MYOTONIC DYSTROPHY FOUNDATION

OCCUPATIONAL THERAPY SUGGESTIONS FOR THE MANAGEMENT OF A MYOTONIC DYSTROPHY PATIENT

Cynthia Gagnon, erg, Ph.D.
Professeur adjoint,, École de réadaptation, Faculté de médecine et des sciences de la santé
Université de Sherbrooke, Groupe de recherche interdisciplinaire sur les maladies neuromusculaires

GOAL OF OCCUPATIONAL THERAPY (OT)

Occupational therapy is a health profession concerned with promoting health and well-being through occupation. Occupation refers to everything that people do during the course of everyday life (CAOT Position Statement on Everyday Occupations and Health, 2003) and can relate to participation. The primary goal of occupational therapy is to enable people to participate in the occupations which give meaning and purpose to their lives [1]. Occupational therapists have a broad education that provides them with the skills and knowledge to work collaboratively with people of all ages and abilities that experience obstacles to participation. These obstacles may result from a change in function (thinking, doing, feeling) because of illness or disability, and/or barriers in the social, institutional or and physical environment (Adapted from the World Federation of Occupational Therapists, 2004).

MYOTONIC DYSTROPHY TYPE 1: CLINICAL FEATURES RELATED TO OT INTERVENTIONS

In that DM1 is a complex multisystemic disorder, only a brief description of clinical features related to OT interventions will be done as a general portrait is available elsewhere [2]. This portrait is related to the adult phenotype only although general recommendations could apply to all phenotypes.

DAILY ACTIVITIES

Category	Social Participation
Nutrition	Meal preparation: 27.5 % reported needing human help or not carrying it out [3]. Taking a meal: Taking a meal is usually adequate although dysphagia may be present. In 40 patients, 45% reported having symptoms of dysphagia. [4] In a radiological study, 20% had aspirations (3/15) with or without symptoms of dysphagia.[5]. The nature of the swallowing defect in DM1 is complex, and investigations revealed abnormalities in smooth as well as in striated muscles [5].

Personal Care	Bathing: 17% - 42% experienced problems (having difficulty, needing human help and/or using technical aids) [6]. Using toilets: 22% experienced problems (having difficulty, needing human help and/or using technical aids){Mathieu, Submitted #3826}. Dressing: 15% experienced problems (having difficulty, needing human help and/or using technical aids) {Mathieu, Submitted #3826}.
Mobility	General mobility is among the most affected area of daily activities. Wheelchair: One study reported that among 51 patients, 6% reported using a wheelchair.[#5117] From a large sample (n = 200), 17.5% where using a wheelchair{Mathieu, Submitted #3826} . Driving: More than 50% are still driving but vigilance should be kept for factors which could influence driving such as myotonia, hypersomnolence, cognitive functions and grip strength{Mathieu, Submitted #3826}. Lower extremity strength, education, technology, support and attitude from family and friends, government and public services, fatigue and gender could partly predict disruption of participation in the mobility-related area [8].
Housing	Social deterioration secondary to muscular dystrophy, intelligence deterioration and reduction of initiative were first described by Thomasen in 1948. [9] Caughey and Myrianthopoulos introduced the term "myotonic's home" because "it was possible to identify a residence by its neglected appearance, the obvious need of repairs, the unkempt yard and garden choked with overgrown grass and weeds, which provided a vivid contrast to the surrounding well-kept homes". [10] Doing major household tasks: 68% experienced problems (having difficulty, needing human help and/or using technical aids)[3]. Natterlund reported that 32.6% of the DM1 patients are not doing activities related to home maintenance and 25.8% do it with problems [11] Maintaining the house: 50% experienced problems (having difficulty, needing human help and/or using technical aids)[3] Lower extremity strength, fatigue, support and attitude of family and friends, education and income could predict disruption of participation with housing related tasks [8].

SOCIAL ROLES

Category	Social participation
Community Life	Getting to public buildings or commercial establishments : 24.7% experienced problems (having difficulty, needing human help and/or using technical aids)[3]
Work	Different studies [12]; [13] showed that 12% to 31% of DM1 patients held a job and that 52% to 66% used to work. In 2007, a reappraisal of the DM1 population from the Saguenay-Lac-Saint-Jean region has shown that 20% are currently working, 66% used to work and 14% never worked. [14] In the same population, 44.5 % reported employment as severely restricted and caused then the highest level of dissatisfaction. [3]. Many aspects of DM1 such as muscular impairment, low education, excessive daytime sleepiness, and apathy, problems with access, equipment and transportation may restrict opportunities to employment as well as leisure. Technology, lower extremity strength, fatigue and pain could partly predict disruption of participation in the work-related area (paid and unpaid work) [8].

	Leisure activities (sports, craft, outdoor or tourist activities) are severely restricted in 22 to 26% of DM1 patients and 24% of this population reported a high level of dissatisfaction about it [3]. From another study, restricted participation in leisure activities was found to be around 63% [6].
Recreation	The following problems to pursue leisure activities were expressed by the patients: physical limitations (29% of the patients); lack of money (28%); fatigue (25%); distance (18%); activities not adapted to their condition (14%); help needed (13%); no transportation available (11%).
	Technology, lower extremity strength, fatigue and pain could partly predict (R2 42%) disruption of participation in the work-related area (paid and unpaid work) [8].

REHABILITATION CONCEPTUAL FRAMEWORK

Rehabilitation professionals are becoming increasingly aware of the importance of evaluating not only the reduction in mental and physical capabilities but also the restriction of participation that may occur in neuromuscular disorders and especially DM1. According to the International Classification of Functioning, Disability and Health (ICF) model, participation (previously called handicap) is defined as involvement in a life situation, and participation restriction is defined as problems an individual may experience in involvement in life situations [15]. The nature, quality and/or duration of participation may be restricted and the comparison is based on an individual without a similar health condition [15]. This refers to the concept of society-perceived participation as opposed to person-perceived participation [16]. This approach, although sometimes useful when comparing populations, can have limited utility in rehabilitation, as it tends to overlook the ability of individuals to make autonomous choices about the way they conduct their lives since the scores are based on a societal and normative perspective of what constitutes optimal social participation. On the other hand, the Disability Creation Process model had operationalized social participation via the concept of life habit, which is defined as "a daily activity or social role valued by the person or his/her sociocultural context according to his/her characteristics (age, sex, sociocultural identity, etc.) and which ensures his/her survival and well-being in society throughout his/her life"[17]. This definition is closer to person-perceived participation, the importance of which has been recognized, especially in chronic conditions where readjustment of life goals and expectations is part of the rehabilitation process [18]. Among several issues, the various clinical phenotypes present in DM1 should also be taking into consideration upon establishing a portrait of participation. Clinically, patients with the mild and adult phenotypes exhibit clearly different pictures and require distinct types of rehabilitation and community follow-ups. Satisfaction related to participation is increasingly gaining attention from literature as it has been associated more strongly with subjective quality of life than the performance component [19]. The individual's feelings about or appraisals of his/her participation has thus been suggested as a promising approach in quality of life assessment as well as in healthcare and community services planning and delivery [20]. Tailoring our intervention towards the areas demonstrating less satisfaction may improve quality of life more than solely focussing on traditional rehabilitation areas such as activities of daily living, which only predict a small proportion of quality of life among a neuromuscular population [11].

OT EVALUATION FRAMEWORK

Occupational therapists evaluate client's occupational performance (social participation), performance components (personal factors), and performance contexts (environmental factors) [21]. OT evaluation should define occupational problems of concern to the client [21].

A) Evaluation Of Occupational Performance (Social Participation)

Several interview procedures are available for the assessment of social participation. A few instruments were recently designed to assess participation from the individual's perspective, such as the Impact on Participation and Autonomy Questionnaire (IPA)[22], the Late Life Function and Disability Instrument (Late-Life FDI)[23] and the Assessment of Life Habits (LIFE-H) [24]. The Canadian Occupational Performance Measure is also often used in clinical practice. Only the LIFE-H has defined metrological properties with a DM1 population (reliability between evaluation and between assessors). The LIFE-H documents the manner in which people carry out activities of daily living and social roles. It is a generic tool that takes into consideration the individual's subjective perception regarding the disruption in the accomplishment of a specific life habit such as preparing a meal or doing volunteer work [25]. Although based on a different conceptual model, the LIFE-H [24] is among the instruments that capture most of the items of the ICF participation dimension when compared with several participation measures [26]. The LIFE-H demonstrated adequate validity [27]. The LIFE-H demonstrates high to moderate test-retest and inter-rater reliability when used with a DM1 population [28].

B) Evaluation of Performance Components (personal factors)

DM1 being a progressive disorder, evaluation of performance components should be evaluated within a functional state of mind and strongly related to occupational performance priority area identified by the person with DM1.

Evaluation of sensory and neuromuscular performance components

Decreased muscle strength is the hallmark feature of all neuromuscular disorders. However, in DM1, other symptoms often precede the decrease of muscle strength. In DM1, slowly progressive muscle weakness is present with a pattern of distal to proximal involvement. In addition, facial weakness, atrophy, ptosis, and weakness of the sterno-mastoid and neck flexor muscles, long finger flexors and foot dorsiflexor muscles are the earlier muscular features of DM1 [29]. Myotonia is a frequent presenting symptom (36 -75.9%) [29, 30]. Upper extremity range of motion will often be affected in relation to decreased muscle strength but no treatment has been shown to be effective. Again, endurance, gross coordination, postural control, fine coordination and dexterity are affected but no treatment has been shown effective. Reflexes are also preserved in DM1. Sensory testing is rarely necessary as myotonic dystrophy has not been associated with any sensory involvement apart from cold sensitivity where counselling can be given [29]. Soft tissue evaluation is rarely of concern.

Evaluation of perception and cognition

Evaluation of perception and cognition is usually done by a neuropsychologist. The OT can provide a unique contribution in evaluating the effect of cognitive-perceptual impairments on participation in daily activities and social roles [21]. In DM1, special attention should be devoted to fatigue, hypersomnolence, executive function and apathy.

Evaluation of psychosocial skills and psychological components

Evaluation of psychosocial skills and psychological components includes the ability to interact in society and to process emotions [21]. It is necessary to gain knowledge about these components in order to help clients maximize function. It includes psychological skills (values, interests and self-concept), social skills (role performance, social conduct, interpersonal skills, and self-expression), and self-management (coping skills, time management and self-control). In the context of DM1, knowledge about these concepts should be gained in order to interpret social participation in relation to well-known features of DM1 which could be present such as avoidant personality traits [31], diminished affect and few interests.

C) Evaluation of Performance Contexts (environmental factors)

DM1 being a progressive disorder, the role of environmental factors and especially the implementation of community services (home services, meal preparation, nursing at home, budget management, etc.) should not be underscored as these are most probably effective measures for alleviating some of the consequences and burden imposed by the disease [3]. As a group, DM1 patients show poor academic achievement, high unemployment, low family income, and high reliance on social assistance compared with the general reference population thus confirming a socioeconomic disadvantage [13; 14] Using socio-spatial modelling of a Saguenay-Lac-Saint-Jean urban area, DM1 was found to be six times more prevalent in disadvantaged neighbourhoods compared with advantaged ones [32.] Such patterns of residential segregation impose a double burden on deprived people: they not only have to struggle with many problems arising from their own lack of income but also they have to live with the social effects of residing in a neighborhood where the majority of their neighbours are also poor [33]. Such a phenomenon can play a role in the perpetuation of poverty in DM1 and can contribute to social exclusion and isolation [33]. Residents of extremely poor neighborhoods often report the absence of regular sources of social support, including a marital partner and close friends. Also, people who receive less social and emotional support from others are more likely to experience less well-being, more depression, and higher levels of disability from chronic diseases [34]. The perception of negative support and attitude of family and friends was an explanatory factor for level of participation in work, leisure and mobility. The perception of obstacles related to access and use of technology and government services is also related to level of participation [8].

OT INTERVENTION AREA

Recommendations are usually based on clinical practice because there are very few studies in occupational therapy. Based on the findings from qualitative studies, a recent review of the literature [1] recommended a client-centered approach that includes the following aspects: educating the patient about the disease because education plays an important role in his or her understanding of the need to implement adaptive strategies (Jönsson et al., 1999; Nätterlund & Ahlström, 1999; Young, 1989); evaluating the patient's perception of his or her life history, personal values, goals, and problems (Jönsson et al., 1999); informing the patient about the adaptive strategies available; and identifying the patient's adaptive strategies, which can be used in occupational therapy to empower the patient to make changes in his or her occupational performance (Jönsson et al., 1999). Occupational therapy interventions had to include training of activities of daily living, skills training (fine motor skills), advice and instruction in the use of assistive devices, provision of splints and slings, counselling on energy conservation strategies, educating patients, families, and caregivers or a combination of the above.

A) Occupational performance treatment

The purpose of OT treatment is to help clients learn or relearn occupational performance that they need to live as independently as possible [21]. Treatment strategy will be mostly geared toward compensation and education. For the compensation approach, three options can be explored with the client: 1) Alter the task method; 2) Prescribe assistive devices or; 3) Adapt the task environment [21]. Education can be a real challenge with DM1 in relation to cognitive functions. From a large study, the highest level of dissatisfaction is related to participation in work and leisure as long as engaging in physical fitness activities (40 – 25 % are highly dissatisfied).

Personal Care: Specific problems that are often encountered may relate to upper limb function (such as picking up a cup, washing hair, wiping oneself after going to the toilet, doing up buttons), to lower limb function (such as walking to the toilet, standing in the shower) or both (such as putting on trousers, getting into or out of a bath). Multisystemic complications such as diarrhea, anal incontinence and dysphagia, may add to these - so considering ameliorating them can assist in personal care. These aspects are usually approached in a problem-oriented way by suggesting adaptive techniques, specialized equipment and/or community services [35].

Mobility: Some patients have an early and severe involvement of the knee extensors, complain of multiple falls and are rapidly wheelchair dependent. Patients who require wheelchairs will often have moderate to severe proximal and truncal weakness. Therefore, when the wheelchair is prescribed, attention must be paid to stability and posture while seated, the ability to stand from the chair and the ability to transfer [35]. The need of an electric wheelchair or a four-wheel scooter can also be explored, although their use is less frequent.

Work: No specific treatment option has been explored.

Leisure: No specific treatment option has been explored.

B) Performance components treatment

Muscle strength: Only strengthening of the hand muscle has been tried in OT in one study with five subjects that is insufficient on which to base clinical guidelines [36]. A recent Cochrane Collaboration review came to the conclusion that in myotonic dystrophy moderate-intensity strength training appears not to do harm but there is insufficient evidence to establish that it offers benefit [37].

Myotonia : A recent Cochrane Collaboration review concluded that due to insufficient good quality data and lack of randomised studies, it is impossible to determine whether drug treatment is safe and effective in the treatment of myotonia [38].

Dysphagia: Conclusion of a Cochrane Review reported that no trial has adequately evaluated treatments in the management of dysphagia for chronic muscle diseases.[39] They reported that the main treatment options are mostly based on stroke population and include dietary manipulation, adoption of safe swallowing techniques, surgical intervention and enteral feeding. No single universally effective treatment for dysphagia in DM1 has been described. This probably reflects the many different mechanisms underlying dysphagia in DM1 [40]. Strategies to facilitate pharyngeal functions in DM1 patients include: 1) strict adherence to reflux precautions; 2) education of friends and family members in the performance of the Heimlich manoeuvre; 3) dietary counselling emphasizing maximum nutritional density within a restricted range of consistencies

(identified as safest, most effective); 4) strategies to facilitate pharyngeal clearing, i.e., careful chewing and bolus preparation to a liquid consistency, repeat swallows, alternating thin with thick consistencies (if possible without aspiration); and 5) airway protection strategies when aspiration risk is elevated [41].

C) Performance context environment

Provision of education and information has been stressed as a mean of assisting patients and families to cope with neuromuscular diseases [42]. Supportive relationships, whether in the form of practical help, emotional support, or provision of information, may facilitate health-promoting behaviours in patients with DM1 and should be encouraged as such [14]. At the community level, designing facilities to promote social interaction may reduce social isolation in patients with DM1. In this respect, belonging to a lay association has improved the level of well-being in patients with DM1 [43].

OT INTERVENTION EFFICACY

Cup & al, (2008)[1] performed an extensive review of the literature to assess whether there is evidence for occupational therapy for patients with neuromuscular diseases. The initial search strategy resulted in a total of 3,534 citations but after screening the majority of the studies (3,528) they did not meet the predefined criteria. Six full-text articles were retrieved and from them, only one was concerned with myotonic dystrophy type 1 [1]. The objective was to evaluate an individualized hand training program with silicone-based putty in five patients with DM1. There was improvement in self-rated performance and satisfaction with performance using the Canadian Occupational Performance Measure [36], as well as muscle strength increase and fine motor control but not grip force and pinch strength. This study provides some indications for the efficacy of a hand-training program in muscle disease with at least 3 of 5 manual muscle tests in the wrist and hand. The intervention was a 12-week, 3 times a week, 45-minute regimen with silicone-based putty and a stretching program.

EVALUATION OF THE NEED FOR REFERRAL TO OCCUPATIONAL THERAPY

Perceived Limitations in Activities and Needs Questionnaire (PLAN-Q) [44, 45]. It is a screening tool to select those patients with NMD that need referral for a one-time consultation by OT, PT and ST. The PLAN-Q only screens patient-opinions and the results demonstrated that the results were not reliable from one time to another. Indeed, patients change their need for referral within a two weeks period.

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