiration, the negative pressure

Table 3. Presentation of the medical device.

Conditions		
Cystic fibrosis, COPD, bronchiectasis, primary ciliary dyskinesia The lungs show a degree of congestion and difficulty mobilising their mucus.		
Mechanism	**Benefits**	**Contraindications**
It helps to liquefy and transport the secretions from the small bronchi of the distal part of the lung to the large bronchi, so that they can then be eliminated by coughing.	Hygienic concept with exhalation kit replacement It has ergonomics to be used by the patient himself (autonomy in the management of airway clearance) or in addition to the intervention of the physiotherapist It liquefies and transports bronchial mucus, even from very distal parts of the lungs No increase in patient fatigue Reduced risk of bronchial collapse No side effects Safe and easy to operate/easy to learn	Pneumothorax that has not been treated or drained Unstable cardiovascular pathologies (recent heart attack, unstable angina, uncontrolled rhythm disturbances, unstable heart failure) Massive haemoptysis

COPD, chronic obstructive pulmonary disease.

pulse generated increases. This sequence of very short negative pressure pulses of increasing intensity broadcasts a vibratory signal in the bronchial tree. It acts on the rheological properties of mucus to reduce its viscosity. The liquefied mucus is simultaneously transported from the peripheral lung area to the central airways and then naturally expectorated by the patient (21–24).

The patient is thus kept at atmospheric pressure for the majority of the expiratory time (70%) to avoid any risk of bronchial collapse, the main obstacle to clearing the peripheral airways. The power of the device can be adjusted to fit each patient (21).

There are no absolute contraindications for this new bronchial drainage device's use.

Indications, benefits, and relative contraindications related to the device are listed in Table 3 (21).

Our patient was recommended the use of this drainage device based on the studies published in the scientific medical literature. Thus, the medical reports published by the teams from Slovakia, Poland, and Romania showed that this device has benefits for patients who are known to have cystic fibrosis, are asthmatic, have COPD, and are in the pulmonary rehabilitation period after SARS COV2 infection. The benefits listed by them are related to reduced sputum viscosity, increased mobility at the level of the diaphragm, improved spirometric values (FEV1 + 2%–5%) and, most importantly, good patient tolerance (25–28).

Discussions

There have already been several studies of the new bronchial drainage device, but no study of this bronchial drainage device in SJMS. Numerous studies have demonstrated the superiority of the bronchial drainage device in lung diseases involving a difficulty in expectorating secretions from the respiratory tract.

Many patients of various ages who have used the bronchial drainage device confirm they easily cope with using the device, and the secretions have become more and more abundant and the expectoration easier since the first drainage sessions (29). Our 82-year-old patient testified that the device is easy to use and intuitive, and the increase in the amount of sputum was noted by the acquired independence, adherence, and consistency in using the device for 3 years already.

The bronchial drainage device was well tolerated with good adherence in nine patients with cystic fibrosis out of a total of 11 patients evaluated as an alternative to conventional respiratory physiotherapy (27,28,30). Our patient used the device for 3 years, during which time she accumulated other comorbidities that negatively affected a rather rare lung disease.

The bronchial drainage device led to an improvement in diaphragmatic mobility, the rheology of bronchial secretions, and an increase in lung function (FEV1 being the most monitored parameter) in patients with cystic fibrosis (25,27,31). We intended to perform another plethysmography on our patient, with the aim of assessing lung function after introduction of the medical device, but due to repeated associated bacterial and/or fungal infections and haemoptysis recurrent episodes, this was postponed. Also, due to her impaired mobility, remote patient monitoring was preferred. Although functional tests have not been carried out since our first contact with the patient, the bronchial drainage device made a marked improvement in her quality of life, as has also been proven in post-Covid 19 patients (26).

The use of the bronchial drainage device was temporarily stopped during haemoptysis episodes; but, in the absence of these, our patient has been using the drainage device

twice a day. We appreciate once again the importance of the autonomy that this device was able to offer the patient. It is important to appreciate the ideal moment to resume the use of the bronchial drainage device because it reduces the rate and duration of hospitalisation (31).

Conclusions

Drainage of secretions in patients with severe lung damage is a challenge. In many cases, due to the significant reduction in muscle mass, the combination of bronchial drainage device together with drug treatment and oxygen therapy can be a win–win solution for patients.

The bronchial drainage device (SIMEOX) is a new therapeutic drainage option that has been in competition for several years with the existing modalities (cough assist device) and it shows considerable benefits brought to patients with respiratory diseases and other associated comorbidities. It can also be used with confidence at home or remotely by home care services for very long periods. If the indications are correct and followed, the bronchial drainage device proves to be useful for these patients.

Conflicts of interest

The authors declare no conflicts of interest related to this manuscript. The patient agreed to the publication of this clinical case.

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