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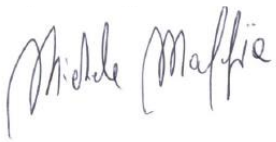

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Obstructive Sleep Apnea (OSA), an emerging health problem

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Abstract
Obstructive Sleep Apnea (OSA) is the most common respiratory disorder in Western societies: according to a first recent worldwide epidemiological study, it was estimated that 936 million patients aged 30-69 years with mild to moderate OSA and 425 million patients aged 30-69 years with severe OSA requiring CPAP treatment.
Recently, the Centre for Research on Health and Social Care Management (CERGAS) at the Bocconi University of Milan has estimated that in Italy, the prevalence of moderate to severe OSA occurs in the 27% of the general population, with an overall prevalence of mild to medium-severe OSA of more than 24 million people aged between 15 and 74 years (54% of the adult population), while from a practical point of view, Italian doctors diagnosed only 460.000 moderate-severe patients (4 per cent of the estimated prevalence) and 230,000 patients were treated (2 per cent of the estimated prevalence), highlighting a substantial gap between diagnosis and treatment.
In addition, OSA patients are often obese and the close correlation between the two conditions suggests that the prevalence of OSA will increase in the short term as obesity increases. At the individual level, OSA leads to a significant decrease in quality of life (HRQoL) and intellectual and mechanical/functional capacities with reduced physical activity, as well as a marked increase in sudden death and risk of cardiovascular and metabolic diseases.
Emerging epidemiological data also suggest that the severity of OSA associated with the severity of chronic nocturnal hypoxemia (CIH) correlates with an increased risk of diabetes mellitus, metabolic syndrome (MS) and cancer. OSA is also an important risk factor for high blood pressure, acute and chronic atrial fibrillation (FAC), chronic coronary artery disease (CAD) and stroke.
It is therefore intuitive that at the social level, OSA also leads to a decline in economic productivity.
This article addresses OSA from a new epidemiological perspective, according to the latest prevalence studies, and addresses emerging problems related to the diagnosis.

Keywords: obstructive sleep apnea, OSA

I. Introduction

Obstructive Sleep Apnea (OSA) is defined as a very common disorder in the general population characterized by loud and habitual nocturnal snoring with repetitive upper airway obstruction (apnea) (Paul E Peppard et al. 2013; Heinzer et al. 2015) sometimes accompanied by daytime sleepiness. The severity of OSA is generally measured by the Apnea-Hypopnea Index (AHI) which defines the number of nocturnal respiratory pauses (apnea) or partial and/or incomplete pauses (hypopnea) and/or obstructive events measured per hour of sleep.
The diagnostic definition of OSA is obtained from both polysomnography recording (PSG) from which the AHI index, which must be ≥5 events/hour of sleep, and from the night-time

cardio-respiratory monitoring (MCRN). In the best-known American epidemiological study, called the Wisconsin Sleep Cohort Study, the prevalence of OSA in the general American population was estimated to be around 24 percent in men and 9 percent in women between 30 and 60 years of age (Punjabi 2008)
Current prevalence predictions indicate a worldwide increment and suggest that OSA is present in both developing countries and Western societies (Kapur 2010)
New data from a first worldwide epidemiological study published in August 2019 on Lancet carried out in 16 countries, and from the analysis of the results of 17 new epidemiological prevalence studies, all carried out with the new instrumental diagnostic criteria of the American Academy Sleep Medicine (AASM) of 2012 (new diagnostic definition of hypopnea and arousal) suggest that the prevalence of the disease worldwide can be estimated at around 936 million patients with mild to moderate OSA, (almost one billion!) aged 30-69 years and 425 million patients with severe OSA aged 30-69 years who need night-time CPAP treatment (Benjafield et al. 2019)

II. OSA and obesity: a close relationship

Obesity is an epidemic disease with a rapid progression in Western society (Armeni et al. 2019). Obesity, defined as body mass index (BMI) >30, is the most important risk factor for the onset of OSA, in particular when the fat distribution is prevalent in the thoracic-abdominal area. In the multi-center Wisconsin Sleep Study, it is reported that in OSA patients body weight gain over a 4-year period is an important predictor of OSA progression; the study showed that in OSA patients, a 10 percent increase in body weight resulted in a 32 percent increase in the AHI metric index, with a relative 6-fold increase in the risk of developing an even more severe form of OSA (Paul E. Peppard et al. 2000) Another multicentre epidemiological study, Sleep Heart Health Study, conducted on native Americans over 65 years of age, also showed that a 10 kg weight gain over a 5-year observation period resulted in a 2.5-fold increase in AHI with a 5.2 percent increase in the risk of developing OSA-related cardiovascular disease (Newman et al. 2005)

In addition, according to another epidemiological study carried out in bariatric surgery, OSA is present in 41 percent of patients with BMI >28 and 78 percent of severe obese patients with BMI >35 who have to undergo bariatric surgery (Domenico Maurizio Toraldo et al. 2013; Lopez et al. 2008). In obese patients with severe OSA, obesity is considered responsible for sleep deprivation and changes in sleep architecture, which leads to comorbid insomnia, which worsens the quality of sleep and life of obese OSA patients (Spiegel et al. 2005). The alterations of the diet and of the alimentary style in paediatric age, can determine alterations of the neuro-hormonal regulation of leptin and greline that can favour, with the years, the development of OSA. A series of clinical studies have demonstrated in the obese OSA patient the presence of elevated serum levels of functionally altered neuro-hormonal leptin; leptin is a hunger inhibitor and intervenes in the regulation of body weight together with greline and affects the neurological centers of breathing during sleep; treatment with continuous positive airway pressure (CPAP) determines an improvement in serum leptin levels with an improvement in obesity and respiratory activity during sleep (Chen et al. 2015; Phillips et al. 2000; Ong et al. 2013).
Recently, a longitudinal controlled clinical study was conducted in Sweden on more than 3,400 severely obese patients with OSA, using laparoscopically adjustable gastric banding to reduce body weight in severe OSA. In this study, the decrease in BMI resulted in a marked improvement in OSA symptoms (quality of life and daytime sleepiness) and in a decrease in the use of drugs for the treatment of diabetes mellitus 2 (DM) and metabolic syndrome (MS). In another clinical study of severe obese patients (BMI >35) with severe OSA (AHI >30/hs), bariatric surgery resulted in significant weight loss and a decrease in OSA severity evidenced by a reduction in AHI from 80 to 17.7 ±10.0 obtained after about 5 months of surgery (Dixon, Schachter, and O'Brien 2005) However, the majority of OSA patients who underwent bariatric surgery, after surgery, after about 6 months demonstrated a significant residual OSA (AHI >15) at a control PSG, and special attention should be paid in the protocol for the repetition of PSG in the follow-up of operated

patients and for CPAP treatment if the AHI index should be >10 (Greenburg, Lettieri, and Eliasson 2009; Lettieri, Eliasson, and Greenburg 2008). Leptin is therefore a hormone involved in appetite suppression, it is released from adipocytes located in abdominal fat and is produced in proportion to the abdominal fat content (Münzberg and Morrison 2015); it acts on respiratory centers to stimulate ventilation in response to peripheral hypercapnia in OSA with the aim of reducing the increased PaCO2 and normalizing PaO2. Physiologically ineffective leptin increase has been associated with worsening of OSA and metabolic syndrome (D M Toraldo et al. 2015; Kalra 2008).

III. Genetic factors

The OSA recognizes multiple predisposing factors of a genetic family type. Given the complexity of the syndrome, it is not possible to recognize a single genetic factor that can determine its manifestation, but a set of genetic-biological factors. The risk of OSA is genetically predisposed in 40% of cases (Kent, Ryan, and McNicholas 2010; Redline and Tishler 2000).
The prevalence of OSA in first-degree relatives of OSA patients ranges from 22 to 84 percent (Mathur and Douglas 1995). Anatomical risk factors for OSA, such as obesity and structural changes in the soft tissues of the upper first airway such as macroglossia and mandibular hypoplasia, show familiarity. Obesity and upper airway alterations often occur from early childhood and studies of monovular twins have shown 57-86 percent heritability from early youth (Silventoinen and Kaprio 2009)
Another study in a Scottish population identified a strong family genetic component for OSA and suggested that hereditary craniofacial malformations were more important than obesity itself (Mathur and Douglas 1995). Indeed, the morphology of the pharyngeal lateral wall, the properties of the craniofacial mucosa and neck fat have a significant level of heritability and therefore susceptibility for OSA.

IV. Economic aspects and quality of life

OSA has a significant economic impact on Western public health systems and must be considered and managed as a chronic condition and not, as is the case in Italy, as an acute pathology (the recognition of chronic pathology implies the adoption of an ad hoc government law). OSA is burdened by direct costs, which are the healthcare costs necessary for the diagnosis and treatment of the disease and the associated comorbidities as well as for the use of hospital admissions, and by indirect or social costs, very high, due to a reduction in work productivity, permanent disability due to road or work accidents. This is compounded by the overall economic impact of the OAS, which is very high.
In the United States, according to an article by the American Academy Sleep Medicine (AASM) of 2016 (Nieto et al. 2012) the annual cost for an undiagnosed OSA patient is estimated to be around $5,500 (considering direct and indirect health care costs), while in diagnosed OSA patients it drops to $2,100 per year (Campos-Rodriguez et al. 2013b) Patients with sleep disorders are less productive than those without such disorders and have a higher level of absenteeism and decreased productivity at work due to fatigue, chronic fatigue and excessive daytime sleepiness (Sherman 2013) Occupational accidents are also more common in patients with OSA (Alghanim et al. 2008) Several studies have shown that the quality of life measured with QOLi questionnaires is unfavourable in OSA patients compared to non-OSA patients and improves after various medical, ventilatory, dental and surgical therapies (Alghanim et al. 2008; Smith and Shneerson 1995; Jenkinson, Stradling, and Petersen 1997). The bedfellow of an untreated OSA patient may also suffer from insomnia due to loud snoring at night. The QOLi of the untreated snoring OSA patient's bed partner significantly improves when the OSA is successfully treated (Gall, Isaac, and Kryger 1993; Parish and Lyng 2003).

V. OSA and cardiovascular disease

The strongest scientific evidence supporting an independent role for OSA in the promotion of early atherosclerosis and the development of adverse cardiovascular outcomes (cerebral strokes, myocardial infarction and complicated vascular aneurysm) can be found in clinical

studies addressing the relationship between OSA and arterial hypertension (Paul E. Peppard et al. 2013) . There are controlled clinical trials that have shown a correlation between OSA severity and the likelihood of developing hypertension with its complications (Hedner et al. 2006; Javier Nieto et al. 2000)
Recent European data show that out of 11,900 patients participating in the European study called Cohort Apnea Study (ESADA), chronic intermittent nocturnal hypoxemia with deep desaturations is a predictive factor for early vascular atherosclerosis and that it can be a key factor for the development of arterial hypertension and early vascular endothelial damage (vascular aneurysm) (Tkacova et al. 2014). The results of another clinical study have shown an increased propensity in OSA patients for the development of hypertension and cardiovascular complications (P E Peppard et al. 2000). In another epidemiological study, carried out on 709 subjects, the presence of severe OSA conferred a very high risk to diagnose hypertension, regardless of other factors such as diet, obesity and smoking history. Other recent clinical data from a Spanish study also confirmed this same relationship (José M. Marin et al. 2012; Kent et al. 2013)
With regard to chronic ischemic heart disease with CAD, recent data suggest that CAD is very widespread in patients with OSA and constitutes about 30 percent of patients with CAD, and vice versa that patients with CAD are more likely to develop an obstructive pathology during sleep type OSA (Mooe et al. 1996). In another epidemiological study called Health Study carried out on more than 6,000 patients, the severity of CAD (measured as atherosclerotic plaque diameter) was correlated to the severity of the AHI metric index (SHAHAR et al. 2001)
In addition, patients with OSA are at high risk of developing paroxysmal atrial fibrillation (FAP); in another study involving 566 patients with chronic ischemic heart disease, severe OSA was found and this was associated with a fourfold probability of developing FAP (Mehra et al. 2006).
In OSA patients, the increased diameter and size of the left cardiac atrial chamber determines an increased presence of risk factors for the appearance of FAP and subsequent failure of the subsequent pharmacological cardioversion (Drager et al. 2010). In addition, recurrence of FAP becomes less likely if it is significantly reduced with the initiation of CPAP treatment (Kanagala et al. 2003). Obstructive sleep pathology is also an independent predictor of ventricular arrhythmia, particularly in subjects with chronic heart failure (Monahan et al. 2012).
OSA is much more common in patients who have had a recent stroke or transient ischemic attack such as TIA, in 32-63 percent of patients compared to the general population, and is associated with increased mortality and worse functional outcomes over time (Bassetti and Aldrich 1999; Good et al. 1996). In addition, the prevalence of cerebrovascular disease seems to increase with increasing severity of OSA. Some prospective studies suggest that the presence and severity of OSA is a predictive factor for stroke (Arzt et al. 2005; Yaggi et al. 2005; Redline et al. 2010).

VI. OSA and cancer

The fundamental pathophysiological mechanism that characterizes OSA is represented by chronic nocturnal intermittent hypoxia (CHI) that generates a chronic inflammatory cascade that causes a diffuse vascular epithelial damage of pro-atherogenic type (Garvey, Taylor, and McNicholas 2009) the same chronic inflammatory mechanism could promote the development of cancer through the release of angiogenic factors.
In a murine model of OSA, the possibility of developing malignant tumors towards blood or skin cells (malignant melanoma) by triggering mechanisms of systemic inflammation, oxidative stress and immune dysregulation has been demonstrated (Isaac Almendros et al. 2013). Data from these animal models suggest that exposure to CIH in humans may determine the origin of malignant cellular neoformation and increase distant metastatic progression and spread (I Almendros et al. 2012; Nieto et al. 2012)
Therefore, several longitudinal clinical studies in humans have been proposed that have assessed the association between OSA and cancer. In an analysis of 1,522 patients participating in the Wisconsin Sleep Cohort Study, subjects with severe OSA were at almost five times the

risk of developing cancer (Campos-Rodriguez et al. 2013a). Similarly, in another Spanish clinical study of nearly 5,000 OSA patients, the severity of the disease expressed by the intensity of nocturnal hypoxemia predicted the development of malignant neoplasms in different body districts (Kendzerska et al. 2014). A Canadian retrospective cohort study has shown that there is a close relationship between OSA and cancer, and among other things within this population an independent association between smoking-related cancer and severity of chronic nocturnal hypoxemia has been demonstrated (Young et al. 1993). To date, no data have been published about the effect of CPAP therapy on cancer, and potentially innovative clinical trials are being conducted in this area.

VII. Diagnosis Considerations

Clinically defined OSA syndrome in combination with excessive daytime sleepiness (ESD) and obesity is estimated to increase in the coming years in both women and men due to increased obesity (Al Lawati, Patel, and Ayas, n.d.) OSA, a chronic disease, if left untreated is associated with an increase in overall mortality and a number of serious social and health consequences such as reduced quality of life, reduced productivity in the workplace and increased road and work accidents (De Benedetto, Garbarino, and Sanna 2017; Jose M. Marin et al. 2005)

The gold standard diagnostic investigation is complete, stationary polysomnography (PSG) with neurological and video-surveillance electrodes that is performed in a sleep laboratory (LS) and is the preferred method to diagnose OSA correctly and completely (D M Toraldo et al. 2017). The treatment, not pharmacological, is the application of CPAP device (continuous positive pressure in the upper airway) which is performed in LS with high health care costs (manual night titration supported by a neurophysiology technician). Due to the high costs and long waiting times to perform the examination, new simplified but equally suitable and important methods for home sleep testing (HST) have been developed. These are low-cost, portable and non-permanent instruments, defined as polygraphs (night cardiorespiratory monitoring, MCRN) which, in addition to making the diagnosis, also offer the possibility of home treatment by self-titling positive airway pressure (APAP). A number of studies compared the complete polysomnography performed in LS with HST, confirming the diagnostic non-inferiority of the latter, and also validated home treatment with CPAP device. The diagnostic and therapeutic non-inferiority of HST vs LS has also been confirmed by other studies (D M Toraldo et al. 2017)

The metric index expressed as AHI/hs is used to diagnose the degree of OSA severity by polygraph/polysomnography recording. A distinction is made between mild (5-15 events/hour), moderate (15-30 events/hour), severe (>30 events/hour) and very severe (>60 events/hour). A threshold value of 5 is considered a sign of normalcy in adult patients.

In the case of AHI <5 the patient can be considered negative for the diagnosis of OSA, unless the daytime sleepiness symptom is present, in which case the patient should undergo LS night polysomnography (doubtful cases). The test is considered positive for the diagnosis of OSA if AHI ≥5 (2,16,17).

In conclusion, OSA (Passàli et al. 2014; Costa and De Benedetto 2017) leads to impairment of quality of life, increased risk of cardiovascular and metabolic complications and poor work performance as well as the possibility of causing road and work accidents.

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The Phonetics of Speech Production and Medical Research

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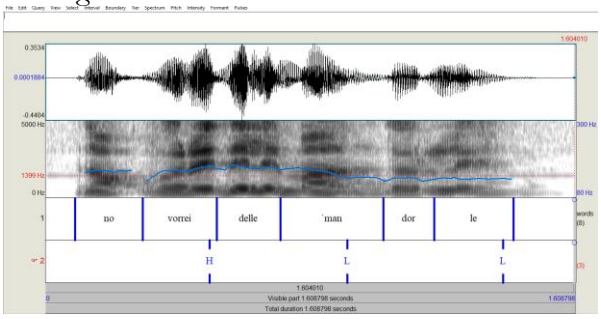
Abstract
The production of speech requires the interplay of a number of cognitive and motoric activities, which make it an interesting object of study from both a linguistic and a medical point of view. In this paper, we discuss, first, the features and domain of application of the most used technologies in linguistic research on speech production, focusing on those that have been applied to medicine. Second, we offer an insight into the main results that have been obtained so far in studying dysarthria in Italian Parkinson's Disease, as an example of the interdisciplinary, experimental research at the border between linguistics and medicine.

Keywords: 3D articulography, ultrasound, phonetics, phonology, medicine, dysarthria, Parkinson's Disease.

1.1 The analysis of speech production

Investigations on speech production may rely on acoustic analyses, which offer information on the issue, though do not allow a direct and detailed observation of the production mechanism. Nowadays phoneticians easily perform acoustic analyses, thanks to the diffusion of recording facilities and dedicated software. However, acoustic data only represent the starting point of studies whose aim is to deeply investigate speech production. Therefore, a typical speech production study involves the recording of both acoustic and articulatory material, and the following analysis of both types of data. Crucially, data acquisition has to be synchronized (via hardware or post-processing) in order to shed light on the articulatory mechanism responsible for the production of linguistic sounds. The following part of this section offers and overview of the main software nowadays used for acoustic investigation, as well as the main technologies exploited in most of the current studies on speech production.
Acoustic analysis, as well as acoustic recordings, may be performed by means of PRAAT (Boersma and Weenink, 2020). As for speech analysis, the software allows the user to segment the audio signal and to time-align multiple levels of text labels, such as those regarding consonant and vowel boundaries, as well as the presence intonational events, words, phrases and larger constituents – Figure 1. The system also allows to semi-automatically performing a wide range of acoustic measurements with reference to the abovementioned labels, i.e. points in time. Crucial information on speech may be already collected by means of this type of material and analyses.

Figure 1: *PRAAT – waveform, spectrogram with overlapping fundamental frequency track, and two levels of time-aligned labels*



However, investigations on speech production nowadays often relies on the use of appropriate, in some cases purpose-built, technology.

1.2 Electromagnetic articulography

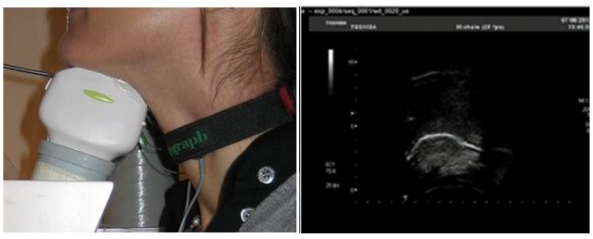
An example of purpose-built technology is the ElectroMagnetic Articulograph (EMA). Various systems are described in the literature (Kaburagi et al. 2005, Stella et al., 2012, Stella et al., 2013), which are used to track the movements of a set of sensors glued on the main speech articulators, such as tongue and lips (see Figure 2, right panel), as well as on more stable parts, such as the front teeth or the forehead, to compensate for head movement. For instance, the Carstens Medizinelektronik GmbH's system AG501 (http://www.articulo graph.de) is used to identify the Cartesian coordinates x, y and z, as well as azimuth and elevation of directionally sensitive magnetic field sensors (8 to 24) at a sampling frequency of up to 1250Hz in real time. The measuring sensors are single axis coils. Nine reference coils, along three arms (blue arms in Figure 2, left panel), arranged to form a three-dimensional frame of reference, emit magnetic fields at different well known frequencies between 7500 and 13750 Hz. During a recording session, the alternating currents induced in the sensors by the magnetic fields of the reference coils are separated by their frequencies, digitized and sent in real-time to the control unit. Dedicated software stores the current values, making them available for the spatial arrangement determination process.

Figure 2: *Speech recording with AG501 (left) and sensors glued on the subject's tongue and lips (right)*

1.3 Ultrasound tongue imaging

Articulatory studies of tongue motion are becoming popular in phonetics, thanks to the adoption of ultrasound systems which have been used for clinical purposes and have been eventually adapted to suit the investigation of speech production. Such systems, which are both non-invasive and non-obtrusive, are able to provide the profile of the tongue during speech production, although the image of tongue apex and radix may be often occluded by the presence of the jaw and the hyoid bone respectively. Ultrasound images are obtained thanks to a high frequency (2-14 MHz) sound waves emitted from an array of piezoelectric transducers (crystals), multiplexed in time: only one crystal emits sound waves in a given time interval, while all the remaining crystals are used to convert the received echoes to voltage values. The ultrasound wave goes through tissues and is reflected when it reaches an interface between tissues or materials with different impedance properties. Ultrasound images are reconstruction of such interfaces, thanks to the processing of the voltage values of the received echoes. In the case of tongue imaging, the probe is placed under the chin and the wave goes upward, though the tongue (Figure 3, left panel). When the wave reaches the upper surface of the tongue, where the impedance changes (think of the mucous membrane and the air in the oral cave), it is reflected to the probe and the surface of the tongue may be reconstructed (Figure 3, right panel). The tongue profile during speech production may be available as a sequence of images that are time-aligned with the audio recording.

Figure 3: *Ultrasound probe position during speech recording (left) and example of ultrasound tongue image (right)*



Nowadays, increased sampling rates allow researchers to get a more convenient number of frames per second during speech, as producing the verbal chain requires quite a high-speed sequence of gestures. For instance, an Aplio XV machine, by Toshiba Medical System corp. (http://www.medical.toshiba.com), allowed us to collect the first ultrasound data related to speech in Italy (Grimaldi et al., 2008). At that time, during each recording session, the ultra-

sound pictures were exported as a continuous video stream (at 25 Hz), by means of a dedicated S-Video output; such stream was acquired together with the audio signal (synchronously), by means of an external a/v analog-to-digital acquisition card, and then recorder in real-time on a dedicated PC. Nowadays, more compact and fast systems may be used, that do not require the realization of a constellation of hardware and software means to ensure the acquisition of all the information needed to analyze speech. Systems such as the "Micro Ultrasound system for speech research", proposed by the Articulate Instruments Ltd (http://www.articulateinstruments.com/alan-wrench/), for instance, may include the hardware synchronization for audio capturing and the AAA software for data analysis.

Linguists, phoneticians, and laboratory phonologist in particular use the abovementioned technologies to test their hypothesis on the linguistic organization of speech production. The way speech gestures are realized and phased with respect to each other is investigated with respect to various languages, as well as with respect to the changes in their organization in the case a second, or foreign, language is produced. However, the testing of the very same linguistic hypotheses, or a better understanding of speech production in general, may be of interest of medical research too (for an overview, see Gili Fivela, Zmarich 2013).

2. Articulatory studies and speech pathology

For many decades, the description, explanation and rehabilitation of various speech articulatory disorders have been based on data derived from phonetic transcriptions, based on transcriber's perception, and acoustic analysis of pathological speech. However, these methods reveal limitations concerning the description, the explanation and the rehabilitation of speech pathologies. As for the former, limits are due to the subjectivity of the auditory perception and of the subsequent evaluation (Shriberg and Lof, 1991), and to the lack of a two-way correspondence between acoustic and articulatory data (Sondhi, 1979). As for the possible explanation, limits are identified, firstly, in the "opacity" introduced by the distance between the cause of the pathology and the measured acoustic or perceptual events, which originate at the periphery of the speech production system; secondly, limits concern the inadequacy of phonetic-phonological theories based on the study of perceptual or acoustic targets, and therefore not suitable to explain motor events of an intrinsically dynamic nature (Weismer, Tjaden, Kent, 1995). Finally, concerning rehabilitation, a limit consists in the inability to provide adequate articulatory feedback, which can be only partially provided by auditory and, even less, by acoustic information.

A useful integration to these traditional methods is then represented by the information on the dynamics and kinematics of speech, that is the description of the movement (e.g., duration, extension) and the description of the physical conditions responsible for a given movement (which, in addition to the already mentioned descriptors, include, e.g., mass coefficients, rigidity, damping; Bingham, 1988). Kinematic and dynamic data offer reliable qualitative and quantitative information about the movements of the articulatory organs (Gracco, 1992); from an explanatory point of view, they can provide a precious alternative to traditional explanations, when framed within a suitable theoretical framework, such as a non-linear dynamic theory (see Port, van Gelder, 1995). For instance, according to Articulatory Phonology, the main unit of the motoric control for linguistic purposes is the so-called articulatory gesture (see the review in Goldstein and Fowler, 2003). The articulatory gesture is dimensioned with respect to the spatial coordinates that represent the vocal tract and with individual quantities that are proportional according to a gestural score that indicates the organization of gestures and the intervals of activation of the tract variables which are relevant to the production (e.g., Lip Aperture or Lip Protrusion, which according to Gestural Phonology – Browman and Goldstein, 1986 - are associated with a series of articulators). The impact of such theory on the investigation of speech pathologies is acknowledged (van Lieshout, Goldstein, 2008), and is tightly linked with experimental investigations performed with the technologies described in the first section of this paper.

In fact, the usefulness of technologies in studying speech articulation has been clear at least since the review by Thompson-Ward and Mur-

doch (1998), on the methods to verify the articulatory capacity in dysarthria, and the review by Barlow et al. (2009), on cinematic measures related to speech. However, as recalled in the latter, Sonoda (1974) already observed the usefulness of the real-time collection of kinematic data by means of orofacial magnetometry, because such data can be used in the rehabilitation of dysarthria, thanks to the visualization of the movement of the articulators. Not surprisingly, the first studies employing ultrasound in the investigation of pathological speech date back to the early 1980s. As Thompson-Ward and Murdoch (1998) recall, Shawker et alii (1984) already carried out a study on non-pathological and dysarthric speakers, and noted that ultrasounds allowed to observe significant differences in the articulation of vowels (/a/, /i/) and consonants (/k/), and could have therefore represented a promising technique. The usefulness of Electromagnetic articulography is also acknowledged and quite widespread (Wong et al., 2010), as for the investigation of various pathologies, such as dysarthria (Rong et al. 2012, Jaeger et al., 2000, McAuliffe et al., 2005; Wong et al., 2010a; Wong et al., 2011), apraxia of speech (Katz et al., 2003, Katz, MacNeil, 2010) and stuttering (van Lieshout et al., 1993, 2004; McClean et al., 2004, McClean, Runyan, 2000; Max, 2004). Further, several corpora of kinematic data have been collected for the investigation of apraxia, stuttering and dysarthria. For instance, van den Berg et al. (2006) for apraxia, van Lieshout et al. (1993), and Ward (1997) for stuttering, and the TORGO database, which includes video, audio and 3D electromagnetic articulation data recordings (AG500) concerning dysarthria (Rudzicz et al., 2008).

Interestingly, a number of studies investigate on the use of technologies, in particular the electromagnetic articulation, in training during therapies related to pathologies that cause articulatory problems in speech. For example, Bose et al. (2001) reported on a one case study on an adult subject suffering from Broca's aphasia and apraxia. They demonstrated the usefulness of the PROMPT system (Hayden, 1984), originally developed for oral language "teaching" and involving the use of auditory, visual and tactile stimuli.

Further, a dynamic field of investigation and application concerns the use of articulatory information for the realization of rehabilitation and training protocols based on biofeedback. The visualization of ultrasound images, for instance, has been successfully used to provide articulatory feedback in the therapy of articulatory problems (Bacsfalvi et al., 2007; Bernhardt et al., 2005). Systems developed from articulographic data are also of considerable interest. In this context, the BALDI systems are worthy of note (Massaro, 2004; for Italian, cf. BALDINI, Cosi et al., 2002, LUCIA, Cosi et al., 2008) and ARTUR (Eriksson, 2005, Engwall, 2008). They are computer training systems used for teaching pronunciation (not only in the case of speech/hearing problems, but also for foreign language learning): thanks to the use of "talking heads", i.e. heads and faces animated by computers, these systems show the user how to produce speech sounds, and help them to self-correct their speech gestures. In particular, ARTUR has been developed on the basis of acoustic, video and EMA data (or MOVETRACK; Branderud, 1985) through the optimization of acoustic-articulatory inversion (Kjellström, Engwall, 2009).

With respect to the use of biofeedback, the contribution of Katz and McNeil (2010) who studied the feedback effect provided in real time to verify its usefulness in apraxic patients is also particularly important. The study, carried out by means of EMA and sensors positioned on the subjects' tongue, describes how it is possible to provide information on the use of biofeedback in real time, regarding the position of the tongue (see also Schulz et al., 2006). The system shows subjects how to reach a target indicated on the computer monitor, and is proved to be a useful aid in the improvement of articulatory problems due to apraxia.

As far as Italian is concerned, the usefulness of articulatory investigations has long been recognized and applied to the analysis of stuttering (Zmarich, 1999 a, b, and following works), and, more recently, to the investigation of dysarthria in Parkinson's Disease.

3. Dysarthric speech in Parkinson's Disease: state of the art of investigations on Italian

Along with the development of Parkinson's Disease, patients often suffer from hypokinetic dysarthria. They show a reduction in the amplitude

and speed of movements (Ackermann, Ziegler, 1991, Duffy, 2005, Darley et al., 1975), which has an impact on speech production too. Besides these very general characteristics, a considerable intra-speaker variability concerning speech abnormalities is observed, which may also depend on various factors, such as the task subjects are asked to perform.

Nevertheless, some established characteristics have been identified. In early stages, there may be mild phonetic impairment, while at later stages articulation becomes less precise, and reading rate becomes slower, with an increased number and duration of pauses which may relate to the difficulties in initiating the articulator movement. Perceptually, hypokinetic dysarthria is characterized by monopitch, monoloudness, reduced stress, imprecise consonants, and inappropriate silences. From a kinematic point of view, a reduction is observed in the movement peak velocity and amplitude of lips and jaw. This is evident from reduced vowel formant transition extents, reduced vowel spaces and reduced consonant spectral distinctiveness (Tjaden, 2008). Besides reduction, incoordination has also been observed, and different gesture coordination relations can imply changes in syllabic affiliation too.

Research in speech production in Italian dysarthric speech by Parkinsonian subjects has been performed by adopting the Laboratory Phonology approach (Pierrehumbert et al., 2000). Specifically, the impact of the disease on speech production has been investigated with reference to its effect on phonological features, rather than by itself. Specifically, most of the analyses performed so far aimed at investigating phonological contrasts involving vowels and consonants, with specific attention to the realization of the geminate vs. singleton differences, as produced by mild-to-severe Parkinson's Disease patients (Gili Fivela et al. 2014, Iraci et al., 2016, 2017a, 2017b, Iraci, 2017, Gili Fivela et al., submitted). Besides the obvious interest in the realization of single vowels and consonants, gemination was chosen in order to get information also on the realization of syllables, as the presence of a geminate rather than a singleton involves a change in syllabic affiliation and a change in the duration of the preceding vowel.

The mail goals in investigating Italian dysarthric speech have been 1) to verify if pathological speakers were able to produce the articulatory correlates of a given phonological contrast, and 2) to identify possible compensatory phenomena speakers may adopt in order to overcoming the motor deficit.

Following the Articulatory Phonology framework's predictions, the hypothesis behind 1) and 2) has been that distinctiveness is not threatened since the vocal tract (which is seen as a dynamical system) would re-organise as a function of the linguistic contrasts to maintain.

In order to reach the above mentioned goals and check the main hypothesis, a corpus of acoustic and articulatory – AG501 – data has been collected, by asking mild-to-severe Parkinson's Disease patients, who developed hypokinetic dysarthria, to read aloud highly controlled sentences. They include minimal pairs differing as for the medial consonant – singleton vs. geminate – and the vocalic composition.

Results of analyses performed so far (acoustics by means of PRAAT and articulatory by means of MAYDAY – Sigona et al., 2015) show that Parkinson's dysarthric speakers show spatial alterations (amplitude of movements) that not necessarily involve a reduction of the range of motion. Specifically, they show even gestures of systematically increased amplitude in the case of horizontal, antero-posterior, displacement of the tongue (Gili Fivela et al., 2014, Iraci, 2017). However, intra-speaker analyses showed that phonological distinctions are preserved as much as possible, through a re-organisation of the vocal tract movements, consisting in re-adjustments of surrounding/secondary articulatory gestures (Iraci 2017, Gili Fivela et al., submitted).

The analysis of the abovementioned corpus is still on-going, and it will be deepened thanks to a funded National project (PRIN 2017JNKCYZ), within which prosodic correlates will also be investigated.

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Mounier Kuhn Syndrome

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Abstract
Mounier Kuhn syndrome or tracheobronchomegaly is characterized by a marked dilatation of the trachea and main bronchi that can extend to the periphery of the lungs. Dilation is due to an alteration of the cartilaginous and membranous structure of the trachea and bronchi. It is a pathology much more frequent in men. The aetiology is unclear. The clinical case in question concerns a 67-year-old man who has reported recurrent epi-sodes of bronchopulmonary infections and hemoptysis , after undergoing instrumental investigations (fibrobroncoscopy and chest CT), the diagnosis of Tracheobroncomegalia or Mounier Kuhn syndrome was formulated.

Keywords: Tracheobroncomegalia, bronchiectasis, diverticula, clearance, chronic inflammation, atrophy, connective tissue disorders (Ehlers-Danlos syndrome and Marfan syndrome)

I. Introduction

Mounier Kuhn syndrome or idiopathic tracheobroncomegalia is a rare disease first diagnosed in 1932. About 300 cases have been described in the literature in the world. The clinical symptomatology is common to that of COPD and bronchiectasis, so it is assumed that the incidence of the disease is greater because it is undiagnosed (underestimated). This condition occurs more frequently in males than in females. The disease is characterized by an abnormal dilatation of the tracheobronchial tree, which can extend all the way from the larynx to the periphery of the lung. The tracheobronchial mucosa has a chronic inflammation and a remarkable atrophy that determines the alteration of the mucociliary system, this favors a variation of the clearance of the secretions, which, together with an ineffective mechanism of cough, as in bronchomalacia, facilitates the stagnation of secretions and it is responsible for recurrent infections (pneumonia), bronchiectasis and diverticula mainly of the pars membranacea). Although the aetiology is not clear, its association with connective tissue diseases, such as Ehlers-Danlos syndrome, Marfan syndrome and cutis laxa, suggest an underlying defect in elastic tissue. From the anatomopathological point of view, both the cartilaginous and membranous portion of the trachea and bronchi are involved, both presenting a thin muscular and elastic atrophic tissue. Therefore, the tracheobronchial airways are dilated during inspiration and collapsed during expiration. The disease may present at all ages but is usually diagnosed in the third or fourth decade of life in adults with recurrent bronchopneumonia. The diagnosis can be made thanks to the use of chest CT and fibrobroncoscopy. The treatment of this pathology is symptomatic. Chest physiotherapy can be proposed to improve mucociliary clerance and antibiotic therapy to treat lung infections that can sometimes lead to the appearance of bronchial suppuration with respiratory failure. Differential diagnosis includes bronchitis, chronic obstructive pulmonary disease, bronchiectasis, William Campbell's syndrome or trachebroncomalacia.
In normal conditions, the dimensions of the trachea are in men the following: transverse portion> 25 mm, sagittal portion> 27 mm. In women, transverse portion> 21mm and in sagittal portion> 23mm. For the bronchi, in women the measurements are linked to right> 19.8

mm and to the left> 17.4 mm; for men on the right> 21, on the left> 18.

II. Clinical Case

He came to my observation at the BRONCOLOGIA service, of the Galateo Hospital of San Cesario in Lecce, a man of 67 years in fair general conditions, non-smoker, repeatedly hospitalized at the above described hospital for recurrent bronchial infections in subject who had been diagnosed with chronic obstructive pulmonary disease (COPD) . The symptomatology was characterized by a persistent cough with low emission of mucopurulent sputum, mild dyspnea on exertion, hemoptysis. Respiratory function tests show only a modest functional deficit of respiratory function parameters (FEV1, FVC and FEV1/FVC).
Blood gas analysis showed a slight reduction of pO2 (76.5 mmHg). Laboratory tests showed an increase in white blood cells (WBC 17,580).

The fibrobroncoscopic examination allowed to find a marked amplitude of the trachea (greater than 30mm in diameter) and of the main bronchi (greater than 25mm in diameter) where there was a partially pale, thin (atrophic) mucosa, in part, congested with presence of bronchiectasis, diverticula of the pars membranacea of the trachea and immediately after the entrance of the left main bronchus, abundant mucopurulent secretions. The CT scan of the thorax confirmed the increase in amplitude of the trachea and main bronchi.

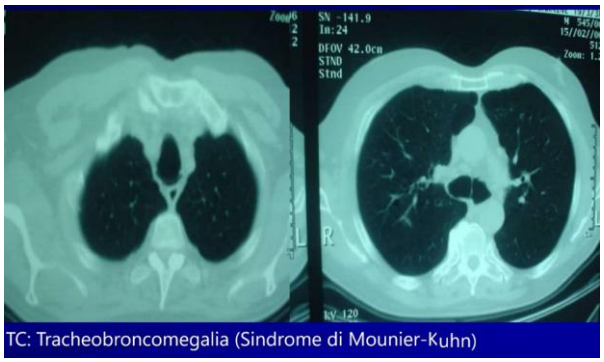

Figure 1. Abnormal dilatation of the trachea and main bronchi

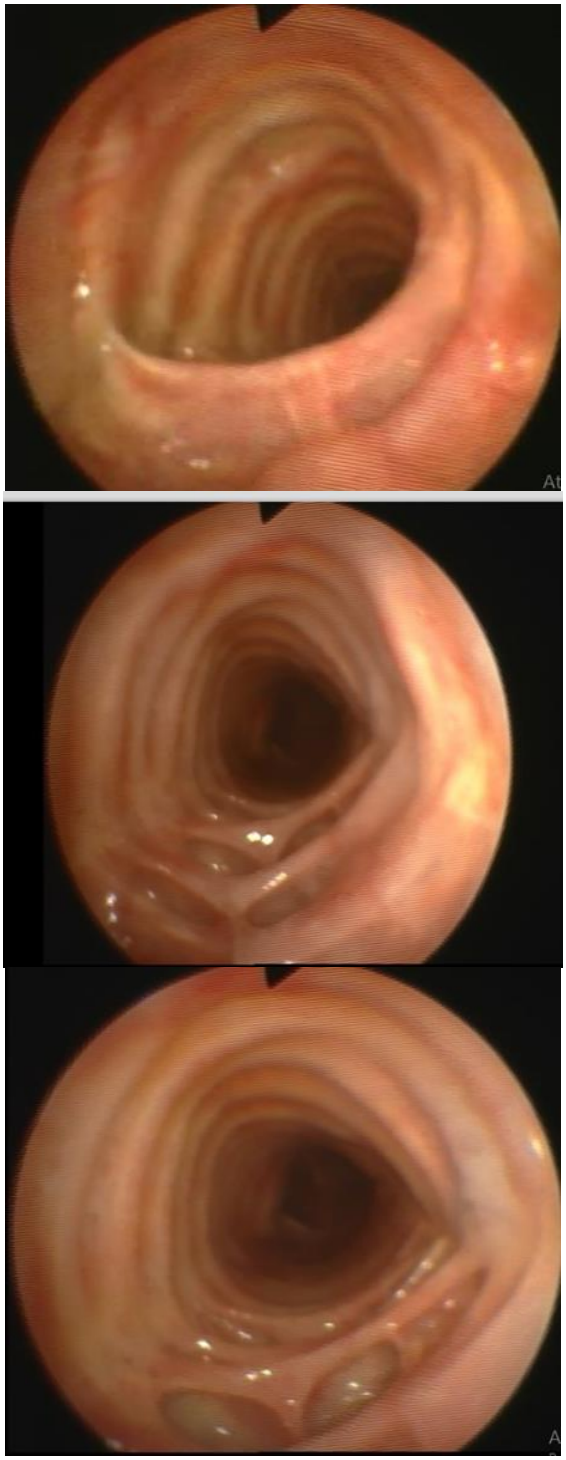

Figure 1. Tracheomegaly with pars membranacea diverticula

Figure 2. Broncomegaly with diverticula located on the posterior wall of the left main bronchus immediately after the carina.

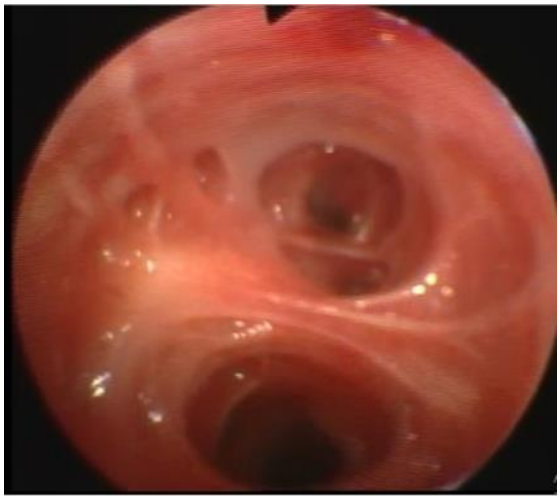

Figure 3. Peripheral bronchiectasis

Figure 4. Mucopurulent secretions present in thesubsegmetary bronchi

III. Conclusions and considerations

The symptomatology of the above described pathology is generally not distinguishable from that of chronic obstructive bronchitis and from bronchiectasis. However, recurrent infections and the presence of a prolonged cough and a loud and hard sound during the auscultation of the chest of patients who complain of an inability to expectorate the secretions should make the diagnosis suspect. Pulmonary function tests typically show a reduction in expiratory flows and a widening of dead space. This pathology cannot be treated surgically due to its extension. However, a symptomatic treatment can be performed (fibrobroncoscopy to aspirate secretions, antibiotic therapy to treat infections, chest physiotherapy and postural drainage to improve the mucociliary clerance).

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E-Health: lacking financial resources and health quality production

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Abstract

The health sector is being studied as an object of individual interest with that of society, in order to pursue equity without sacrificing efficiency, citizens of different Italian regions can access services that are equal in quantity and quality. In health care, the problem from the economic point of view is relevant as the health needs are infinite, while resources are limited and scarce.

In the field of health, the objectives have evolved, passing from the elimination of infectious diseases to the extension of life. The World Health Organization defines health as a right to be counted among other fundamental rights that belong to people.

The analysis of the work carried out focuses on the concept of healthcare seen through the eyes of economists, of how to deal with the fundamental economic questions of what, how and for whom to produce dictated by the distance between need and resources that drives the search for greater productivity and fair re-source allocation systems; discussing the need and expectations for public health intervention.

The Italian Health Service has founded its origins on the recognition of health as an individual right linked to citizenship and as a collective asset to be protected. The financing is based on the principle of the welfare state, which connects the payment to the contribution capacity and the use of services to anyone who manifests a need within the Italian territory.

Since the beginning of the 1990s, a process of reorganization of the health system was initiated, which was mainly oriented towards the research and improvement of tools for assessing the quality perceived by the user. The search for tools that can reduce as much as possible the inconvenience that the citizen may encounter in contact with highly bureaucratic systems and structures such as health care tend to be.

The socio-demographic evolution of the population and the need to balance the available resources with the quality of the healthcare provided, require the definition of new ways of providing health services. In the field of public health, a growing demand for quality in the provision of care and assistance services has intensified in recent years. The increase in interfaces between telematic networks and health and social services is part of the general transformation process of world society due to the progressive increase in the degree of use of ICT services.

The improvement of the quality of health services concerns the management of total quality understood as the management process aimed at establishing the organizational activities that allow the improvement of overall performance.

The success of the organization lies in the continuous and careful communication of the needs related to health by users caused by bad lifestyles, therefore prevention and socio-health policies should reduce the probability of citizens getting sick.

From the analysis carried out, the proposal that is made is that the mission assigned to hospitals on the basis of their classification allows users to be "empowered" to health-related problems without compromising the principles of effectiveness, legal efficiency and health law.

If the goal of our communities is to improve the health level of the population, it is important to promote health policies not only in terms of health services, but also aimed at removing or in any case reducing the impact of a series of risk factors. Therefore, it is hoped that future health policies will increasingly target individual and social risk factors, with health interventions also in sectors such as education, working conditions, transport and nutrition.

Keywords: e-health, risk, health care, empowerment, health quality.

1.Introduction:

The process of health production, input-output and outcome

The health sector, analyzed from the point of view of economists, allows us to study the distribution of "scarce" resources of the community and the multiple needs of citizens. In healthcare, the needs to be met are infinite, therefore resources will never be sufficient in the face of needs to be met. In a civil and advanced country, the user chooses a health system intended as "a state of complete physical, mental and social well-being and not just as an absence of disease. Therefore, the principle of the right to health protection must be made operational, choosing an organizational model that ensures quality and cost containment at the same time. The economic analysis consists in ensuring that the conditions of allocative efficiency, production efficiency and equity exist in the health system

In the case of health, the Region decides how many and which health structures to prepare to be able to provide the LEA necessary to meet the needs of the population, one must then ask what are the criteria for deciding which structures to actually implement, and in what proportion or how many beds read for acute compared to beds for long-term care, rehabilitation, palliative care; and then, what is the best way to provide health services. The only way to evaluate what the effect of a choice could be is to resort to economic models; in order to describe the production of health within a complex economic system, it is necessary to highlight the types of choice necessary to produce health starting from the amount of resources available to the community.

The first decision to be taken concerns the allocation of resources to be allocated to health, i.e. the best allocation is that which guarantees that maximum resources or satisfaction are achieved with the resources available.

The resources dedicated to the health production process are called inputs; once the inputs are obtained, a set of decisions must be made which consist in organizing the production of health services defined as outputs. The connection that takes place between inputs and outputs represents the process of producing health services which summarizes all the daily decisions of thousands of people with different functions and professionals who contribute to producing health services.

The related economic problem is production efficiency, i.e. producing the maximum performance by using the best available inputs. Finally, it is necessary to link the services produced or the outputs to the final result in terms of health or outcome.

In healthcare, unlike what happens in other production sectors, a distinction must be made between output, i.e. the performance and services produced, and outcome that is health, which is the final aim of the whole process. The effectiveness of health services has significant economic implications, since health workers efficiently allocating resources destined to health by producing outputs, the benefits obtained consist in the production of health.

To provide effective performance, it is necessary to rely on guidelines based on the results of the so-called EBM or Evidence-Based Medicine and on protocols. EBM takes into consideration all the scientific evidence produced and compares it with each other with particular techniques to provide scientific groups and professional associations with elements for the formation of guidelines.

The guidelines are recommendations for conduct and indicate, based on the available knowledge, the appropriate clinical behavior in the average user, leaving the management of the individual case to the operator. They refer to the complete management of the user and not only to the therapies, identifying the times and ways of involving family members or the best care settings.

The protocol is a formalized model of professional behavior for which the operators who use it are responsible. The protocol consists in the description of a succession of physical, mental, verbal actions, mostly incorporated in decision algorithms; that is, the protocol identifies the best care strategy compatible with the actual situation in which the individual operator works and allows to complete the economic evaluation with the clinical point of view, in order to ensure the best result in terms of outcome.

2. Electronic health: secure and interoperable information system

Digital healthcare is a complex reality, that is, a macro area for the large number, a macro-phenomenon that can be described according to the representative models of the markets, in which the subjects and processes involved are high, both in number and in complexity.

The World Health Organization defines the concept of e-Health as the "state of complete physical, psychic, social well-being and not just as absence of disease", on the other the digital one, intended as the use of technologies applied to diagnostic tools, to the organizational processes of health systems, to the market.
Among the figures included in the dynamics of delivery and use of health services, individuals, public and private, natural and legal persons are involved, who deal with the sale and purchase of goods and services functional to the activities related to the introduction of new technologies in the health sector.
Around the sixties, physicists, mathematicians and doctors began to deal with e-health, paying special attention to biomedical profiles. Since the end of the nineties and, with greater continuity from the beginning of the New Century, examining the action strategies promoted by the European Union and the consequent operational plans put in place by the member countries, many documents have been adopted at Community level which have concerning e-Health.
The rationalization of public health expenditure, organization of highly complex structures and systems, collaboration between healthcare professionals through telemedicine tools, access to information on assistance and care by users, aggregation and immediate availability of personal and clinical data in digital health records, are just some of the many profiles examined by the European Commission, which also pays considerable attention to the growing and current phenomena of international mobility, multiculturalism that increasingly characterize contemporary society and whose implications, in the matter of health care, are of significant importance.

The health system, as a public service, cannot be exempt from a process of structural renewal, in this sense it becomes essential to rethink traditional organizational forms and define a patient-based institutional system, in which the dialogue between technology and user push towards a simplification process of the bureaucratic apparatus. The results of this approach include the acquisition of a good level of empowerment by the user, which, in fact, benefits individuals, health systems and civil society.

New technologies play a key role in that they facilitate access to information and facilitate collaboration between organizations and professionals. Especially in a historical period, such as the current one, characterized by strong international mobility, appropriateness and optimization of treatment processes, the use of ICT is central to the "European" socio-health system. In particular, thanks to the electronic health records, the user's medical history is obtained in an updated and complete manner, accompanied by prognosis, diagnosis, therapeutic indications, diagnostic images.
In order for this hypothesis to become a reality, however, it is essential to implement the interoperability of the systems which, to date, are still manifested as a critical node for health information systems.
The digitization of the data contained in the medical records produces the positive effects both for the individual user and for the Public Administration as this innovation produces results in terms of efficiency and effectiveness. Significant are the actions undertaken by national governments aimed at reducing the complexity of the bureaucratic apparatus, to improve the timing and quality of the provision of services offered to users. Equally significant are the public interventions adopted to guarantee the security of health information systems, thanks to the which, users and operators are encouraged to use communication networks and to exchange health data.
The European Commission has already spoken several times to encourage Member States to strengthen national policies and legislation, in order to promote a homogeneous development of infrastructures which could witness a significant growth in demand for goods and services available on the Net.

The hope for the entire Action Plan designed by the European Commission is of strong synergy between the member countries. In order for the European digital healthcare market to be able to record positive trends, according to the Commission, it is necessary to allocate further funding for research in e-healthcare, to guarantee greater legal certainty by regulating the sectors of healthcare through the network, labor law on accidents and occupational diseases, develop information networks based on fixed and wireless, broadband and mobile infrastructures, and on GRID technologies to improve the interoperability of electronic medical records. Equally essential is, to increase the sensitivity and culture of users towards the issue of e-Health, through concrete health education interventions. To this end, the provision of accessible information systems, such as telematic websites on public health, as well as collaboration between Member States for the dissemination and sharing of good practices are decisive.
In compliance with the principle of subsidiarity, each government has the task and duty of guaranteeing the right of access of its citizens to health, providing efficient and effective means, of which an adequate regulatory framework shared between the member states is an essential part. Benefits of interoperability, and therefore of the flow of sensitive user data within the Union, are, first of all, the rapid accessibility of the user's clinical and health information and the consequent increase in quality and safety in care.

3. The Electronic Health Action Plan 2012-2020

The President of the Task Force made up of high-level experts in the field of electronic health, said that the digitization of health and healthcare is still significantly delayed due to regulatory tasks; these obstacles must be overcome in order to fully exploit the benefits of a robust and interoperable eHealth system in Europe. The bureaucratic-administrative systems of the Governments are burdensome as a consequence the fragmentary nature of the law prevents, better dissemination and usability of information and health services.
To comply with the revitalization of the digitization of healthcare, already called for by the European Digital Agenda and the Europe 2020 strategic plan, and to respond to new challenges in the e-healthcare sector, at the end of 2012 the European Commission defined a specific action plan, Electronic Healthcare 2012-2020. Healthcare whose operational objectives can be summarized with greater interoperability of e-health systems, research, development and innovation in digital health in order to improve efficiency and effectiveness of the services offered to consumers. What emerges from the Action Plan is the solicitation for a strong interaction between all subjects involved in any way involved in health dynamics. The term eHealth indicates the use of ICT in health products, services and processes aimed at improving citizens' health, efficiency and productivity in the health sector in order to attribute greater economic and social value to health. The benefits of e-Health for citizens, patients, health workers are remarkable in that it makes health care centered on citizens and aims to reduce cases of errors in the health sector.
Therapies performed with IT aids, which can effectively integrate ordinary clinical care, improve the cost efficiency of the treatments. The use of the electronic medical record favors the free movement of European Union citizens within the Union.
The strategy outlined in the Action Plan is based on the use and development of electronic healthcare to face the most urgent healthcare challenges for:
- improve the management of chronic diseases and improve effective practices aimed at the prevention and promotion of health;
- make health systems more sustainable and efficient by encouraging innovation;
- improve health security, solidarity and equity;
- improve legal and market conditions for developing eHealth products and services.
The action plan aims to achieve greater interoperability of e-health services, to support research, development and innovation in e-health and well-being in order to counter the scarce availability of tools and services oriented to customer.

4. The Regional Health Plan

Over the past few years, the Regions have assumed an increasingly direct and responsible role in health matters; in addition to carrying out planning and regulatory activities, they also have a direct involvement in the management and financing of health services. The purposes that each Region must achieve in health matters are included in the Regional Health Plans, through which the strategic aspects of the interventions to be put in place for the protection of health, as well as for the better functioning of the services, are identified.
The regional health plan must be prepared, taking into account the objectives identified by the national health plan; in order to ensure coordination, the Regions send the draft plans to the Minister of Health, in order to obtain their opinion.
Within one hundred and fifty days from the entry into force of the National Health Plan, the Regions must adopt their own Regional Health Plan, the preparation of which is the responsibility of the Regional Council.
An important participatory role is played by local autonomies, by private non-profit social formations, engaged in social and health care, by trade unions of public and private health workers and, finally, by structures accredited by the NHS. In the event that the Health Plan is not adopted by the Region, the provisions contained in the National Health Plan apply. The Region which has remained in default within one year of the entry into force of the PSN must take action within a term established by the Minister of Health, not less than three months. After this deadline, the Council of Ministers, on the proposal of the same Minister of Health, after consulting the Agency for Regional Health Services, in agreement with the Permanent State-Regions Conference, adopts the necessary measures to implement the PSN in the Region.
The Legislative Decree, 229/1999, amending the Legislative Decree 502/1992, has confirmed, in general, to the Region all its powers, providing that their exercise in the planning function takes place in a more immediate relationship with the Municipalities . To this end, Legislative Decree 229/1999 provided for the establishment, with regional law, of a permanent Conference for regional health and socio-health planning, of this assembly ensures the connection or inclusion in the representative body of local self-government, where established, between Regions and Municipalities, a relationship equivalent to that already operating at national level between State and Regions. For each local healthcare company, some representatives of the local self-government, that is the Mayor or the president of the Conference of Mayors depending on the size of the company itself, are part of this Conference; it also includes representatives of regional associations of local self-government. The Conference examines the draft Regional Health Plan, verifies the implementation plans of hospital companies and makes any comments on the matter.
The Regional Health Plan, the content of which must comply with the indications of the National Health Plan, has the purpose of defining the objectives to be achieved over a three-year period for the regional area of action; as well as the consequent lines of action to be pursued. In the Regional Health Plan, if on the one hand the training path of strategic decisions is extremely rigid, on the other the contents and implementation methods, entrusted to regional laws, are very flexible. The Regional Health Plan must take into account several factors, such as the analysis of the financial needs, the monitoring of essential levels of assistance, the performance and priority interventions, the monitoring of health expenditure.
The report on the health status of the country aims to present the results achieved with respect to the objectives set by the national health plan. In addition to this primary task, the report illustrates the health conditions of the population present in the area, exposes on the resources used and the activities carried out by the NHS, provides indications for the development of health policies and the planning of interventions. The function of the report is, on the one hand, to verify the application of the objectives set by the PSN on the national territory, on the other, to illustrate the results achieved by the individual Regions, on the respective regional territories, in relation to the provisions of the health regionalplans.

5.The quality of health services

Legislative Decree 502/1992 in Article 10 emphasizes the need to guarantee the quality of services by adopting the "verification and review of the quality and quantity of services, as well as their cost" method. The qualitative and quantitative indices must condition the organizational models of the healthcare companies as well as the information system of the same and the relationships between public and private subjects as well as the legislation regarding employment contracts.
The competences on the matter are shared between the Regions and the Minister of Health. The Regions are responsible for verifying compliance with the provisions relating to minimum requirements and for classifying the supply structures in order to determine the relevant activities regarding the assessment of the quality of health services. The Minister of Health is responsible for exercising the supervisory powers, for establishing by decree issued in agreement with the State-Regions Conference and having heard the competent Orders, contents and methods of use of the efficiency and quality indicators and reporting, during the course of the report on the health status of the country, regarding the checks of the results achieved.
The quality of health services is analyzed using efficient management methodologies.
Risk management is a methodology used in the health sector but changed from the financial one. Risk management is a structured methodological approach, since it is aimed at assessing risks that are already identified or identifiable as such. Without knowledge of the risk, there is no possibility to prepare or take corrective actions for improvement.
Healthcare involves the technological and at the same time the professional or human dimension; from this characteristic derives its assimilation to highly risky sectors that have already implemented strategies aimed at optimizing and increasing the quality of the services provided within the context of a wider and more innovative vision of risk management.
In the healthcare sector, we speak more precisely of clinical risk management, i.e. a corporate function that is central to the clinical governance model and whose role is to identify and prevent risks, that is, manage risk with undoubted positive effects on perceived and expected quality of the services provided.
The quality model of the health system is aimed at obtaining an efficient and effective level of quality of health care, understood as access to services, the relevance of collective needs, the practical effectiveness of individual users, equity and impartiality in the treatment, the social acceptability in the provision of the service and the efficiency and economy of the service rendered.
In any case, regardless of the model used, the priorities linked to the respective principles depend on the needs and expectations of each interested party. In the context of health care, the interesting parts include the beneficiaries of a service or treatment, the professionals and health personnel who provide the service, the managers and those who pay for the service.
The improvement of the quality of health services regards the management of total quality, understood as continuous improvement of quality, with reference to a management process aimed at establishing organizational activities of continuous improvement that involve all the components of an organization, in an integrated and overall aimed at improving overall performance. The cardinal principles that underpin total quality management include that the success of the organization lies in the adherence of all the components of the need of the users who benefit from the service; quality is a consequence of the production processes within which the causal interactions are complex.
Total quality management focuses on work processes, the implementation of total quality management implies focusing on operational processes, the explicit identification and measurement of user needs; the existence of multifunctional working groups, necessary to identify and solve quality problems; learning and continuous improvement, through research activities in order to increase the quality of the health services offered and best meet the numerous needs required by users.

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Health Pedagogy and Narrative-based Strategies to Promote Healthy Eating Behaviours and Prevent Obesity in Schoolchildren: an Experimental Protocol Designed in Salento

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Abstract
Overweight and obesity in paediatric population are becoming serious public health problems, reaching epidemic proportions in the world and being recognized as the "new pandemic of the twenty first century". Food choices and nutritional habits are strictly linked to the cultural dimension of communities and symbolic representations of food. Even in Italy, young generations are moving away from the culture of Mediterranean Diet (MD) – recognized as Intangible Heritage of Humanity by UNESCO – being at risk of losing both their identity and the health benefits associated with MD. In this frame, health pedagogy (i.e. nutritional education) can play a fundamental role in promoting the adoption of healthy eating behaviours since childhood, addressing at the same time such crucial binomials as food-health and environment-sustainability. In the last decades, a huge variety of school-based obesity prevention programs have been introduced. Among those, according to scientific evidence, narrative-based strategies may be helpful in promoting healthy nutritional habits among schoolchildren. Actually, narration is a useful tool in didactic practice, impacting on the emotional and motivational dimension of learning, thus represent-ing a valuable vehicle of health contents. On these bases, as DREAM Laboratory of Health Pedagogy, we have proposed to the Department of Prevention of the Local Health Authority ASL Lecce – specifically involving the Food Hygiene and Nutrition Service (SIAN) – to implement a research protocol aimed at assessing if narrative-based strategies could be more effective than other educational approaches in promoting the adoption of healthy eating behaviours among schoolchildren.

Keywords: health pedagogy; food habits; schoolchildren; narrative pedagogy; storytelling.

1. Introduction: background

The culture of food, together with language or dialect, is a key element of community identity, being one of the intangible assets passed from a generation to another within an uninterrupted transmission of knowledge (Napier et al. 2014; Hedegaard, 2016; Anna 2001). The Expo 2015 held in Milan has represented an extraordinary non-formal setting for generating awareness about food and health topics. It has been a great international event during which countries, companies and NGOs implemented the general theme ("Feeding the Planet") according to the different traditions, cultures and creativity of each national, regional or local community, in the perspective of defining a universal right to nutrition as "source of health" for all people and ecosystems. Health protection is a fundamental individual right (art. 32) stated in the Italian Constitution, while the right to have access to adequate food (both in terms of quantity and quality) is recognized in the Universal Declaration of Human Rights, as well as in the Convention on the Rights of the Child and Adolescent adopted by the United Nations.

Preserving specific food knowledge and flavors of different communities means protecting an integral part of their intangible heritage, which - in the case of the Mediterranean Diet (MD) -

also assumes therapeutic and preventive values, as confirmed by scientific evidence regarding the positive effects of MD on cardiovascular, metabolic and osteo-articular diseases.
Following dietary regimens with a high antioxidant content, such as those of MD, has been associated with a reduced prevalence of cardiovascular diseases and cancer in several studies. Beneficial effects of monounsaturated fatty acids (MUFA), widely contained in MD, on blood pressure and on the coagulation system have already been documented; it is now recognized that the adherence to MD is associated with the finding of a reduced waist circumference both in men and in women (Martinez-Gonzalez et al., 2009). The use of olive oil, typical of MD, reduces LDL-cholesterol levels and at the same time lowers cellular oxidative stress, thus removing free radicals from the bloodstream. Olive oil has also been shown to reduce the inflammatory response at the level of atherosclerotic lesions by inhibiting endothelial activation in the early stages of the atherosclerotic plaque genesis, displaying antithrombotic mechanisms and vascular protection (Capurso et al., 2014). Young generations who move away from the culture of the MD - recognized as Intangible Heritage of Humanity in 2010 by UNESCO – are at risk of losing both their identity and the benefits that derive from MD for individual health (Naska & Trichopoulou, 2014).
The philosopher Feuerbach asserted that "we are what we eat" and that individuals put into food all their humanity. This means that psychological variables have also an important role in determining healthy or pathological attitudes (anorexia or bulimia, diabetes, metabolic syndromes). That's why nutrition education can play a decisive role (Delgado-Noguera, 2011) since early stages of life. In this frame, health pedagogy can be helpful in promoting among young generations healthy food habits (i.e. adherence to MD), which may also impact on sustainable development. Moreover, MD has a huge range of specific local food products and biodiversity that young generations should preserve and pay attention to. Finally, food represents an opportunity for a significant relationship between parents and children, grandchildren and grandparents: by cooking and eating, community assets are transmitted and the symbolic languages of food are learned (Curtis et al., 2011).

2. *Rationale of the proposed research*

Overweight and obesity in paediatric population are becoming serious public health problems, reaching epidemic proportions in the world and being recognized as the "new pandemic of the twenty first century" (Malecka-Tendera & Mazur, 2005). Obesity has negative physical and psychological health consequences during childhood and adolescence (Ng. et al., 2014). Moreover, children's obesity is a strong predictor of adult obesity, which results in negative health and economic effects both at individual and social level. For this reason, the World Health Organization (WHO) suggests that health and "nutrition literacy" should be included in the core curriculum as children enter school, and should be supported by a "health-promoting school environment" (WHO School Policy Framework, 2008; Paakkari & Paakkari 2012; Nutbeam 2000). Different approaches have been displayed for the prevention of overweight and obesity in school setting (Birch & Ventura, 2009); Blom-Hoffman et al., 2004; Brown & Summerbell, 2009).
In a previous systematic review, we have assessed the efficacy of narrative-based strategies to promote healthy eating behaviors among schoolchildren (Pulimeno et al., 2018). On these bases, the DREAM Laboratory of Health Pedagogy has proposed to the Department of Prevention of the Local Health Authority ASL Lecce – involving specifically the Food Hygiene and Nutrition Service (SIAN) – to implement a research protocol aimed at assessing if an educational intervention in school setting based on narrative pedagogy could be more effective than other educational and preventive approaches in promoting the adoption of healthy eating behaviors among schoolchildren, and particularly the adherence to the Mediterranean diet. The general objective of the research is to plan, implement and evaluate a specific education intervention to foster healthy nutrition in a sample of primary school children through narration (storytelling activities), in the framework of health and well-being pedagogy. Our specific objectives consist in:

- Developing a multidimensional questionnaire to evaluate (during the experimental intervention) the degree of adherence to the Mediterranean diet, and the preservation of traditional food knowledge, as well as to assess the symbolic representations and the cultural dimensions underlying food choices;
- Assessing the efficacy of Narrative Pedagogy as educational intervention to promote healthy nutrition and positively impact on food choices of schoolchildren and their weight. The interventions will be based on original and engaging stories that foster the adoption of healthy lifestyles (linked to eco-sustainable models).

2. *Narrative Pedagogy for health education*

Narration has always been at the basis of education (Demetrio, 2012). The idea of learning from stories (i.e. miths, fables, fairytales etc.) can be regarded as an alternative educational strategy, useful to overcome the risk of frontal teaching and able to create a friendly classroom environment (Walsh 2011).
As highlighted by Bruner (1990; 2010), narrative thinking allows us to map the reality and look at it from multiple points of view. Thanks to narrative, we can build and re-shape our yesterday and our tomorrow. Each narrative experience - with the help of memory and imagination - mobilizes energies and nurtures a relational potential between the narrator and the listener (Diekelmann, 2009).
Being a medium of relationship in didactic practice, narration may impact on the emotional and motivational dimension of learning, becoming a valuable vehicle for health contents, including the promotion of healthy food habits. Moreover, narrative pedagogy is able to present even complex contents from the perspective of pleasure, stimulating the ethical and aesthetic sense of knowledge in the vision of a non-directive but promotional pedagogy, based on facilitation and joy. The mechanisms of interpretation, projection and reflection of narrative allow the internalization of contents (including those related to health) and potentially stimulate behavioural changes or modify cultural and symbolic representations (De Graaf, 2016).
Narrative pedagogy can engage children in activities that develop "critical and problem solving thinking" concerning risk factors for individual and collective health.
In the current scenario, where the traditional food knowledge of the communities has been replaced with global market products (i.e. artificial milk in substitution of breastfeeding and fast-food or "junk-food" instead of vegetables and fruit snacks), narrative pedagogy can play a useful role in nutritional education of young generations, becoming a successful vehicle of health contents. It is not a coincidence that publishers (as well as media) have enriched their publications with titles dedicated to food choices and cooking even for children.
In this context, the storybook is proposed as a "friendly" intermediary that satisfies children's desire for adventure and fun. The pleasure of knowledge has an emotional value and increases the motivation to learn. It is known that an optimal level of learning is achieved (along with a possible change in students' behaviours), only if children feel involved. That is why active methodologies are more effective than passive learning techniques (Michel, 2009).
Teachers could incorporate narrative-based strategies in the repertoire of teaching skills as useful instrument for sharing knowledge, values, and behaviours in a transversal and interdisciplinary way, in order to satisfy the real needs of children, who naturally learn through fun, play and communication (Piaget, 1964).
Teaching narratively consists in using stories instead of abstract concepts or theories, building up a safe and non-competitive learning environment, that stimulates creativity and encourages collaboration among schoolchildren, who feel more free to express themselves (Ironside, 2003).
School-based narrative interventions can take also advantage from technological devices and web resources (digital storytelling) – that may positively impact on the classroom daily setting (Oomen-Early, 2015; Wyatt, 2008).

3. *Methodology of the research*

A specific research protocol has been written and approved by the Ethical Committee of the Local Health Authority ASL Lecce. The methodology involves the use of a mixed approach based on quantitative and qualitative techniques. An experimental intervention of Health Pedagogy based on narrative strategies

(with the use of original storybooks specifically produced for this purpose by our PhD candidate) has been designed to prevent obesity and promote healthy eating behaviors among primary schoolchildren. Evaluation questionnaires (pre-intervention and post-intervention) aimed at assessing the cultural dimension (including aspects concerning sustainability) and the symbolic representation of nutritional choices have been developed.

Food frequency questionnaires were used to report dietary habits concerning a standard week and to evaluate the consumption of the so-called junk foods along with the degree of preservation of traditional food knowledge. Both questionnaires needed to be completed by children at home with their parents. The adherence to nutritional models that are supposed to represent a community heritage - such as Mediterranean Diet - have been assessed by KIDMED questionnaire directly administered in the classroom by personnel from Local Health Authority ASL Lecce, that had also the task of recording anthropometric measures (weight, height, Body Mass Index and waist circumference) according to anonymous and standardized procedures.

A total of 120 primary school children (aged 8-9 years old) have been enrolled in Otranto, Giurdignano, Uggiano La Chiesa and Lecce for a total of 8 classes participating into the study. Children have been divided into three different groups: (1) a group receiving traditional educational intervention with "frontal" lessons (one per month, for a total of six months) and presentation of slides addressing the topic of healthy nutrition; (2) a group receiving the multicomponent narrative-based intervention, which includes artistic laboratories (one per month, for a total of six months), aimed at stimulating the adoption of healthy eating behaviours through the original storybooks *"Tino and Rina, the fantastic adventure of life"* and *"Myrian vs. Raymond, the destiny girl and the prince of the world"*; (3) a control group in which the topic of healthy nutrition is not addressed at all. At the end of the study (May 2020), a storytelling laboratory will be offered also to children involved in the control group as a present for their participation and collaboration.

After the acquisition of informed consents from all the parents, the multidimensional questionnaire has been proposed to all children (closed answers, with compilation assisted by the parents at home) to evaluate the psychological and cultural variables related to food choices, as well as the degree of preservation of traditional food-related knowledge, but also the sustainability of supplying food (if produced from one's own garden, at 0 km, organic/bio food or purchased in full in supermarkets). A food frequency diary was also proposed to all children to assess eating habits at baseline along with the KIDMED questionnaire, specifically developed for the evaluation of the adherence to the Mediterranean diet (MD).

For all children, we have recorded at the beginning of the study the following data: weight, height, Body Mass Index (BMI) and waist circumference (CV). These latter quantitative measures will be repeated at the end of the study (May 2020) along with re-administration of all the previously described questionnaires. At the end of the study all the parents will be provided with the results of the questionnaires and the overall data of the basal and final measurements, presented anonymously.

Statistical analyses will be performed to evaluate the differences between the groups and to assess the effectiveness of the proposed narrative-based strategies in improving eating behaviors and its potential to impact on cultural representations and sustainable models (variables measured through the multidimensional questionnaire) as well as on quantitative outcomes (weight/waist circumference/BMI).

The organization of the data matrix will be performed on Excel sheet showing the individual cases in the rows and the individual variables in the columns (e.g. sex, age, weight, height, waist circumference, BMI, class). Monovariate analyses will be used to determine the trend of each variable in the population under examination for each group (e.g. verify the prevalence of overweight with BMI> 26 or the adherence to the Mediterranean diet), in order to verify the baseline characteristics of the children and apply corrective factors in the bivariate/multivariate analysis, as well as to exclude extreme inhomogeneity of the groups. Graphical representations of the frequency distributions of the variables and their amplitude will be produced. By using a double entry

table, two categorical variables ordered and un-ordered will be placed in relationships to compare the observed and expected frequencies. The analyses will mainly consist in the calculation of the before-after difference. At the end of the study, statistical analyses (χ^2; Wilcoxon test) will be performed to evaluate the differences between the groups and to assess the effectiveness of the educational intervention based on Narrative Pedagogy in improving food choices and habits (and in particular the adherence to Mediterranean diet) and consequently the quantitative outcomes (weight / waist circumference / BMI). The significance of the difference test and the Wilcoxon test will indicate the existence of a relationship /association if the p values will be < 0.05. The control of the hypotheses will be checked with bivariate or multivariate analysis.

3. *Expected Impact*

The impact of the research relies on the possibility of validating a multidimensional questionnaire concerning cultural and psychological determinants of food choices and to assess the efficacy of a multi-component narrative-based intervention in school setting (in the framework of health pedagogy) at improving eating habits of schoolchildren.

In case of significant results, the Local Health Authorities could take advantage from the use of narratives in their ordinary health educational activities in school setting.

The promotion of healthy dietary habits and working for the prevention of childhood obesity also impacts on the protection of the environment (Micha 2018; Jones, 2012), fostering the recovery of traditional and more sustainable models based on waste reduction, re-use, recycling, and responsible consumption (circular economy) that can limit the exploitation of the territory, and safeguard natural resources of the planet (such as soil and water). Finally, it must be underlined the potential impact on school learning environment (that could become more friendly and cooperative, thanks to storytelling activities). Moreover, we expect that children will get closer to storybooks, thus improving their reading's attitude and language and communication skills (Kuciapiński, 2014).

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