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## Speech therapy in the management of difficult-to-treat chronic cough — preliminary results

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### Abstract

**Introduction:** The efficacy of management of chronic cough in adults is limited. Speech therapy is one of the few therapeutic methods which seems to be useful in patients with persistent chronic cough. However, the method has not been available in Poland so far. The aim of the study was to implement speech therapy and assess its efficacy in the management of patients with difficult-to-treat chronic cough.

**Material and methods:** Patients, who were diagnosed and managed due to difficult-to-treat chronic cough, were enrolled into the study. Speech therapy was developed on the basis of the technique described by Vertigan. The entire therapy consisted of eight weekly sessions, each lasting 45 minutes. Before and after speech therapy, cough severity and its impact on the quality of life was assessed by the Visual Analogue Scale (VAS) and Leicester Cough Questionnaire (LCQ). Additionally, cough challenge test with capsaicin was performed.

**Results:** Eighteen women were enrolled into the study, 15 of them (83%) attended all treatment sessions (median age 66 years, median duration of cough 60 months). There was a significant decrease in cough severity measured by VAS (46 vs 28 mm,  $p = 0.016$ ) after completion of speech therapy. A significant improvement in patients' quality of life measured by LCQ (10.7 vs 14.6 points,  $p = 0.004$ ) and an increase in the threshold of cough reflex measured by capsaicin challenge were also demonstrated.

**Conclusions:** Speech therapy resulted in a decrease in cough severity and improvement of quality of life of females with refractory chronic cough. Our results support the use of speech therapy as add-on treatment in females with difficult-to-treat cough.

**Key words:** chronic cough, cough management, difficult-to-treat cough, speech therapy

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### Introduction

Chronic cough is a common complaint affecting as many as 10–20% of adult population [1–3]. It has a negative impact on general health status, including physical, psychological and social domains of quality of life [2]. According to a cross sectional European survey, 93% of patients with chronic cough sought medical attention and 72% of patients attended more than three visits because

of this symptom [4]. Therefore, chronic cough and its management are related with a significant economic burden [2].

Chronic cough is commonly a symptom of smoking-related bronchitis. As smokers still account for a significant proportion of the adult population in Poland (30% of men and 21% of women), smoking related bronchitis is a frequent cause of cough in our country [5]. However, smokers rarely seek medical advice because of cough. The most

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common causes of chronic cough in non-smoking adults with a normal chest radiograph are: upper airway cough syndrome (UACS) i.e. chronic rhinitis or rhinosinusitis; gastroesophageal reflux disease (GERD); asthma and non-asthmatic eosinophilic bronchitis (NAEB) and treatment with ACE inhibitors. In many patients cough may have two or more coexisting causes [1–3]. Our previous study showed that 42.5% of patients with chronic cough had a single underlying disease, while 45% and 12.5% of patients were diagnosed with two and three coexisting causes, respectively [6]. In the majority of patients chronic cough can be successfully diagnosed and treated, but in 5–10% cough persists, despite a thorough diagnostic and therapeutic work-up [7]. In specialized cough clinics, the proportion of patients with persistent, difficult-to-treat cough is even higher. The group of patients with persistent cough includes both patients with unexplained chronic cough (of unknown cause or cough with inconclusive diagnostic work-up) and difficult-to-treat cough (chronic cough with known underlying cause(s) which failed to respond to specific treatment) [7]. Unexplained and difficult-to-treat chronic cough significantly impairs patients' quality of life [7–9], nevertheless the number of therapeutic options that can be effectively applied in such patients is very limited. These include pharmacotherapy with gabapentin, pregabalin or baclofen and non-pharmacological intervention called speech and language or voice therapy [7, 10]. The latter consists of education, respiratory and speech exercises and teaching the strategies to reduce cough [10, 11]. The results of recent studies suggest that speech therapy might be useful in the management of persistent, refractory cough [12–14]. However, this method has not been available in Poland to date.

Therefore, the aim of this study was to develop and implement our own protocol of speech therapy and to assess its efficacy in the treatment of patients with difficult-to-treat or unexplained chronic cough.

## Materials and methods

### Study design

This was a prospective, single arm, single center study performed between 2015 and 2017. The study was registered at ClinicalTrials.gov (NCT 03457610). The protocol of the study was approved by the Institutional Review Board of the Medical University of Warsaw (KB/218/2015) and all enrolled patients signed an informed consent.

### Patients

The study group was recruited from patients diagnosed and treated due to chronic cough in the Department of Internal Medicine, Pulmonary Diseases and Allergy of the Medical University of Warsaw. As there was a significant female predominance in our cohort of patients with chronic cough, we decided to enroll only women. The remaining inclusion criteria were as follows: 1) age between 18 and 80 years, 2) chronic cough lasting more than six months, 3) unsuccessful management of cough despite at least 3-month treatment, 4) no symptoms of acute airway infection in the previous 4 weeks preceding the study. The main exclusion criteria were: 1) inability to participate in speech therapy sessions, 2) cough lasting less than 6 months, 3) active cigarette smoking, 4) current therapy with ACE inhibitors, 5) symptoms of acute airway infection (other than cough), and 6) abnormal chest radiogram or CT scan suggesting any acute or chronic pulmonary disorder (including infection, neoplasm or interstitial lung disease). In all patients, a panel of the diagnostic procedures was applied to diagnose the cause of chronic cough. These included: spirometry with bronchial obstruction reversibility testing, methacholine challenge, induced sputum analysis, computed tomography of the chest and paranasal sinuses, multichannel/pH impedance and ENT assessment. Then, an appropriate treatment concordant with the causative diagnosis was applied. Patients who failed to respond to treatment or patients with unexplained chronic cough were offered speech therapy in the frame of the current study. The patients could continue their earlier treatment of cough but no new therapies other than speech were allowed at this point.

### Speech therapy

Speech therapy was conducted using a protocol developed by our team on the basis of the technique described by Vertigan *et al.* [11, 12, 15]. It started with patient education on the pathophysiology of chronic cough with emphasis on the lack of benefit of repeated coughing, its harmful effects on the larynx and airways and the advantages of voluntary cough suppression. The patients were informed on the significance of their potential to voluntarily control the cough. Additionally, cough triggers and specific techniques to suppress the cough and avoid laryngeal irritation were presented. Speech therapy included breathing exercises (nose and pursed lip breathing), swallowing exercises (dry swallows or with sip of water, chewing gum or sucking candies in order

to swallow saliva more frequently and attempt to delay the cough) and vocal exercises. Moreover, vocal hygiene was implemented to maximize hydration and reduce larynx irritation. It included increase of water intake and avoiding exposure to environmental smoking or substances known to have drying effect on the larynx (caffeine, alcohol). The effectiveness of the interventions was monitored during the entire therapy. The therapy consisted of eight weekly sessions (2 individual and 6 group sessions), each lasting 45 minutes. After each session the patients were encouraged to repeat the exercises at home.

### Assessment of treatment efficacy

Before and after speech therapy, cough severity and its impact on the quality of life were assessed by the Visual Analogue Scale (VAS; range 0–100 mm) and Leicester Cough Questionnaire (LCQ; range 3–21 points), in accordance with ERS recommendations. The higher numbers in the visual analogue cough scale reflected more severe cough, while the higher LCQ value corresponded with better quality of life in patients with cough. A validated Polish version of the LCQ was used [16]. The minimal clinically important difference (MCID) for LCQ total score is 1.3 points; there is no validated value of MCID for VAS in chronic cough [17–19].

Besides VAS and LCQ, cough challenge test with capsaicin was performed before and after therapy to assess the sensitivity to inhaled irritants expressed by C2 and C5 (concentration of capsaicin which induces 2 and 5 coughs during challenge test with capsaicin, respectively) [17, 18]. Finally, the patients were asked to answer three questions on their subjective rating of the effect of speech therapy (complete vs partial vs no response) and the appraisal of the usefulness of each part of speech therapy. Complete response was defined as complete cough resolution, partial response as decreased cough severity and no response as no change in cough severity before and after speech therapy.

### Statistical analysis

Statistical analysis was performed with Statistica 13.1 (StatSoft, Tulsa, USA) software package. Data on patient characteristics were presented as median and interquartile range (IQR) unless otherwise specified. Differences between continuous variables measured before and after speech therapy were analyzed by Wilcoxon test. All p-values were 2-tailed and  $p < 0.05$  was considered statistically significant.

## Results

Eighteen patients were initially enrolled. Fifteen (83%) attended all speech therapy sessions and thus completed the whole treatment program. Only those patients were included in the final analysis. Of the 3 patients who failed to complete the program, two were excluded because of irregular attendance to the sessions and one due to acute upper airway infection. The detailed characteristics of the study group are presented in Table 1. All patients were nonsmoking women, median age 66 years with median duration of cough 60 months.

After 8 weeks of speech therapy, a significant decrease in cough severity measured by VAS (46 vs 28 mm,  $p = 0.016$ ) and significant improvement in patients' quality of life measured by LCQ (10.7 vs 14.6 points,  $p = 0.004$ ) were noted (Table 2). Clinically important increase ( $> 1.3$  points) in total LCQ score was demonstrated in as many as 13/15 (87%) patients. The sensitivity of cough reflex measured by capsaicin challenge test was lower, but the result was significant only for C2 (Table 2). The increase in cough reflex threshold measured as C2 and C5 was found in 8 (53%) and 9 (60%) patients, respectively (Fig. 1).

After completion of the speech therapy program, one patient declared complete response, while all the remaining (14/15) declared partial improvement. All patients declared that breathing exercises were the most useful component of speech therapy.

**Table 1 . Baseline demographic and clinical characteristics of the study group**

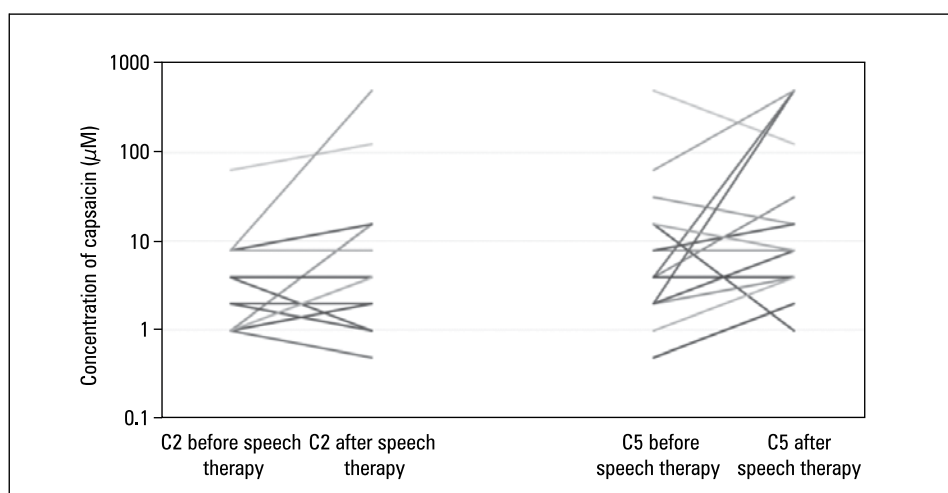
	Patients n = 15
Age, years	66 (56–71)
Sex F/M	15/0
Smoking history S/EX/NS	0/2/13
Cough duration, months	60 (42–156)
BMI, kg/m <sup>2</sup>	26.2 (23.1–28.7)
Cough causes, number of pts.	
GERD	10
UACS	6
Asthma	4
Other causes	1
UCC	1

Data are given as median and IQR (in parenthesis) or number of patients. BMI: body mass index; EX: ex-smoker; F: female; GERD: gastroesophageal reflux disease; M: male; NS: never smoker; S: smoker; UACS: upper airway cough syndrome; UCC: unexplained chronic cough

**Table 2. Results of the speech therapy**

	Before speech therapy	After speech therapy	p-value
Severity of cough measured by VAS, mm	46 (38.5–73)	28 (21.5–65)	0.016
Quality of life measured by LCQ, points	10.7 (8.4–11.9)	14.6 (11.9–17.1)	0.004
LCQ physical domain	3.62 (3.2–4.1)	4.75 (4.3–5.4)	0.001
LCQ psychological domain	3.57 (2.4–4)	4.87(4–6.1)	0.004
LCQ social domain	3.5 (2.7–3.7)	5.0 (3.6–5.7)	0.009
Sensitivity of cough reflex measured by capsaicin challenge, $\mu\text{M}$	C2–1.96 (0.98 –7.84)	C2–3.92 (1.96 –15.68)	0.03
	C5–3.92 (1.96–15.68)	C5–7,84 (3.92–78.4)	> 0.05

Data are given as median and IQR (in parenthesis). The effect of speech therapy was analysed using Wilcoxon test. C2: concentration of capsaicin, which induces 2 coughs; C5: concentration of capsaicin, which induces 5 coughs; LCQ: Leicester Cough Questionnaire, range 3–21 points; VAS: visual analogue scale, range 0–100 mm



**Figure 1.** Effect of speech therapy on results of capsaicin cough challenge test. C2: concentration of capsaicin, which induces 2 coughs; C5: concentration of capsaicin, which induces 5 coughs

## Discussion

The results of our study demonstrated that speech therapy based on the methods described by Vertigan *et al.* [11, 12, 15] was an effective therapeutic approach for patients (in our group women) with persistent, difficult-to-treat cough. Speech therapy significantly reduced cough severity and improved patients' quality of life. These results are particularly important in the context of the European Respiratory Society (ERS) and American College of Chest Physicians (ACCP) guidelines which state that the above parameters measure the most important aspects of cough assessment [17, 18]. It should also be emphasized that all our patients showed partial or complete response to applied therapy.

We would like to stress that the favorable results were obtained in patients who had been

unsuccessfully treated for a long time before the study onset. The median duration of cough in this group was 60 months, but the longest duration was 30 years. Only one patient diagnosed with unexplained chronic cough had not received causative treatment before the study onset. Failure in management of chronic cough is a well-known phenomenon [7, 20]. In previous studies, 36–55% of patients with known cause of chronic cough did not respond to specific therapy [4, 20, 21]. According to the ACCP guidelines, persistent untreatable cough affects approximately 5–10% of all patients with chronic cough and even up to 46% of patients managed in specialized cough clinics [7]. It seems that the failure of cough treatment may result from hypersensitivity of cough reflex [7, 20, 22]. It is hypothesized that repeated coughing causes laryngeal injury, resulting in neural inflammation, increased expression of

cough receptors and both peripheral and central neuromodulation. Subsequently, cough threshold decreases producing cough even with low level irritant stimuli. Repeated cough maintains the pathophysiological processes in the airways and nervous system resulting in difficult-to-treat or refractory cough, currently referred to as cough hypersensitivity syndrome.

Another important feature of our study group was the fact that all patients were women. Similar to other authors, we observed a higher prevalence of women among patients with chronic cough. In international surveys, women accounted for approximately 67% of population with chronic cough [20, 21]. It may result from higher sensitivity of cough reflex observed in females [20]. Although in clinical practice we also see men with chronic cough, the predominance of women is highly significant. Therefore, we decided to include only females to our study. This approach also made our study group quite homogenous. Nevertheless, our team is also currently working on a study in men, and we believe it will be interesting to compare the results obtained in women and men.

We believe it is critically important to use various, possibly the most objective and recommended tools measuring cough in both clinical trials and routine practice. Cough severity combines aspects of cough intensity (measured by VAS), frequency (by cough recorders) and influence of cough on quality of life (assessed by dedicated questionnaires). One of the most common cough questionnaire is LCQ, which had recently been validated in the Polish population [16]. Following the recommendations [17, 18], we implemented the measurements of both cough severity and its impact on patients' quality of life. This approach enabled more objective assessment of the therapeutic efficacy. We also assessed cough intensity by cough reflex measurement using capsaicin cough challenge test. Although we observed an increase in the cough reflex threshold in more than one half of patients, the results were somewhat ambiguous. Similar results were recently reported by Chamberlain Mitchell *et al.* [14], who confirmed the efficacy of speech and language intervention in treating chronic cough, but did not observe an increase in cough inducing capsaicin concentration. It can be speculated that in refractory cough, different receptors or signaling pathways may be involved than in that stimulated by capsaicin. According to the ERS and ACCP recommendations, the capsaicin test might be useful in measuring cough intensity, but its use is not mandatory [17, 18, 20]. Although

cough recorders are recommended for objective cough frequency measurement, they are not easily available and we did not have access to this tool during the study [17, 18]. Therefore, we based on subjective tools for cough assessment.

Management of difficult-to-treat chronic cough is certainly a challenge and there are only two potentially successful methods: pharmacological — based on centrally acting neuromodulators such as gabapentin or pregabalin and nonpharmacological intervention — such as speech and language therapy [7, 10]. The latter was adapted from the treatment of hyperfunctional voice disorders. Although the first studies on speech therapy as a method of chronic cough treatment had been published 30 years ago, the first randomized trial was reported by Vertigan *et al.* [12] only in 2006. Speech therapy relies on multifactorial interventions, which may comprise several elements: education on cough, identification of cough triggers, teaching cough suppression techniques, breathing exercises, vocal hygiene and hydration techniques. Sometimes additional forms of counselling are used. According to the literature, speech therapy is usually delivered by speech and language therapists or physiotherapists and consists of 2–4 sessions [10–15]. In our study, speech therapy was implemented and performed by a phoniatician (DR) and speech and language therapist (AŁ). The mechanism of cough reduction by speech intervention is still a matter of debate. Perhaps, breathing exercises allow to relax the throat, neck, and shoulder muscles, what helps to reduce the adductor activity of the vocal folds during expiration in patients with chronic cough and paradoxical vocal cord movement [10, 15]. Moreover, it is believed that swallows may interfere with cough and alter its threshold. Similarly, proper hydration and avoidance of laryngeal irritants, as well as conscious suppression of cough may desensitize cough reflex, increase its threshold and reduce cough frequency.

To our knowledge, there were only a few studies on the effect of speech therapy in the management of chronic cough [12–14, 23, 24]. All of them confirmed the efficacy of speech intervention. The recently published randomized multicenter trial also confirmed that physiotherapy combined with speech and language intervention is an effective method of reducing cough in patients with refractory chronic cough [14]. Therefore it was so important for us to implement that technique and assess its efficacy in Polish population, since such a therapeutic approach had not been commonly used in Poland before. To our knowledge there has

been only one case report published so far, which referred to trial of speech therapy in management of difficult-to-treat chronic cough in Poland [25].

Although our protocol of speech therapy intervention in chronic cough was mainly based on that presented by Vertigan *et al.* [11,12,15], we also introduced some modifications. The main components of our therapy were education on cough pathophysiology and vocal hygiene, identification of cough triggers, instruction on cough suppression techniques, conscious breathing, diaphragmatic breathing, relaxed breathing control techniques, speech and vocal exercises. Individual sessions followed by group sessions was our concept of providing patient psychological support. The patients were strongly encouraged to repeat the exercises at home.

Our study has some limitations. The most relevant is the small study group. However, we would like to emphasize that these are only preliminary results, which need to be confirmed in a larger study. On the other hand, in a recently published multicenter randomized control trial concerning speech and language intervention, there were only 75 patients enrolled and 34 of them were allocated to the speech and language therapy arm [14]. Therefore, as speech therapy had not been available in Poland, our intention was to sum up the preliminary results and to present the technique. Second, our study included only women, and this can be considered a selection bias that could have influenced the results. On the other hand, persistent, difficult-to-treat cough is more common in women. In other similar studies women accounted for even 68–74% of the study population [12, 14]. Regardless of the above, this method certainly needs to be evaluated in men with difficult-to-treat cough. Third, this study was a single arm, observational study without control group or randomization, so the results cannot be compared with other interventions and control patients. Fourth, the effect of speech therapy was measured only once, just after the end of the intervention. Therefore, there is no evidence that these favorable results are long-term. Finally, we did not use cough monitors to assess the frequency of chronic cough. There is no doubt that cough monitors are useful for objective measurement of cough frequency which significantly increase the reliability of studies on cough. However, we have implied all other accessible tools to evaluate the impact of speech therapy on cough intensity. Despite all these limitations, the results of this study seem to indicate that speech therapy is a useful technique

in the management of patients (in our group women) with difficult-to-treat chronic cough.

## Conclusion

Basing on the method described by Vertigan *et al.* [11,12,15], we developed, implemented and evaluated speech therapy which resulted in a decrease in cough severity and improvement of quality of life of patients with refractory chronic cough. Our results support the use of speech therapy as add-on treatment in females with difficult-to-treat cough.

## Conflict of interest

The authors declare no conflict of interest.

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