

A Scoring System for the Early Detection of Oral Submucous Fibrosis Based on a Self-administered Questionnaire

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Abstract

Objectives: The aims of the present study were to evaluate the frequent clinical complaints of oral submucous fibrosis (OSF) and to develop a scoring system for early detection of the disease by a self-administered questionnaire. **Methods:** A total of 296 subjects were recruited, including 123 OSF patients without oral cancer and 173 betel quid chewers without OSF or oral cancer. A self-administered questionnaire was used to collect the symptom profile from study subjects. Their maximal mouth opening (MMO) between upper and lower incisor edges was measured and recorded by well-trained nurses. A binary logistic regression model examining the likelihood of OSF based on the eight symptoms of interest was used to develop the scoring system. **Results:** Among 79 OSF subjects with an MMO <35 mm, the most frequent complaint was trismus (87.3%), followed by burning sensation (76.0%) and xerostomia (72.2%). Among 44 OSF subjects with an MMO ≥35 mm, burning sensation (68.2%) was the most frequent complaint, followed by trismus (54.5%) and xerostomia (54.5%). Six frequent complaints including trismus, burning sensation, xerostomia, sore throat, numbness, and oral ulceration were utilized to develop a scoring system for the early detection of OSF. The scoring system had an area under the receiver operating characteristic curve of 0.90. **Conclusion:** This study suggests a screening questionnaire of frequent complaints for the early detection of OSF. [J Public Health Dent 2002;62(1):28-31]

Key Words: oral submucous fibrosis, early detection.

Oral submucous fibrosis (OSF) is a betel quid chewing-specific precancerous condition (1, 2). The onset of the disease is often insidious and its clinical symptoms are variable. It may eventually lead to severe mouth opening limitation, trismus (3). Several remedies have been tried to improve the mouth opening of OSF patients with trismus, but none of them was satisfactory (4). Because of the malignant tendency and possible trismus complication, regular follow-up of OSF patients is recommended.

This disease affects approximately 0.5 percent (5 million people) of the population in the Indian subcontinent and is now a public health issue in many parts of the world, including the United Kingdom, South Africa (2), and many Southeast Asian countries. In

Taiwan, betel quid chewing was prevalent among the working classes in the past years. Recently, the betel quid chewing population has been growing. The socioeconomic level of chewers is also going up and there are more young betel quid chewers (BQCs). In Taiwan, the impact of OSF is becoming a pressing public health issue. We conducted this study to evaluate frequent clinical complaints of OSF and to develop a scoring system for the early detection of OSF by a self-administered questionnaire. The performance of the scoring system was also evaluated.

Methods

Study Subjects. The consecutive OSF patients were recruited from the Department of Dentistry of National

Taiwan University Hospital from January 27 to December 31, 1999. Every male patient was asked to fill out a one-page questionnaire before clinical examination. The questionnaire included basic demographic characteristics, eight frequent symptoms of OSF, as well as betel quid chewing history. The eight symptoms included trismus (mouth opening limitation), burning sensation while eating spicy food, xerostomia, mal-taste (defective gustatory sensation), sore throat or pain in ear region, oral numbness, oral ulceration, and dysphagia. The English version of these eight questions is shown in Table 1.

The maximal mouth opening (MMO) between upper and lower incisor edges of each study subject was measured and recorded by well-trained nurses. Every patient was examined and diagnosed by oral pathologists. The oral pathologists were blinded to the results of the questionnaire. Palpable or strip fibrous bands in a chewer involving one or more oral structures, including buccal mucosa, labial mucosa, fauces, soft palate including the uvula, floor of the mouth, and the tongue were regarded as OSF (5). Scars and oral frenulae were not considered as fibrous bands. Oral symptoms were not regarded as diagnostic for OSF in the absence of fibrous bands. A total of 123 consecutive OSF patients without oral cancer were recruited. To prevent a keloid-like response after biopsy surgery, further histopathologic confirmation was carried out on 31 (25.2 percent) patients with suspected malignant transformations. All OSF subjects were BQCs.

One hundred and seventy-three BQCs who had never been diagnosed

with OSF or oral cancer were also recruited from five townships including Makung, Chutung, Potzu, Kaoshu, and Sanchi. These townships were selected for the cooperation of local health personnel and residents. They also represented both urban and rural areas in Taiwan. The same questionnaire was used to collect the information from these community subjects.

Statistical Analysis

Development of a Scoring System for Early Detection. We used a binary logistic regression model to examine the likelihood of being affected with OSF based on the eight symptoms of

interest. To develop the model, we utilized the SAS/LOGISTIC program with the data set of 44 OSF subjects having an MMO ≥ 35 mm (5-7) and 173 community BQCs. The initial model was developed from the eight symptoms. None of the unaffected BQCs complained of maltaste or dysphagia. These two symptoms of interest, maltaste and dysphagia, were dropped from the model to avoid the problem of being unable to derive fixed estimates. The remained six symptoms were included in the final model. The intercept estimated from the model was subtracted by $\log$ (prior odds of OSF), that is, $\log (44/173)=-1.369$ (8).

All the estimated parameters in the model were then exponentiated and rounded. A composite score for each study subject was obtained by multiplying the scores according to whether or not the subject had a particular symptom. This composite score represented the model-based likelihood ratios (LRs) of being an OSF case for the particular subject with the specific combination of symptoms (8). According to this scoring system, a study subject was classified as a case or noncase if his composite score (the model-based LR) was greater or less than the cutoff point selected to achieve a desired level of sensitivity and/or specificity.

Evaluation of the Scoring System

To evaluate the screening value of the scoring system, we drew the receiver operating characteristic (ROC) curve of the logistic regression model (9). The trapezoid rule was utilized to calculate the areas under ROC curve (AUC) (9). The sensitivities and specificities of the scoring system at various composite score cutoff points were calculated. Linear interpolation was applied to estimate the sensitivity of the scoring system at a particular level of specificity if necessary. However, we used the sensitivity to indicate the probability of correct classification of OSF patients, and the specificity to indicate the probability of correct classification of unaffected BQCs. To generate the ROC curves (Figure 1) of the logistic regression model including eight symptoms, an OSF subject who had the symptoms of maltaste and/or dysphagia was classified as an OSF case when the probability level was less than 1.0.

Results

Demographic Characteristics. The age range for 123 OSF subjects and 173 unaffected BQCs were 17-66 years and 39-72 years, respectively. With regard to the ethnicity, 100 (81.3%) OSF subjects and 140 (80.9%) unaffected BQCs were Fukien Taiwanese. With regard to the education level, all of our study subjects were literate or had at least an elementary school education.

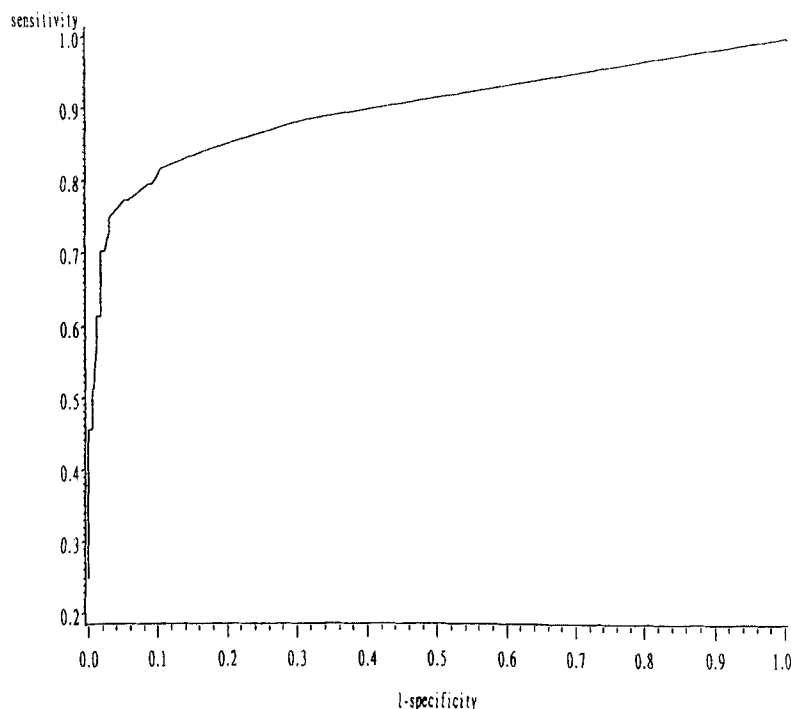
Symptom Profile of OSF Patient

The number and percentage of clinical symptoms among the study subjects are listed in Table 2. All of the eight symptoms were more prevalent among OSF cases than among unaffected BQCs.

TABLE 1
Questionnaire for Early Detection of Oral Submucous Fibrosis

1. Do you often feel stiff cheeks and difficulty in opening mouth?	Yes	No
2. Do you often feel burning sensation in mouth while eating hot or spicy food?	Yes	No
3. Do you frequently feel dryness in mouth?	Yes	No
4. Do you frequently feel change of taste?	Yes	No
5. Do you frequently feel pain in ear/throat region?	Yes	No
6. Do you frequently feel numbness in mouth?	Yes	No
7. Do you frequently have vesicles and/or ulceration in mouth?	Yes	No
8. Do you frequently feel difficulty in swallowing?	Yes	No

FIGURE 1
Receiver Operating Characteristic (ROC) Curve for Initial Model
[Area Under ROC Curve (AUC)=0.903]



affected BQCs. Among OSF patients with MMO ≥ 35 mm, burning sensation (68.2%), trismus (54.5%), xerostomia (54.5%), and ulcer (47.7%) were the most prevalent symptoms. Among OSF patients with MMO < 35 mm, trismus (87.3%) was the most prevalent symptom that differed significantly ($P < .001$) from the prevalence among OSF patients with MMO ≥ 35 mm (54.5%). The proportions of xerostomia and dysphagia were also significantly ($P < .05$) different between these two groups of OSF patients.

Screening of OSF with the Scoring System. The scoring system derived from the logistic regression model using six symptoms is shown in Table 3. For a BQC having complaints of burning sensation and oral ulceration, his composite score was $0.17 \times 4.84 \times 2.57 = 2.11$, which was also the LR of being affected with OSF. The background score (0.17) was the baseline LR of being affected with OSF. Complaint of any symptom increased the composite score. Trismus and burning sensation had more contribution on composite score than the other four symptoms. Xerostomia, sore throat, numbness, and ulceration of oral cavity had similar load on composite score.

The sensitivity and specificity of the scoring system at various cutoff points of the composite score are shown in Table 4. When the cutoff point was set as 1.0, the sensitivity and specificity of detecting early-stage OSF were 82.0 percent and 85.8 percent, respectively.

The ROC curve for the model including eight symptoms is shown in Figure 1. The AUC of the ROC curve was 0.903. The ROC curve for the model including six symptoms was similar to that of the model with eight symptoms showing an AUC of 0.895 (Figure 2).

Discussion

The eight symptoms investigated in this study have been documented to be prevalent among OSF patients (3,5). According to our data, proportions of these symptoms are significantly ($P < .001$) higher among OSF patients than among unaffected BQCs. This result implied that the eight symptoms are useful to differentiate OSF patients from unaffected BQCs. Moreover, trismus was the most important symptom for an early-stage OSF patient (MMO ≥ 35 mm) to be differentiated from ad-

TABLE 2
Distributions of Clinical Symptoms of 123 Patients Affected with Oral Submucous Fibrosis (OSF) and 173 OSF-free Betel Quid Chewers (BQC)

Symptoms	OSF (n=123)		BQC (n=173)
	MMO < 35 mm (n=79) No. (%)	MMO ≥ 35 mm (n=44) No. (%)	No. (%)
Trismus	69 (87.3)	24 (54.5)	3 (1.7)
Burning sensation	60 (75.9)	30 (68.2)	23 (13.3)
Xerostomia	57 (72.2)	24 (54.5)	24 (13.9)
Maltaste	18 (22.8)	9 (20.5)	0 (0.0)
Sore throat	24 (30.4)	11 (25.0)	3 (1.7)
Numbness	18 (22.8)	9 (20.5)	2 (1.2)
Ulcer	37 (46.8)	21 (47.7)	21 (12.2)
Dysphagia	18 (22.8)	3 (6.8)	0 (0.0)

MMO=maximal mouth opening.

TABLE 3
Scoring System for Early Detection of Oral Submucous Fibrosis Derived from Logistic Regression Model Using 6 Symptoms

Terms	Score
Background	0.17
Clinical symptoms	
Trismus	19.89
Burning sensation	4.84
Xerostomia	2.91
Sore throat	2.72
Numbness	2.08
Ulcer	2.57

Composite score = $0.17 \prod (\text{Score}_i)$, $i=1$ to K positive symptoms.

vanced OSF patients (MMO < 35 mm). As shown in Table 2, a substantial difference in symptom profiles between early-stage OSF and advanced OSF was noted. To increase the sensitivity for detecting early-stage OSF cases, we excluded 79 severe OSF cases and utilized 44 early-stage OSF cases to develop the scoring system for early detection of OSF.

Excepting the precancerous characteristic, limitation of mouth opening is the most important clinical problem of OSF. Trismus impairs the ability to eat, speak, and even to receive dental treatment. About one-half of our early-stage OSF patients complained of trismus. It seemed that the progression of mouth opening limitation was not so

TABLE 4
Sensitivity and Specificity of Scoring System for Early Detection of Oral Submucous Fibrosis at Various Cutoff Points of Composite Score

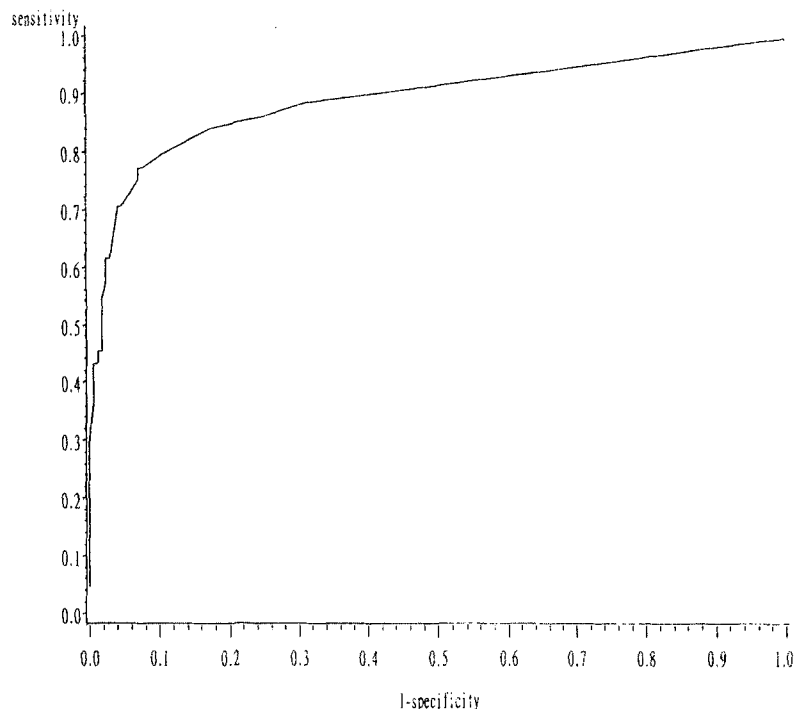
Cutoff Point of Composite Score	Sensitivity (%)	Specificity (%)
0.2	98.5	9.1
0.5	86.3	75.5
0.6	85.6	78.0
1.0	82.0	85.8
1.8	77.3	92.9
2.0	75.2	93.1
5.0	58.2	97.7

insidious among these early-stage OSF patients. In our scoring system for early detection of OSF, trismus had the greatest contribution to the composite score.

The similarity of ROC curves and AUCs between the model including eight symptoms (Figure 1) and the model including six symptoms (Figure 2) suggests little information was lost by dropping maltaste and dysphagia.

While utilizing 123 OSF subjects in the development of the scoring system for identifying OSF cases in general (without regard to MMO), the AUC of the ROC curve for the model including six symptoms was 0.942. The ROC curve for the model including eight

FIGURE 2
Receiver Operating Characteristic (ROC) Curve for Final Model
(Area Under ROC Curve (AUC)=0.895)



symptoms was also similar to that of the model with six symptoms showing an AUC of 0.946. The general scoring system including six symptoms was composed of background score (0.087), trismus score (63.86), burning sensation score (3.54), xerostomia score (4.26), sore throat score (3.29), numbness score (1.27), and ulcer score (2.57). When the cutoff point of composite score was set as 1.0, this general scoring system derived from the logistic regression model using six symptoms had sensitivity and specificity of 83.7 percent and 91.9 percent, respectively, for detecting OSF.

As shown in Table 3, it appears that most of the variation can be explained by just two variables: trismus and burning sensation. However, the ROC curve for the model including only trismus and burning sensation showed an AUC of 0.871 for early detection of OSF. When the cutoff point of composite score was set as 1.0, the sensitivity and specificity of detecting early-stage OSF were 81.8 percent and 86.7 percent, respectively. The ROC curve for the model including only trismus and burning sensation showed an AUC of 0.914 for general (without regard to MMO) detection of

OSF. When the cutoff point of composite score was set as 1.0, the sensitivity and specificity of detecting OSF were 75.6 percent and 98.3 percent, respectively.

For calculating sensitivity and specificity here, however, we have applied the scoring system to the same data set from which it has been derived. This does not quite provide sensitivity and specificity, but provides the best possible power of discrimination. To estimate true sensitivity and specificity, the scoring system should be applied to another series of OSF cases and unaffected BQCs in a blinded fashion. That is, the questionnaire should be completed by subjects before clinical examination and the clinician should not be aware of the results of the questionnaire while conducting the clinical examination.

As shown in Table 4, the higher the cutoff point of composite score, the lower the sensitivity and the higher the specificity. We may choose an appropriate cutoff point according to the purpose and situation of using the scoring system. For example, we may set a higher cutoff point for screening of a community with low OSF prevalence to obtain a higher positive pre-

dictive value and fewer false positives. On the other hand, we may set a lower cutoff point for screening a high-risk group, such as patients in a BQC special clinic, to reach a higher negative predictive value and fewer false negatives.

The questionnaire we developed here is an easy, self-administered, and noninvasive tool for the early detection of OSF. It could be used in clinics as well as in communities. In conjunction with a two-step design, it could also be used to estimate the prevalence of OSF in the general population. This questionnaire and the scoring system may be used to detect OSF in order to intervene in its progress toward malignant transformation and severe mouth opening limitation. Moreover, symptoms and signs observed in the present study are similar to those reported in the previous studies. This questionnaire is considered to be applicable to other populations because the terms of oral symptoms included in the questionnaire can be easily translated into other languages.

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