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un mondo libero dalla SM

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INTRODUCTION: Italy is classified as a high-risk area for MS, with highest rates in the island of Sardinia, and no evidence of the latitude gradient. The Italian MS patient society (AISM) estimates that in Italy there are 75,000 cases with an incidence of 2,000 cases per year. The latest rates published in Italy show a prevalence of 140 cases per 100,000 in 2009 [1] with the exception of Sardinia, where prevalence raised up to about 224 cases per 100,000 in 2009 [2]. The incidence was of about 5.5 cases per 100,000 in Continental Italy in 2009 [1] and 9.7 cases per 100,000 in Sardinia in 2011 [3]. In Tuscany (Central Italy), the latest estimates show a prevalence of 56 per 100,000 (1991) [4] and a mortality of 0.4 for males and 0.6 for females (2002-2006) [5]. Nowadays, in Italy, prevalence is absolutely higher than the above estimates. Indeed, prevalence is rising due to annual incidence that is higher than annual mortality [6]. In Tuscany a population MS register has been founded but, to date, it's not yet completed. To monitor disease epidemiology, comorbidities and care pathways, but also to describe the disease burden and to plan its prevention, treatment and management strategies and resource allocation, population-based studies are preferable. Administrative data offer a unique opportunity for population-based prevalence study of chronic diseases such as MS.

GOALS: We want to update the prevalence of MS in Tuscany using a validated case-finding algorithm based on administrative data and to demonstrate the progressive increment of prevalence in three consecutive years (2011-2012-2013).

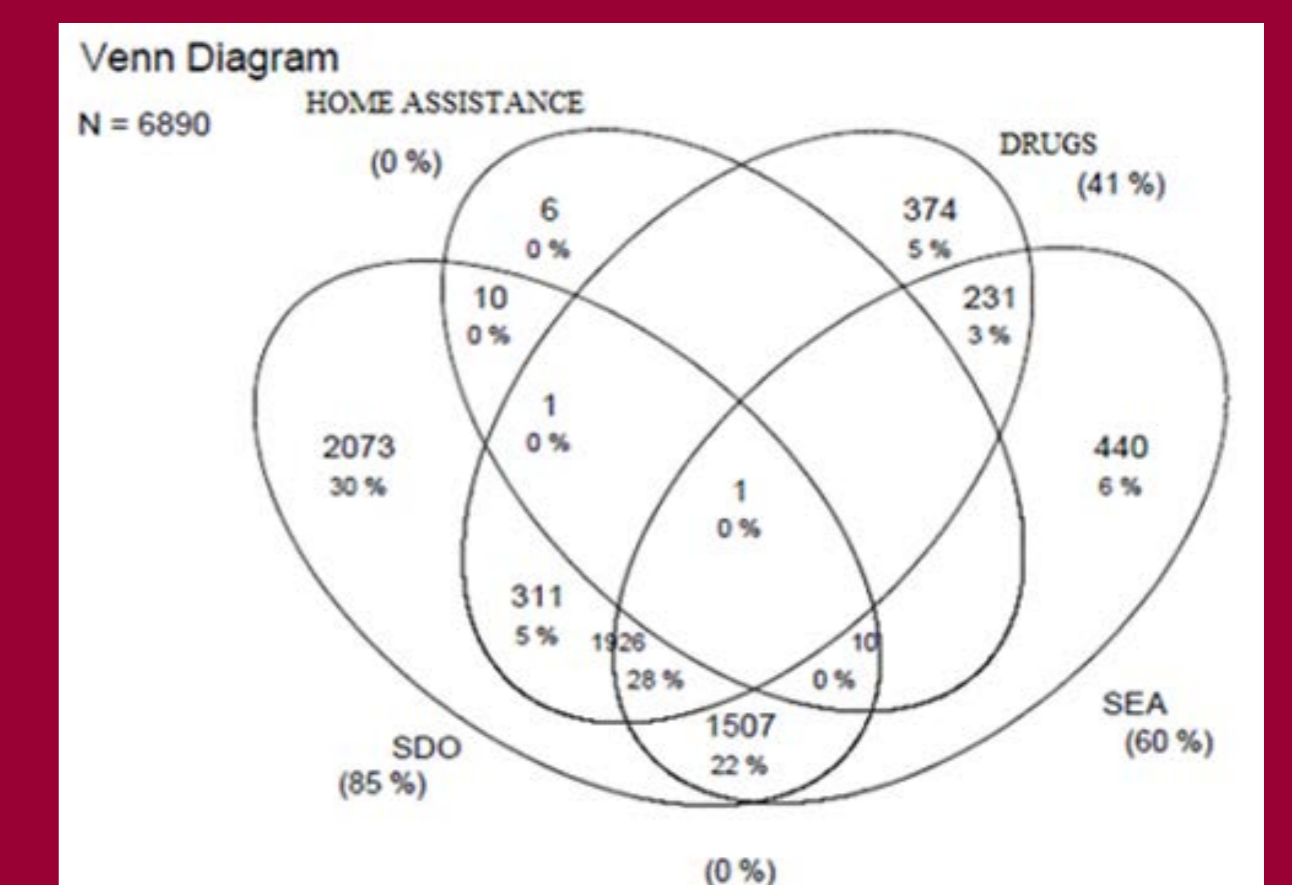
METHODS Case ascertainment and data sources: We created a case-finding algorithm in which we linked hospital discharge records, drug dispensation records, disease-specific payment exemptions and long-term care up to the end of the years 2011/2012/2013. The cohort was composed by all inhabitants alive at the prevalence day who met at least one of the following criteria: A) at least one hospital discharge record (SDO) with MS diagnosis, B) one active payment exemption (SEA) for MS, C) at least two drug prescriptions with different dates for at least one of the five drugs that are specific for MS (glatiramer acetate, interferon beta 1A, interferon beta 1 B, fingolimod, natalizumab), D) MS diagnosis in home and residential long-term care data. Then, we calculated crude prevalence at the index dates (31 December of each considered year).

Gold standard cohort: To test sensitivity, we used a true-positive reference cohort of individuals with MS, using data extracted by the Tuscan MS register. This cohort of patients was formed by all individuals with MS alive as at 12/31/2011 that had been inserted in the registry by specialists of two MS centers, the one in Livorno and the one in Siena. In total, the cohort was formed by 302 individuals with a definite MS diagnosis. To test specificity, we used another cohort of individuals who were presumably not affected by MS. For this purpose, we created a presumably true-negative reference cohort composed by individuals who were resident in Tuscany and alive as at 12/31/2011, had never undergone either cranial or spinal cord CT scan or MRI and had never received a neurological outpatient visit within the NHS. We obtained a cohort of 2,644,094 subjects presumably not affected by MS.

RESULTS: As at prevalence date, we identified 6,890 cases in 2011, 7,057 in 2012 and 7,330 in 2013 with a crude prevalence rate of 187.9, 191.1 and 195.4/100,000, respectively. Most identified individuals with MS were 16-64 years old. The female: male ratio slightly increased from 2.0 in 2011 to 2.1 in 2012-2013.

For the first year (2011) we analyzed in details the sources of our data. A total of 5,839 cases (85%) had at least one MS-related hospitalization, 4,115 cases (60%) had an MS specific payment exemption, 2,844 cases (41%) had at least two prescriptions for an MS specific drug, and only 28 cases were in long-term care for MS (Figure 1).

Figure 1. Venn diagram of sources of data.



When sensitivity was tested, we linked the true-positive reference cohort with the one obtained through our algorithm in 2011. We calculated that 296 individuals of the true-positive reference cohort were included in our cohort, and only 6 individuals with a certain MS diagnosis were not included. So, the sensitivity was of 98%. Analyzing the 6 false negative cases, we found that actually one had a drug exemption being indeed a missing datum, whereas the others could not be captured because they did neither have drug exemption nor a recent hospitalization and they were not using specific drugs. Regarding specificity, we linked the true-negative reference cohort with our cohort of 2011 and we obtained a specificity of 99,99% with only 353 patients false positive.

DISCUSSION: Our results are higher than the latest available prevalence rate of 56 cases per 100,000 reported in 1991 [4]. One of the possible reasons for this difference is the different methodology used for capturing patients. In fact, the previous study was based only on referrals from general practitioners, while the present study uses a case-finding algorithm based on various data sources, thereby allowing capturing a larger cohort of patients. However, additional factors occurred after 1991 have to be taken into consideration:

- revision of diagnostic criteria and the wide use of MRI have facilitated and anticipated the diagnosis;
- life duration is increased in general population. In addition in MS patients therapeutic approaches and management of symptoms and comorbidities ameliorated life expectancy.

The current prevalence differs from the latest available data (2009) in Italy (188, 191 and 195 vs 140 cases per 100,000) [1]: the reported prevalence increase might not be accounted solely for the two previous points. Rather, it could reflect a possible actual increase of incidence, as hypothesized by several authors [1; 6; 7; 8; 9], but not yet demonstrated.

CONCLUSIONS: With our study, we confirmed that Tuscany is a high risk area for MS and that the prevalence is increasing over time since 1991. Our algorithm based on administrative data can accurately identify patients cohorts. Our future aims are to investigate the geographical distribution of MS in Tuscany and to create an integrated dataset with administrative and clinical data to monitor not only prevalence but also disease care pathways and proxy to outcomes.

Disclosure statement: The authors declare that there are no conflicts of interest.

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