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Original article

Understanding leprosy in a nonendemic area: a pilot study on knowledge, attitudes, beliefs of medical professionals from North-Western Italy

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Summary. *Background:* Describing knowledge, attitudes and beliefs about leprosy amongst Medical Professionals in a nonendemic area (Parma Province, North-Western Italy). *Methods:* A cross-sectional study was carried among a sample of Medical Professionals (MP; No. 242) during June and July 2019 as an on-line self-administered questionnaire including 21 true/false items about epidemiology, diagnosis, and clinical characteristics of leprosy. Effectors of better knowledge status (KS) and higher risk perception (RP) were assessed through calculation of respective multivariate odds ratios (OR) and 95% confidence intervals (95%CI) in two logistic regression analysis models. *Results:* A total of 102 questionnaires were retrieved (participation rate 42.1%; 67.6% of respondents <50 year-old). Of them, 10.8% had previously interacted with at least one leprosy case. Knowledge status (KS) was unsatisfying (59.7% correct answers), and also RP was relatively low, as 91.2% of them acknowledged leprosy as a severe disease, but only 42.2% identified leprosy as highly communicable. Knowledge gaps affected particularly understanding of epidemiology and non-dermatological issues. Moreover, 30.4% of respondents ignored that a treated leprosy case may remain in the community before disease eradication. The main effector of KS was having interacted with a leprosy case (OR 4.881 95%CI 1.245-36.905), while RP was negatively associated with a better KS (OR 0.094 95%CI 0.027-0.334), and working as general practitioner (OR 0.133 95%CI 0.031-0.562). *Conclusions:* While individual expertise of European MP on leprosy slowly disappears, significant knowledge gaps and the high share of misconceptions collectively stress that refresher training may improve early diagnosis and management of incident cases. (www.actabiomedica.it)

Key words: leprosy, adult, cross-sectional studies, surveys and questionnaires, health personnel

Introduction

Leprosy, also known as Hansen's disease, is a chronic infection caused by *Mycobacterium leprae* that mainly affects skin, peripheral nerves and upper respiratory tract (1). Transmission of leprosy occurs through inhalation of bacilli from untreated multibacillary leprosy patients, but false beliefs and misunderstandings are still diffuse (2-5). Actually, leprosy is more than a biological disease: it is associated with a significant social stigma originating from socio-cultural beliefs that often lack any scientific rationale, and hindering all aspects of leprosy control. In fact, leprosy patients are likely to conceal their condition fearing social re-

jection, ultimately impairing their chances to receive an early appropriate treatment, with increased risk of permanent disabilities (1-6).

This is particularly frustrating, as leprosy is a curable disease. Since mid-1980s, global prevalence has decreased by over 90%, from 5.3 million cases in 1985 to around 192,713 cases at the end of 2017 (1, 3, 6). In 2017, around 210,671 cases were reported from 150 countries with higher detection rates occurring in South East Asia (119,055 registered cases, 7.72 new cases/100,000 population), South America (31,527 cases, 2.86 new cases/100,000 population) and Africa (30,654 cases, 1.90 new cases/100,000 population) (2, 6-9).

Once an endemic disease, the epidemiology of leprosy in Italy has changed with the nearly disappearing of locally-acquired cases. At the moment, the near totality of newly reported cases (since 2010, 0 to 12 cases/year) occur in migrants from endemic countries (10-13). Given the very low incidence and the eventual unfamiliarity of medical professionals (MP), potential knowledge gaps may impair early identification, referral, diagnosis and treatment of leprosy patients, unwillingly contributing to the social stigma usually attached to the disease (13-18). On the other hand, as an untreated multibacillary patient can release more than 10,000,000 bacilli per day, which can survive for 4-5 weeks in the Italian climate, reports following the ongoing migratory crisis have underlined the possibility of reintroduction of leprosy from endemic areas because of missed or late diagnoses (7, 19).

Our objective was therefore to assess the extent of knowledge of leprosy among a sample of MP, and their attitudes and beliefs about treatment and management of patients with leprosy.

Material and Methods

Study design and population. In this cross-sectional questionnaire-based study, MP operating in the Province of Parma, Italy, have been asked about their knowledge, attitudes and beliefs on leprosy. A convenience sample had been collected among the MP participating to a medical online forum (in total, 242 members). All participating MP had received by mail a link to an online questionnaire. The survey had been conducted by means of Google Forms®, and the purpose, the risks, and the benefits of the study had been outlined in the email containing the link.

Questionnaire. The questionnaire was made available up to the end of July 2019, encompassing a total of 26 items divided in 5 areas of inquiry:

1) *Characteristics of the participants*: age, sex, seniority, practice settings;
2) *Previous interaction with leprosy cases*. Participants had been asked whether they had or not: (a) previously interacted with a leprosy case; (b) contributed to the diagnosis of a leprosy case; (c) managed a leprosy case diagnosed by another professional; (d) previously participated of formation courses on leprosy diagnosis and management; (e) previously stayed and/or worked in Sub-Saharan Africa, Indian Subcontinent, South America.
3) *General knowledge about leprosy*. A total of 21 true-false statements covering some misconceptions on leprosy had been presented to the participants (e.g. "*vaccinations increase the occurrence of allergies*"; false). Items had been designed through an accurate analysis of similar studies (4,20-24). A general knowledge score (KS) had been then calculated as the sum of correctly and incorrectly marked recommendations: when the physicians answered correctly, +1 was added to a sum score, whereas a wrong indication or a missing/"don't know" answer added 0 to the sum score. A similar knowledge test had been previously validated and successfully applied in order to assess the degree of misconceptions held by the participants, particularly towards influenza, tetanus, and vaccination intentions (25-29);
4) *Risk perception*. Perceived risk may be defined as a function of the perceived probability of an event and its expected consequences, and therefore assessed as the mathematical product of subjective probability and disease severity (26,27,30). Therefore, we asked the participants about the perceived infectivity of leprosy ($\mathbf{L}^{\mathbf{INF}}$), and its presumptive severity ($\mathbf{L}^{\mathbf{SEV}}$)

through a 5-point likert (i.e., 1 = "very low, 5 = "very high"). The risk perception score (RPS) was eventually calculated as it follows:

$$\mathbf{RPS = L^{INF} \times L^{SEV}}$$

5) Attitudes and Beliefs. Eventually, participants received a set of three statements about management of leprosy cases, and more precisely: (a) "*Leprosy is a curable disease*"; (b) "*A patient affected by leprosy cannot stay in the community until the eradication of the pathogen is eventually achieved*"; (c) "*A patient affected by leprosy should be customarily hospitalized*". Participants were asked to rate their perception through a 5-point Likert scale (from "*totally disagree*" to "*totally agree*"), and results have been eventually dichotomized as "*somehow disagree*" and "*somehow agree*".

Data analysis. Two independent researchers ensured the accuracy of data entry: one read the responses from each questionnaire, while the other researcher reviewed the entered data. The primary investigator examined unclear responses to determine the correct answer. We calculated the described indices for KS and RPS, and all scores had been normalized to per cent values (min. 0.0, max 100) in order to compare the results more easily. A preventive reliability test was performed on GKS through determination of Cronbach's alpha. Continuous variables were expressed as mean ± Standard Deviation (SD). Categorical variables were reported as per cent values. Univariate confrontations between proportions were evaluated through the Chi^2 test (with continuity correction), whereas continuous variables were compared through Student's t-test for unpaired data, or ANOVA when appropriate (Tukey's post hoc test). Relations between the cumulative scores were explored through the calculation of the Pearson product-moment correlation coefficient (i.e., Pearson's r). In regression analyses (SPSS 25, IBM Corp. Armonk, USA), KS and RPS were dichotomized as ≤ vs. > median values in order to calculate multivariate Odds Ratios (ORs) with their respective 95% confidence intervals (95% CI) for their effectors among individual factors. Regression models included all factors that, at univariate analyses, were associated with a KS and RPS > median with a p value < 0.05. In all calculations, significance level was $p < 0.05$.

Ethical considerations. Before giving their consent to the survey, participants were notified that all information would be gathered anonymously and handled confidentially. Participation was voluntary, and the questionnaire was collected only from subjects who had expressed consent for study participation. As individual participants cannot be identified based on the presented material, this study caused no plausible harm or stigma to participating individuals. Eventually, no preliminary evaluation by the Ethical Committee was reputed necessary.

Results

Descriptive analysis. As shown in Table 1, 102 out of 242 potential participants (42.1% of the original sample) have participated to the inquiry: 48.0% were of male gender, and the majority of them was ≤ 50-year-old at the time of the survey (67.6%). The majority of them worked as general practitioner (GP, 76.5%). Overall, 10.8% of them had previously interacted with a leprosy case, participating to the diagnosis in 6.9% of cases, but none of them had actually managed a case diagnosed by other professionals. Interestingly enough, no one among GPs had reported a previous interaction with a leprosy case. Previous participation to post-degree formation courses on leprosy was reported by 6.9% of respondents, while a total of 8 MP (7.8%) had lived and/or worked in endemic areas (i.e. Sub-Saharan Africa, Indian Subcontinent, South America).

Assessment of Knowledge Status (Table 2). After normalization, the mean KS was 59.7% ± 22.4 (actual range 19.1% - 100%; median 57.1%), and internal consistency coefficient amounted to Cronbach's alpha = 0.845. Even though 93.1% of the sample correctly reported a bacterium as primary cause of leprosy, 91.2% appropriately identified hands, feet and face as main targets of the disease, and 70.6% that a patient is unable to spread pathogens after the beginning of the therapy, a significant share of participants exhibited uncertainties about leprosy's epidemiology and clinical features. Not only a third of responders was not aware that bacteria are transmitted through droplets,

Table 1. Characteristics of the Medical Professionals participating to the survey (Number = 102)

Variables	No./102, %	Mean ± S.D.
Age group (years)		
< 30	12, 11.8%	
30 - 39	21, 20.6%	
40 - 49	36, 35.3%	
50 - 59	19, 18.6%	
60 or more	14, 13.7%	
Gender		
Male	49, 48.0%	
Female	53, 52.0%	
Previously stayed / worked in South America, Asia or Africa	8, 7.8%	
Occupation as General Practitioner	78, 76.5%	
Previous interaction with a leprosy case (any)	11, 10.8%	
Previously diagnosed a leprosy case	7, 6.9%	
Previously managed a leprosy case diagnosed by another professional	0, -	
Previously participated to formation courses on leprosy diagnosis and management	12, 11.8%	
Attitude towards leprosy (agree / totally agree)		
Leprosy is a curable disease	82, 80.4%	
Leprosy is a severe disease	93, 91.2%	
Leprosy is a highly communicable disease	43, 42.2%	
A patient affected by leprosy cannot stay in the community until the eradication of the pathogen is eventually achieved	31, 30.4%	
A patient affected by leprosy should be customarily hospitalized	28, 27.5%	
Cumulative scores (%)		
Risk Perception Score		64.1% ± 25.6
> median (60.0%)	36, 35.3%	
Knowledge Score		59.7% ± 22.4
> median (57.1%)	47, 46.1%	

but around half of them reported contaminated water and handshake as able to spread contagion (50.0% and 43.1%, respectively). Actual figures of leprosy in Italy were identified by 69.6% of respondents: interestingly enough, while the large majority of participants associated new cases with people having a migration background (92.2%), only 57.8% were aware that nearly half of them are irregular migrants, and 40.2% that South America is no longer the most frequently reported geographic origin of new cases. Moreover,

Table 2. Knowledge of 102 Medical Professionals participating to the survey about leprosy, its epidemiology, diagnosis and management (Cochrane alpha = 0.845)

	Correct Answer	**No., %**
The primary cause of Leprosy is a bacterium	TRUE	95, 93.1%
Skin lesions of Leprosy are very painful	FALSE	55, 53.9%
Leprosy is actively transmitted through droplets	TRUE	68, 66.7%
Leprosy may be transmitted through handshake	FALSE	44, 43.1%
Leprosy may be transmitted through contaminated water	FALSE	51, 50.0%
Leprosy affects central nervous system	FALSE	16, 15.7%
Patients are unable to spread leprosy after the beginning of the therapy	TRUE	72, 70.6%
Kidney and liver are among the main targets of the Leprosy	FALSE	49, 48.0%
Hypopigmented skin lesions are among possible clinical signs of Leprosy	TRUE	54, 52.9%
Nodular skin lesions are among possible clinical signs of Leprosy	TRUE	57, 55.9%
Lagophthalmos is a clinical sign of Leprosy	TRUE	68, 66.7%
Flaccid paralysis of the feet is a possible early complication of Leprosy	FALSE	46, 45.1%
Claw hand is a possible late complication of Leprosy	TRUE	58, 56.9%
Numbness of hands and/or feet are diffuse complications of Leprosy	TRUE	74, 72.5%
Hands, feet and face are among main targets of Leprosy	TRUE	93, 91.2%
Nowadays, less than 20 cases of leprosy are diagnosed yearly in Italy	TRUE	71, 69.6%
Most of leprosy cases diagnosed in Italy occur among foreign-born people	TRUE	94, 92.2%
Nearly half of new leprosy cases occur among irregular migrants	TRUE	59, 57.8%
In Italy, diagnostic delay may be greater than 2 years	TRUE	73, 71.6%
In Italy, the majority of new cases occur in migrants from South America	FALSE	41, 40.2%
Italian National Center of Genoa San Martino nowadays is not operative	FALSE	41, 40.2%

while 71.6% of the sample was aware that diagnosis delay may be very long even greater than 2 years, ongoing activity of the national referral center of San Martino - Genoa was reported by only 40.2%.

Even greater uncertainties were reported about signs and symptoms of leprosy, as only 53.9% of participants were aware that skin lesions are not painful, and a similar proportion correctly reported hypopigmented (52.9%) and nodular skin lesions (55.9%) as possible signs of leprosy. Moreover, around half of respondents reported kidney/liver involvement as a possible leprosy complication. Even though involvement of peripheral nervous system was appropriately reported by 56.9% (hand claw sign) to 66.7% (lagophthalmos), and even 72.5% (numbness of the hand/feet), only 15.7% of respondents were aware that leprosy usually does not affect the central nervous system, and more than 50% misreported flaccid paralysis as a sign of leprosy.

Assessment of attitudes and beliefs. In total, 80.4% of participants agreed or totally agreed in recognizing leprosy a curable disorder, while 91.2% rec-

ognized leprosy as a severe disease, and only 42.2% characterized Hansen's disease as highly diffusive. Focusing on the management of new diagnoses, 30.4% agreed/totally agreed that a patient affected by leprosy cannot stay in the community until the eradication of the pathogen is eventually achieved, and 27.5% that a leprosy patient should be customarily hospitalized following first diagnosis. In summary, a RPS score of 64.1% ± 25.6 was identified (range: 20.0 - 100%), with a median value of 60.0%.

Univariate analysis. KS and RPS were not significantly correlated ($r = -0.106$, $p = 0.289$). As shown in Table 3, a better KS was positively associated with male sex ($p = 0.006$), previous interaction with a leprosy case ($p = 0.028$), a better RPS ($p < 0.001$) and negatively associated with the statements that a patient cannot remain in the community until the pathogen is fully eradicated ($p = 0.001$) and that a leprosy case should be customarily hospitalized. Similarly, higher RPS was negatively associated with the male sex ($p = 0.016$), occupation as GP ($p < 0.001$), recognizing leprosy as a curable disease ($p = 0.017$), and better KS ($p < 0.001$).

Regression analysis (Table 4). Regression analysis for KS included gender, previous interaction with a leprosy case, acknowledging leprosy should be always hospitalized, and that a patient affected by leprosy cannot stay in the community until the eradication of the pathogen is eventually achieved. Regression analy-

Table 3. Factors associated with Knowledge Score (KS) and Risk Perception Score (RPS) greater than their median values (respectively, 57.1% and 60.0%; univariate analysis)

Variable	KS		P value	RPS		P value
	> 57.1% (No./47, %)	**≤ 57.1%** (No./55, %)		**> 60.0%** (No./36, %)	**≤ 60.0%** (No./66, %)	
Age > 50 years	19, 40.4%	14, 25.5%	0.162	7, 19.4%	26, 39.4%	0.066
Male Gender	30, 63.8%	19, 34.5%	0.006	11, 30.6%	38, 57.1%	0.016
Occupation as General Practitioner	20, 42.6%	17, 30.9%	0.311	3, 8.3%	34, 51.5%	< 0.001
Previously stayed / worked in South America, Asia or Africa	6, 12.8%	2, 3.6%	0.180	1, 2.8%	6, 10.6%	0.308
Previous interaction with a leprosy case (any)	9, 19.1%	2, 3.6%	0.028	3, 8.3%	8, 12.1%	0.798
Previously diagnosed a leprosy case	5, 10.6%	2, 3.6%	0.317	3, 8.3%,	4, 6.1%	0.981
Previously participated to formation courses on leprosy diagnosis and management	8, 17.0%	4, 7.3%	0.224	6, 16.7%	6, 9.1%	0.416
Attitude towards leprosy (agree / totally agree)						
Leprosy is a curable disease	41, 87.2%	41, 74.5%	0.174	34, 94.4%	48, 72.7%	0.017
A patient affected by leprosy cannot stay in the community until the eradication of the pathogen is eventually achieved	6, 12.8%	25, 45.5%	0.001	12, 33.3%	19, 28.8%	0.801
A patient affected by leprosy should be customarily hospitalized	3, 6.4%	25, 45.5%	< 0.001	13, 36.1%	15, 22.7%	0.224

Table 4. Factors associated with Knowledge Score and Risk Perception Score > median values (i.e. 57.1% and 60.0%, respectively). Adjusted Odds Ratio (OR) and their respective 95% confidence intervals (95%CI) were calculated through regression analysis: the models included all factors that were significantly associated with higher RPS and KS at univariate analysis ($p < 0.05$)

	KS > median (57.1%)		**RPS > median (60.0%)**	
	OR	**95%CI**	**OR**	**95%CI**
Male Gender	4.188	0.961; 12.887	1.575	0.431; 5.570
Occupation as General Practitioner	-	-	0.133	0.031; 0.562
Previous interaction with a leprosy case (any)	4.881	1.245; 36.905	-	-
Attitude towards leprosy (agree / totally agree)				
Leprosy is a curable disease	-	-	6.105	0.789; 47.243
A patient affected by leprosy cannot stay in the community until the eradication of the pathogen is eventually achieved	0.167	0.019; 1.463	-	-
A patient affected by leprosy should be customarily hospitalized	0.261	0.028; 2.427	-	-

sis for RPS included gender, working as GP, and acknowledging leprosy as a curable disease. In summary, previous interaction of the respondents with a leprosy case was a significant predictor for a better knowledge score (OR 4.881; 95%CI 1.245-36.905), while working as a GP was a significant negative effector for higher RPS (OR 0.133; 95%CI 0.031-0.562).

Discussion

In European Union countries, leprosy has become a rare disease, with infrequent autochthonous transmission, and only a small number of new cases reported each year, mainly among migrants and/or refugees (1, 12, 19, 31). Not coincidentally, only 11 (10.8%) of respondents had previously interacted with a leprosy case, and while 6.9% had reportedly contributed to the clinical diagnosis of leprosy, none of them had managed a leprosy case. Such figures are significantly lower than that reported in similar studies, but it should be stressed that all available researches have been performed in endemic countries (4, 5, 21-24). However, following the recent increase in travel and migration, leprosy can be reported from any country, and early identification and treatment of new cases is a public health priority that should not be forgotten by every MP who could interact with any new leprosy case (7, 19).

Unfortunately, our survey suggests that Italian MP may be affected by significant knowledge gaps, largely underestimating the potential health threat represented by leprosy. First at all, even though Hansen's disease was diffusely acknowledged as a curable one, around a third of respondents were unaware what a treated leprosy case can stay in the community, not requiring forced hospitalization, and 42.2% of participants characterized leprosy - a not particularly infectious disease, as highly communicable. In other words, a significant share of respondents shared significant misunderstandings that ultimately maintain and propagate the social stigma affecting leprosy patients (1, 5, 23, 31, 32).

Second, because of the inappropriate understanding of actual epidemiological features it is possible that MP may actually bear a low suspicion index when facing potential cases. This is particularly worrisome, as even the understanding of clinical features was largely unsatisfying, and otherwise consistent with a recent review on leprosy in Italy, stressing that diagnostic delay may reach years and even decades (13). Even if Hansen's disease is too often mistakenly understood as a primarily skin disorder, knowledge gaps included the recalling of significant dermatological signs and symptoms (5, 21, 23, 24). Moreover, most of non-dermatological features were ignored by around half of the respondents. More specifically, a significant propor-

tion of MP was not familiar with complications such as lagophthalmos (33.3%), and claw hand (43.1%), and an even larger share of participants associated leprosy with features such as flaccid paralysis of the feet (possibly confounded with the "*foot drop*", 54.9%), disorders of kidney and liver (52.0%), and even impairment of the CNS (84.3%). Such misunderstanding of the leprosy clinical features were previously reported even from countries where leprosy is still endemic, and collectively indicate the possibility of missing or delaying the diagnosis if the patients present with relatively less common clinical manifestation (5, 21, 23). Similarly, we can speculate that the understanding of leprosy as a primarily skin disease possibly affected the knowledge gaps we identified about its transmission: nearly half of the respondents believed that leprosy may be transmitted through touch (43.1%) or even by means of contaminated water (50.0%), and such results are in line with two previous studies from Guyana (21) and Sri Lanka (5), in which aerosol transmission of *M leprae* was acknowledged by 30 to 60% of respondents, while direct contact transmission was recalled by around one fifth of study participants.

Health Belief Model (30, 33) suggests that beliefs about the perceived susceptibility to a health threat, its severity, and perceived benefits and/or barriers towards a particular protective action, may be significantly influenced by personal experiences. Not coincidentally, higher KS was significantly associated with having previously interacted with a leprosy case: available studies suggest that higher familiarity with Hansen's disease is associated with better understanding of the disorder (5, 21, 23, 24). The low RPS associated with the GP status may be similarly explained by means of the Health Belief Model. Because of restrictive regulations to access healthcare services, Italian GPs have few opportunities to interact with higher risk groups such as migrants and particularly irregular migrants, that more frequently refer to MP from specific services, including Emergency Departments (34). Unsurprisingly, no one among the sampled GPs had a previous interaction with leprosy cases, and unfamiliarity with the disorder can easily mean underestimating its actual severity (30,33).

Despite its potential public health impact, our study is affected by several limitations.

First and foremost, our survey had a limited sample size, including 102 professionals from a very delimited geographic area, and only 42.1% of the original sample eventually participated to the survey. However, as the original questionnaire was not shared outside the original recipients, it is unlikely that the final sample included professionals from nearby provinces, furtherly compromising the representativity of the sample.

Second, since the recruitment of the participants has been voluntary, it is not possible to rule out the existence of a selection bias. Participating voluntarily could be due to a proactive attitude or greater knowledge about the health issue we assessed. As the fact of not participating could be understood as a negative attitude or a lack of knowledge about vaccination, actual understanding of Hansen's disease among Italian MP may even worse that that we reported.

Third, we cannot rule out that our results may have been affected by a significant social desirability bias, with participants reporting the "*socially appropriated*" rather than their authentic behaviors, so that our result could have ultimately overstated the share of participants who actually recognized leprosy as a severe and highly communicable disease, and conversely underestimated the number of MPs who associated leprosy with refugees or irregular migrants (26, 28). Similarly, the high share of participants identifying leprosy as a highly contagious disorder, and the diffuse understanding of leprosy as a mainly dermatological disorder, suggest that the knowledge test may have been characterized by a high number of "*common-sense"* rather than "*evidence based*" answers.

Conclusions

Our explorative study suggests that MPs from Northern Italy exhibit a very high prevalence of false beliefs and misunderstanding about leprosy, particularly among GPs, whose unfamiliarity with the disease may explain the very long diagnostic delay affecting leprosy cases in Italy. Some of such knowledge gaps may unwillingly by significantly contribute to social stigma associated with the Hansen's disease, even in high income countries. Ultimately, our results stress

the urgent need of tailored training and educative programs, specifically aimed to GPs and MPs less likely to interact with high risk groups such as refugees and irregular migrants.

Disclosures. The facts, conclusions, and opinions stated in the article represent the authors' research, conclusions, and opinions and are believed to be substantiated, accurate, valid, and reliable. However, as this article includes the results of personal researches of the Authors, presenting correspondent, personal conclusions and opinions, parent employers are not forced in any way to endorse or share its content and its potential implications. Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article.

Conflict of interest: Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article

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