



Case Report

Ankylosing Spondylitis and Respiratory Symptomatology – A Case Study in Mediterranean Health Center – Igalo

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Abstract	Keywords
<p>Rheumatic diseases are considered as public health issue. Special place goes to Ankylosing spondylitis (AS). Patients with AS diagnosis were treated in Mediterranean Center Igalo. Spirometry parameters, parameters of mobility of spine and lumbar spine (Schober's test), width of chest were measured at the beginning and at the end of this therapy. Also, modified questionnaire related to overall health condition was developed. High correlation between summary value of modified questionnaire (SCORE) and parameters of pulmonary function were recognized respectively: a) -0.78451 (SCORE-PEF), b) -0.90055 (SCORE- FEV1) and c) -0.77011 (SCORE-FVC). Also, correlation between Schober's test and parameters of pulmonary function was: a) Sch./FEV1=0.19, b) Sch./PEF=0.30; c) Sch./ FVC = 0.39. All investigated parameters indicated significant improvement at the end of treatment. Insomnia was developed in all patients (100%), while 33% of patients noticed it after four weeks of treatment. Anxiety was notable at the beginning of treatment in 76% of patients while depression syndrome was recognized in 63% of patients. At the end of research, 30% of patients with AS were anxious while 20% of patients with AS suffered of depression syndrome. The highest improvement was noted for parameter FVC. Also, improvement in chest elasticity was noted. Research results suggested modernization of therapeutic modalities for treatment of patients with AS in Montenegro.</p>	<p>Ankylosing spondylitis Schober's test Spirometry</p>

Introduction

Rheumatic diseases present the main cause of chronic invalidity. Also, they are recognized as public health issue. Special place goes to Ankylosing spondylitis (AS) - (Morbus Bechterew or Marie-Strümpell's disease).

Ankylosing spondilitis (AS) is inflammatory rheumatic disease included in group of spondyloarthropathies. Disease symptoms usually develop during 15-30 years of age while only 5% of patients were faced with this disease after 40 years of age. The disease reflects through pains in lumbo-sacral part of spine which

further spreads to gluteal region and legs. Pains are usually thudding. They develop in early morning hours and wake the patient up while immobility usually lasts for half an hour (Calin and Elswood, 1990).

Patients also have pains in muscles which explain that these changes originate directly from inflammatory process or due to irritation of roots of spinal nerves. Changes in respiratory (pulmonary fibrosis, interstitium, pleure) system, gastrointestinal system appear in later stadium of disease. Also, inflammations in urogenital tract can also be observed (Van der Linden and van der Heijde, 2000). Also, there is an increased risk of atherosclerosis, cardio-vascular diseases which are the main cause of lethality of these patients (Alvarez and López de Castro, 2000).

Measurement of mobility of cervical, torakal and lumbosacral part of spine indicated on limited mobility in all directions (Menell, Schober) (Maksymowych, 2004). Restrictive breathing disorder is developed in patients with advanced changes in spine and bone-vertebral joints. Spirometric tests confirmed certain level of damage in pulmonary parenchyma (Burdon et al., 1983; Di Marco et al., 1983). The most often difficulties are dyspnoea, unproductive cough, intolerance to efforts (Javaheri and Sicilian, 1992; Holland, 2010). Dyspnoea and physical limitation lead to difficulties in performing of daily activities (Holland, 2010; Harris-Eze et al., 1994; Agusti et al., 1991). The quality of life of patients with AS can be reviewed via questionnaire Medical Outcomes Short Form-36 (SF36), Medical Research Council scale for dyspnoea (MRC) and questionnaire related to anxiety and depression (Hospital Anxiety and Depression-HAD) (Vesovic-Potic et al., 2009). Conventional antirheumatic drugs showed small effect in treatment (Dougados et al., 1995; Maksymowych et al., 1998; Maksymowych et al., 2001).

New biological drugs are used all over the world since 1998. They block inflammation stimulators in joints and its envelopes: Factor of necrosis of alpha tumor (TNF-1), interleukin 6 (IL-6). Overall clinical condition of AS patients is improved, disability is decreased and difficulties are lowered. However, this therapy is expensive and personalization is necessary (Braun and Sieper, 2002). Physical therapy and regular gymnastic are very important for maintaining of existing condition, prevention of invalidity progression and improvement in functioning of freeze joint and spine mobility (Karatepe et al., 2005).

Living in adequate climate conditions is recommended since difficulties often increase on cold temperatures. The optimal body weight should be maintained, including avoidance of hard physical work. Stationary treatment in one of rehabilitation centers is necessary. Many forms of physical therapy are useful (kinesiotherapy, hydro, manual and underwater massage, mud and paraffin wraps, thermal procedures, ultrasound, laser, LED, various forms of electrotherapy) (Van der Heijde and Spoorenberg, 1999; Dagfinrud et al., 2005; Fernández-de-Las-Peñas et al., 2006).

The main objective of this paper is to investigate connection between parameters of pulmonary function including results of mobility of spine, (Schober), results of questionnaire of overall health condition (SF-36), MRC scale of dyspnoea and results of provided questionnaire related to HAD after four weeks of therapy cycle.

Materials and methods

Experimental part of research included sixty AS patients treated in Mediterranean Center for physical therapy and rehabilitation – Igaló. Male and female patients were included in this research, and they were 28 to 70 years of age. Four weeks of rehabilitation included kinesiotherapy – practices for neck, upper and lower extremities, swimming, hydro-gymnastics, underwater massage, mud packs, ultrasound and electrotherapy. Parameters of pulmonary function, Forced vital capacity (FVC), Top expiratory flow (PEF) and Forced expirium volume (FEV1) were measured at the beginning and the end of the therapy. Patients responded to questionnaires (SF36 and HAD and MRC). Mobility indexes were estimated: Distance Wall – Occipito (Occ. wall), index of sagittal mobility of cervical (Phy. gl.), lumbal spine (Schober's test) and width of chest (Tho-exp.).

Questionnaire SF-36 is a short form of health survey which included 36 questions. It represent theoretical basis for empirically confirmed operability of two overall health concepts, physical and psychological, including their two common manifestations, functioning and advantages. The paper for investigating of level of HAD represented special kind of questionnaire. It represents emotional condition of patient, level of anxiety and depression. According to overall result, mild (0 – 10), moderate (10 – 25) and high (25– 40) level of anxiety and depression can be observed. MRC dyspnoea scale is the questionnaire which presents level of hard

breathing. Questionnaire results provide data on mild, moderately hard and very hard breathing. Scoring or results calculation is provided via following order: first step is data entry; Second step is review of inserted data in order to provide answers to all questions; Third step is summation of obtained results; Fourth step is transformation of results into new score; Final step includes review of previous action. Considering that some questions from these three questionnaires were repeated, a modified questionnaire, adapted to this research, was made.

Statistical software package DATA FIT 9-version 2008 was used for statistic analysis of data. This software was a tool for testing of significance of correlation between parameters of pulmonary function and questionnaire score. Multifactor nonlinear regressive analysis was developed in order to prove statistical connection

between overall physical condition, quality of life and emotional condition of AS patients according to pulmonary function parameters. Statistic software package known as Free statistic software (V 1.1.23-r7)-part Chrombah Alpha (V.1.01) was used for review of consistence of modified questionnaire results.

Results

There was total of 60 respondents, who were 53.6 ± 16.4 years of age, male and women. 43.3% of those respondents were women. Average duration of disease is $15.6 + 4.52$ years. Review of internal consistence of modified questionnaire using Chrombah alpha coefficient resulted with high values of coefficient (Table 1) at total level. Also, these high values were noted for individual parts related to SF 36 and HAD scale (max $\alpha=1$).

Table 1. "Cronbach's Alpha" coefficient for questionnaires.

Cronbach's Alpha Statistic				
	Cronbach's Alpha	Std. Alpha	G6 (smc)	Average R
TOTAL	0.8944	1	1	1
Modified questionnaire	0.95	1	1	1
MRC Scale	0.7674	1	1	1
SF 36 Scale	0.8204	1	1	1
HAD Scale	0.8697	1	1	1

Table 2. Correlations between spine mobility and spirometry.

	FEV1	PEF	FVC	Phy. gl	Occ. wall	Tho-exp	Sch. test
FEV1	1	0.55542622	0.847705662	-0.118029685	-0.299967394	0.155618571	0.19829463
PEF	0.55542622	1	0.325032632	0.000824033	0.151767348	0.005525719	-0.304300185
FVC	0.847705662	0.325032632	1	-0.1420091	-0.479051243	0.305627345	0.397144877
Phy. gl	-0.118029685	0.000824033	-0.1420091	1	0.419522451	-0.478293621	-0.362544124
Occ. wall	-0.299967394	0.151767348	-0.479051243	0.419522451	1	-0.377006845	-0.497229514
Tho-exp	0.155618571	0.005525719	0.305627345	-0.478293621	-0.377006845	1	0.275276793
Sch. test	0.19829463	-0.304300185	0.397144877	-0.362544124	-0.497229514	0.275276793	1

FVC-Forced vital capacity; PEF-Top expiratory flow; FEV1-Forced expirium volume Occ. Wall-Distance Wall-Occipito; Index of sagittal mobility of cervical (Phy. gl.), Schober's test (Sch. test) and width of chest (Tho-exp.).

Regarding anxiety and depression scale, average moderate intensity (22.5 ± 5.3) was registered among patients. The most important predictor of insomnia, anxiety, depression, overall health condition and specific facts about patients was presented as total result (SCORE) of both questionnaires. Its mean value for all respondents was 45 ± 4.2 , which implies to moderately tough quality of life.

Correlation between summary value of modified questionnaire and parameters of pulmonary function had following individual values: a) -0.78451 (SKOR-PEF); b) -0.90055 (SKOR- FEV1); c) -0.77011 (SKOR-FVC). Correlation between PEF and FEV1 was 0.555, while

this value between PEF and FVC was 0.322. Correlation between parameters FEV1 and FVC was 0.847. Results of nonlinear multifactorial regression analysis provided a model explained by following equation:

$$\text{SCORE} = \text{EXP} (0.11821*VC + 0.11276*PEF - 0.122666*FEV1+4.66)$$

Value of regression coefficient $R^2=0.83$ implied to high reliability of model (max $R^2=1$, $p \leq 0.05$). In other words, it implied to the fact that it is possible to express total SCORE via parameters of pulmonary function.

Correlation between Schober's test and parameters of pulmonary function was following:

FEV1 =0.19; PEF=0.30; FVC = 0.39. Other correlations between spine mobility and spirometry are presented in Table 2.

Discussion

The main Mediterranean health center in Igalo is spa and health resort which confirmed quality of its services for AS patients through several decades of work and successful treatments. It concluded that thermal water in combination with mineral mud creates optimal conditions for treatment of AS disease (Van der Linden and Van der Heijde, 2000). All investigated parameters of pulmonary function, indexes of mobility in vertebral and thoracic spine, Schober's test and total score from modified questionnaire indicated on significant improvement at the end of treatment. Respiratory symptomatology for patients with AS is mostly indicated by dyspnoea, nonproductive cough and intolerance to efforts (Maksymowych, 2004; Burdon et al., 1983). Dyspnoea and physical limitation lead to difficulties in performing of everyday activities. Patients have rapid and shallow breathing. Rigid lung tissue leads to increasing of elastic load during aid inspiration so it is necessary to decrease total forces in chest and maintain the needed ventilation (Di Marco et al., 1983).

Disorder in gas exchange develops due to destruction of capillary network in lungs or callosity of alveolar-capillary membrane. Disorder in relation ventilation – perfusion and decreasing of partial pressure of O₂ in pulmonary veins (Javaheri and Sicilian, 1992). This leads to lung hypertension and insufficiency of the right heart (Holland, 2010). Disorder in function of skeletal muscles is noted as an important cause of intolerance to efforts (Harris-Eze et al., 1994). There is small knowledge on potential mechanisms which lead to disfunction of skeletal muscles in patients with AS. Loss of physical abilities leads to avoiding of physical activity. Furthermore, it leads to dyspnoea and deterioration of respiratory symptoms. Application of corticosteroids contributes to myopathy of peripheral respiratory muscles (Dougados et al., 1995; Maksymowych et al., 1998).

Patients used anti-rheumatic drugs and corticosteroids during treatment, continuously or with short breaks. Biological therapy was not applied for such treatments. Insomnia and anxiety were present as leading symptoms of depression. Depending on complexity of diagnosis for patient with AS, psychological status changed, which

lead to changes in quality of life. Insomnia was registered for all patients (100%), and after four weeks of treatment it was still a problem for 18 (33%) patients. At the beginning of treatment, 42 (76%) patients noted anxiety while 36 (63%) patients noted depressive syndrome. At the end of the research, 18 (30%) patients with AS noted anxiety while 30 (20%) patients noted depressive syndrome.

Rehabilitation program for patients with AS whose disease include restrictive disorder in lungs ventilation includes following: kinesiotherapy, aerosol therapy, various medications, hygiene and dietary regime, smoking cessation, psychological support, evaluation of the patient and test for the maximum cardio-pulmonary load. The aim of program of respiratory rehabilitation is decreasing of symptoms, improvement of breathing function, improvement in quality of life and better performing of daily activities including rational use of healthcare (Van der Heijde and Spoorenberg, 1999; Fernández-de-Las-Peñas et al., 2006).

Kinesiotherapy improves and increases following: motivation for better handling with efforts, decreases mood disorder, alleviates symptoms and improves respiratory and cardio-vascular function. Before practicing, clinician should determine optimal medical treatment including bronhodilators, long oxygen therapy and treatment of existing illnesses. Parameters from load tests are often better predictors than result for lung function at rest (Maksymowych at all, 2001; Braun and Sieper, 2002). However, there is no consensus to decide which parameter from load test can be observed as best predictor. Among parameters for lung function, the greater improvement at the end of treatment was noted for parameter FVC. At the same time, FVC had a high correlation with Schober's test and total score of modified questionnaire.

Kinesiotherapy program includes exercises for neck, upper and lower extremities (ergometer bicycle for hands, load lifting and elastic tapes). Practices for upper extremities lower dyspnoea during activity. Bicycle riding and walking is the most often form of physical practicing for patients with AS. The practice conducts in intervals where longer sessions are replaced with shorter ones, rest periods are divided and lower intensity practices are also included. Training in intervals leads to significantly smaller changes in symptoms although patients are burdened with high absolute load which helps in maintaining of practicing effects (Karatepe et al., 2005; Ince et al., 2006).

Resistance exercises have higher potential for improving of mass and strength of muscles unlike endurance exercises. Practicing sessions include 2 to 4 units with 6 to 12 repetitions with intensities from 50% to 85% relating maximum value. Training of strength leads to decreasing of dyspnoea during exercising. Ankylosing spondylitis patients who performed four weeks of inspiratory muscle training associated to conventional exercise training had an increased chest expansion, a better aerobic capacity, resting pulmonary function and ventilatory efficiency than those who performed conventional exercise training only (Widberg et al., 2009; Scholten-Peeters et al., 2004). The Global Postural Re-education (GPR) method results in greater improvement with a group physical therapy program than with home exercises. This can be explained by the mutual encouragement, reciprocal motivation, and exchange of experience in group therapy (Ince et al., 2006; Widberg et al., 2009). Since a decrease in chest expansion is secondary to ankylosis in AS, there is also pulmonary involvement. This may even further decrease the low psychological status and quality of life in patients with AS. By performing the following exercises, the chest expansion can increase, leading to better functional capacity (Widberg et al., 2009).

In addition to conventional exercises (flexibility exercises for cervical, thoracic and lumbar spine and major muscle groups) and respiratory exercises (pursed-lip breathing, expiratory abdomen augmentation, and synchronization of thoracic and abdominal movement), aerobic exercises such as swimming and walking are recommended (Risk et al., 2004). Research has shown a significant increase in chest expansion following swimming programs in patients practicing swimming and/or walking aerobic exercises (Scholten-Peeters et al., 2004). Aerobic exercises lead to a bigger chest expansion and therefore a better functional capacity. It also decreases the chance of respiratory failure. Spa therapy has shown significant positive short- and long-term effects on pain, stiffness, well-being and functioning of patients with AS (Risk et al., 2004).

Conclusion

This study showed that in AS spinal mobility measure are associated with physical function general health, emotional role, mental health domains of quality of life and parameters spirometric measure. It is showed that treatment residence in Mediterranean spa – rehabilitation resort is necessary for patients with AS. Treatment

results showed health improvement in patients regardless of fact that conventional therapy methods were applied. Introduction of contemporary therapy modalities in treatment of patients with AS in Montenegro would probably lead to much better results. Further investigations should be aimed to this assumption.

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