
Vulnerability and presenting symptoms in burning mouth syndrome

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Objectives. To investigate if burning mouth syndrome (BMS) patients have differing health perceptions, medication, and life experiences compared with controls and to examine the role of vulnerability factors and differentiate them from the presenting symptomology in patients with BMS.

Study design. A nonprobability convenience sample of patients presenting with BMS and age- and sex-matched controls were recruited from Queen's University, Belfast, King's College London, and Baylor College of Dentistry, Dallas. Participants completed a questionnaire to assess 9 aspects of their medical and social history, including early and past life experiences. The subjects completed the Hospital Anxiety and Depression Scale to assess current distress.

Results. Participants with BMS had significantly higher experiences of adverse early life experiences compared with controls. They had statistically significantly higher mean scores for anxiety and depression compared with controls. A hierarchical multiple logistic regression analysis showed that the characteristics of BMS included cancer phobia, gastrointestinal problems, and chronic fatigue.

Conclusion. BMS is a complex disorder. People who experience adverse life experiences may become vulnerable to developing BMS in later life.

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Burning mouth syndrome (BMS) is a condition in which the patient reports one of several patterns of burning in their mouth and lips but the affected site(s) appear clinically normal.¹ Typically patients are around 60 years of age and females are more commonly affected than males,² but this depends somewhat on the site affected.³ Psychological factors have been reported as frequently accompanying BMS.⁴⁻⁶ Although depression was principally reported in the earlier literature,⁷⁻⁹ most recent studies point to chronic anxiety as being more important in BMS.¹⁰⁻¹² However, as with any chronic condition it is difficult to link cause and effect, as any long-term illness can produce psychological disturbance.¹³⁻¹⁷

BMS is an interesting condition, as its etiology, though multifactorial, can also be conceptualized as a psychogenic-physical continuum.¹⁸ Perceiving the etiology within this theoretical framework provides a basis for the unpacking of the etiological factors into those which might be thought of as vulnerability factors and those which are associated with the presenting

symptom of BMS. The proposition that vulnerability factors located in life experiences of an individual sensitize them to react with physical symptoms to life stress may be pertinent in understanding those patients who present with conditions such as BMS. Some evidence for this hypothesis may be gained from the work of Essex et al.¹⁹ They have shown that maternal distress can sensitize children to react with increased circulating cortisol in response to stressful life events in later life. The role of early life experiences, such as maternal depression and/or distress, might be important in understanding the type of patient who presents with BMS. Bergdahl and Bergdahl's²⁰ conclusions support this view. In their 1999 paper they state that "BMS should be seen as a marker of illness and/or distress."²⁰

Previous research has suggested that BMS may present with both physical and psychological symptoms. This observation has allowed the proposition to be made that a psychogenic-physical etiological continuum exists.¹⁸ However the place of life experiences as potential vulnerability factors within this etiological framework remains unknown.

The aim of the present investigation was to investigate, first, whether BMS patients differed compared to those without BMS with regard to health perception, medication, and life experiences and, second, to examine the role of vulnerability factors and differentiate them from the presenting symptomology in patients with BMS.

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PATIENTS AND METHODS

A nonprobability convenience sample of patients presenting with BMS were recruited from 3 centers; all of the patients employed identical diagnostic criteria and identical management regimes for BMS.² The diagnostic criteria used by all participants was a burning sensation of the clinically normal oral mucosa. The 3 centers were the Department of Oral Surgery, Oral Medicine, and Oral Pathology, Queen's University Belfast, School of Medicine, King's College London, and the Department of Periodontics, Baylor College of Dentistry, Dallas, Texas. Age- and gender-matched controls were recruited from the 3 localities to mirror the demography of the BMS patients participating in each center. Informed ethical consent was given by all patients to participate.

Inclusion criteria for patients presenting with BMS were that the oral mucosa was normal on examination, no medical/physical causation (including drug side-effects) for BMS could be identified, and hematologic screening was normal (full blood picture, folic acid, serum ferritin, and glucose). BMS patients were excluded from the study if they had visible oral lesions or *Candida* infection, a medical/physical causation was found, hematologic screening was abnormal, cognition impairment, or if they refused to participate. Inclusion criteria for control patients were that they matched the age, sex, marital, and employment status of a BMS participant. Exclusion criteria for controls included experience of BMS, cognitive impairment, and refusal to participate.

A questionnaire was developed using qualitative research methods from in-depth work with a previous group of BMS patients. An example of this approach has been reported elsewhere.¹⁸ The questionnaire inquired about 9 broad aspects of the patient's medical and social history including early and past life experiences. The first part of the questionnaire concerned the patient's previous and current health status including the patient's perception of their health over the last year. Perceived health status was assessed using a 5-point Likert scale with scores ranging from 5 (very good) to 1 (very poor). In addition, the site and duration of burning in the oral cavity and elsewhere, the presence or absence of cancer phobia, and current use of prescribed medication as well as self-purchased over-the-counter medicines were also assessed. In females a distinction was made between those who were still menstruating and those who were postmenopausal. The nature of the menopause (i.e., natural or hysterectomy induced) was noted as was the administration of hormone replacement therapy (HRT).²¹ For those women still menstruating the presence of premenstrual syndrome (PMS) was noted.²²

A number of questions were asked relating to life experiences, included experiences of loss and separation

involving the mother and father. Details were also sought on family history regarding parental depression or complaints of BMS, recent bereavements, relationship to the deceased, patient reaction, and management of bereavement. Additional questions for females included feelings at the onset of menstruation, feelings towards marriage, the ability to experience satisfaction at an intimate level, and experiences of pregnancy, labor, and delivery. Male respondents were asked about their ability to gain satisfaction during intimacy with their partners. Demographic factors of age, sex, present employment status, and marital status were also detailed.

The final part of the questionnaire was the Hospital Anxiety and Depression Scale (HAD)²³ which was used to assess current distress. This 14-item reliable and validated inventory assesses anxiety and depression²¹ in patients with BMS.¹¹ The HAD is simple and easy to administer with 2 separate scales to assess anxiety and depression. There are 4 possible responses to each question which range from "not at all" (scoring 0) to "most of the time" (scoring 3). Scores greater than 10 are said to be indicative of anxiety states and depressive illness.²³

Coding of the questionnaires and statistical analysis

The completed questionnaires were coded for data entry. Statistical analysis was by SPSS (version 11, SPSS, Chicago). The data from the 3 individual sites were pooled together for statistical analysis and comparisons made between BMS and control subgroups. Chi-squared analysis was used to compare BMS and control subjects' demography, perceptions of health, and life experiences. Student's *t*-tests were used to compare the HAD anxiety and depression subscales between BMS and control subjects. Hierarchical multiple logistic regression analysis was utilized to characterize and develop an explanatory model for patients presenting with BMS. Alpha statistical significance level was set to the conventional .05.

RESULTS

The sample

A total of 160 questionnaires were obtained. There were no patient refusals. Three questionnaires, however, were not able to be evaluated so the overall response rate was 98%. Eighty-four questionnaires were completed by BMS patients and 73 by control subjects. Equivalent proportions ($P = .88$) of BMS and control subjects were recruited from the 3 locations.

Demographic profile of the sample

The age range of the patients was 25 to 97 years with a mean age of 65 years. Eighty-eight percent ($n = 138$)

Table I. Comparisons between BMS and control subjects' general health

Health item	BMS	Control	χ^2	df	P
	(n = 84) n (%)	(n = 73) n (%)			
Heart/chest disease	16 (47)	17 (51)	0.42	1	.52
Arthritis/rheumatism	39 (66)	20 (34)	8.10	1	.02
Backache	37 (73)	14 (28)	10.66	1	.001
Endocrine	2 (50)	2 (50)			1.00*
Skin problems	23 (77)	7 (23)	7.99	1	.004
Recurrent GIT problems	38 (78)	11 (22)	16.15	1	<.001
Constipation	30 (59)	21 (41)	.76	1	.38
Nausea	6 (100)	0 (0)			.02*
Gum/mouth trouble	58 (85)	10 (15)	49.99	1	<.001
Anxiety	32 (67)	16 (33)	4.81	1	.03
Depression	29 (73)	11 (28)	7.78	1	.005
Fear of cancer	34 (74)	12 (26)	10.89	1	<.001
Dizzy/giddy	15 (83)	3 (17)	7.11	1	.007
Disturbed sleep	39 (66)	20 (34)	8.09	1	.02
Chronic fatigue	31 (76)	10 (24)	11.78	1	.003

*Fisher exact probability test.

of the patients were female and 12% were male. In terms of employment 44% (n = 69) were retired, 45% (n = 71) were employed, and the remainder were unemployed. Eighty-five percent of subjects (n = 133) were or had been married. Of the married patients, 57% (n = 90) were still in their first marriage, 11% (n = 17) were widowed, and 17% (n = 26) were divorced. The marital status of the remaining subjects (15%) could not be ascertained. Equivalent proportions of control subjects (49%) and BMS subjects (51%) (P = .50) were in their first marriage.

Perception of health status of the sample

When asked about their perception of their general health over the last 12 months, 60% (n = 93) of the total sample stated that they perceived their health to be good or very good. In the previous 12 months, control subjects had significantly greater mean scores for perceived good health status (3.82; 95% confidence intervals (CI) 3.60-4.05) compared with BMS subjects (3.42; CI 3.17-3.66) (P = .02).

A significantly greater proportion of subjects with BMS than controls stated that they had suffered somatic complaints, including arthritis/rheumatism, back pain, recurrent gastro-intestinal disorders,²⁴ suffered chronic fatigue,²⁵ skin problems, persistent problems with their mouths, nausea, anxiety and/or depression, fear that they had cancer, dizziness or giddiness, and disturbed sleep patterns (Table I).

The majority of the sample as a whole (60%) took prescribed medications. Significantly larger proportions of BMS compared with control subjects took tranquilizers, hypnotics, and laxatives. Overall, in terms of

Table II. Comparisons between BMS and control subjects' medications

Medication	BMS	Control	χ^2	df	P
	(n = 84) n (%)	(n = 73) n (%)			
Prescribed tranquilizers	15 (88)	2 (11)	8.70	1	.003
Prescribed Hypnotics	16 (76)	5 (24)	4.59	1	.03
Prescribed anti-depressants	15 (58)	11 (42)	.12	1	.72
Prescribed contraceptive pill	1 (17)	5 (83)			.09*
Prescribed antacids	7 (78)	2 (22)	2.08	1	.15
Other prescribed medication	54 (58)	39 (42)	1.17	1	.27
Self-medication antacids	16 (70)	7 (30)	2.46	1	.12
Self-medication analgesics	23 (64)	13 (36)	1.78	1	.18
Self-medication laxatives	13 (77)	4 (24)	3.70	1	.05

*Fisher's exact probability test.

medication between the 2 study groups no other statistically significant differences were shown (Table II).

Subjects with BMS had significantly higher mean scores for the HAD subscale for anxiety (7.40; CI 6.34-.45) compared with controls (4.04; CI 2.14-5.94) (P = .003). In addition BMS subjects had significantly higher mean scores for the HAD subscale for depression (4.94; CI 4.05-5.83) compared with controls (2.41; CI 1.04-3.77), (P = .003).

Women's health status

Twenty-one percent (n = 30) of all female subjects were still menstruating and 17% (n = 12) stated that they experienced PMS. A significantly greater proportion of the control subjects (70%) compared with BMS patients (30%) were still menstruating (P = .005), and a significantly larger proportion of control subgroup (75%) compared with BMS patients (25%) stated that they experienced PMS (P < .05).

All the other females (n = 108) were postmenopausal. The age at which the menopause began ranged from 33 to 67 years. Fifty-five percent (n = 59) of the women had had a natural menopause. A significantly larger proportion of female BMS (71%) compared with control (23%) subjects had had a hysterectomy (P = .02). Among the postmenopausal females, 57% were taking or had taken hormone replacement therapy (HRT) for between 2 months and 31 years. Equivalent proportions of female BMS patients (56%) compared with control subjects (44%) were receiving HRT (P = .56).

Twenty-three percent (n = 31) of all female subjects stated that they were unprepared for their menarche. Sixty-five percent of BMS women compared with 36% of controls stated that they were unprepared for their menarche (P = .15). Almost a third of the BMS women stated that they "did not understand what was happening," 18% felt they had hurt themselves, 18% thought

Table III. Comparisons between BMS and control subjects' life events

Life event	BMS	Control	χ^2	df	P
	(n = 84) n (%)	(n = 73) n (%)			
Mother dead	59 (55)	48 (45)	1.21	2	.54
Mother ill when subject a child	42 (56)	31 (42)	1.44	2	.49
Mother depressed when subject a child	23 (70)	10 (30)	4.09	2	.04
Mother had BMS	2 (33)	4 (66)	3.47	2	.18
Father dead	38 (57)	29 (49)	.47	2	.49
Father ill when subject a child	22 (55)	18 (45)	.12	2	.94
Father depressed when subject a child	13 (65)	7 (35)	1.92	2	.17
Separated from parents when a child	21 (52)	20 (49)	.83	2	.65
Separation anxiety	10 (44)	13 (57)	1.10	2	.58
Recent bereavements	73 (58)	52 (42)	6.12	1	.04

they were bleeding to death, and 23% were frightened by the pain and cramps.

Nine percent (n = 12) of all the women reported that the thought of intimacy in marriage frightened them. The BMS female patients reported that the reasons for fears of intimacy included not knowing what to expect (36%), being apprehensive (25%), fears that the marriage would fail (13%), and intimacy (coitus) was wrong even in marriage (13%).

Seven percent (n = 8) of the total sample stated that they were unable to achieve orgasm. Of the women who were able to come to orgasm 45% (n = 70) achieved this only with clitoral stimulation and 28% (n = 44) reported that they had vaginal orgasms. Six women who were sexually dissatisfied stated the reason was their partner's impotency and the practice of coitus interruptus.

Comparing female BMS and control subjects, significantly larger proportions of BMS women (77%) compared with controls (23%) felt their experience of pregnancy and delivery of their children had been traumatic (P = .004). The average number of pregnancies in patients and controls, was 2. However 21% of all female subjects had suffered the loss of between 1 and 3 children.

Male health

Two of the men in the total sample stated that were impotent.

Life experiences

Comparisons between the life experiences of control and BMS patients showed that a significantly larger proportion of BMS compared with control subjects had a mother who was depressed and had suffered a recent bereavement. No other significant differences were shown (Table III).

Twenty-nine percent (n = 24) of BMS patients still had a mother living and of the 71% (n = 133) whose

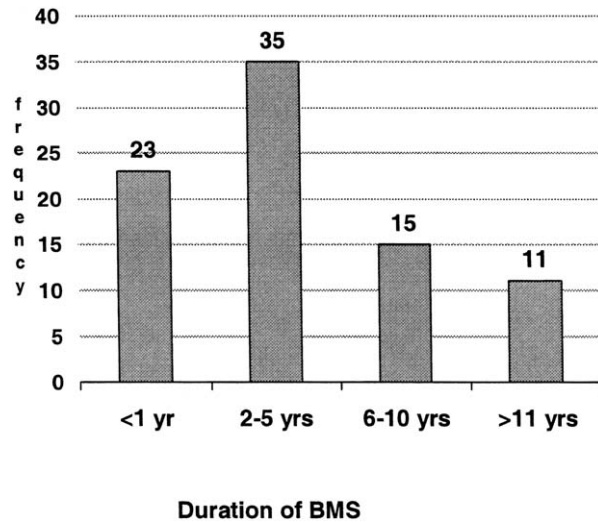


Fig 1. Duration in years of burning for BMS patients.

mother had died, 16% (n = 21) were under 21 years when this happened. Nineteen (27%) remembered their mother being away when they were children because she was in hospital (n = 14) for prolonged periods, she had died (n = 3), or she had left home (n = 2). Over 50% (n = 42) remembered their mother being ill, and 27% (n = 23) recalled their mother suffering from depression. Two percent (n = 2) remembered their mother complaining of a burning mouth.

In relation to the patients' history of their father, 19% (n = 15) still had a father living, but of the patients whose father had died in 22% (n = 14) of cases this happened when the patient was under 21 years of age. Twenty-six percent (n = 22) recalled their father being away when they were small, either on work or on business (26%), in the armed forces (25%), in hospital (10%), because of divorce (10%), or as a prisoner of war (5%). Forty-six percent of BMS respondents stated that their fathers had been ill (n = 38), and 20% (n = 13) recalled their father suffering from depression.

The duration and onset of the BMS

The duration of the burning was variable, as summarized in Fig 1. The median length of time of experiencing BMS was between 2 and 5 years. Twenty percent (n = 16) of the BMS sample reported burning at sites other than the mouth. Experience of burning vulva and heartburn were most commonly cited.

Eighty-three percent (n = 70) of patients had had an operation prior to the onset of the burning. A third of BMS patients (n = 21) had had gynaecological surgery, 13% (n = 11) had teeth extracted, 11% (n = 9) had abdominal surgery, 6% (n = 5) had urinary tract surgery, and 36% (n = 30) had various other operations.

Table IV. Characteristics of patients presenting with BMS

<i>BMS patient characteristics</i>	<i>Relative odds (exp B)</i>	<i>(± 95% CL)</i>
Cancer phobia	2.93	(1.28, 6.71)
Chronic fatigue	2.70	(1.09, 6.69)
Recurrent GIT problems	3.15	(1.34, 7.47)

Predicting the characteristics of patient presenting with BMS

Subjects categorized as BMS patients were designated a score of 0. Control subjects were awarded a score of 1. Using this designation of BMS, an hierarchical multiple logistic regression analysis was used to characterize patients presenting with BMS. The independent variables were introduced in 2 blocks. The first block consisted of demographic variables (age, gender, and location) defined by 3 dummy variables with the youngest aged group, male subject, and Dallas sample, acting as baseline. This block acted as a control to remove demographic variance, and was forced into the equation as a first step. The second block consisted of those variables which had been shown to differentiate between BMS and control subjects. The logistic regression results are expressed as relative odds (with 95% confidence limits). These indicate the likelihood (on an odds scale) that patients presenting with BMS will have certain characteristics compared with the likelihood among control patients who do not have the characteristics. Patients most likely to present with BMS were 2.9 times more likely to be cancerphobic, 2.7 times more likely to suffer from chronic fatigue, and 3.2 times more likely to have recurrent gastrointestinal tract (GIT) problems. No other significant factors could be shown (Table IV).

Three further logistic regression analyses were undertaken to predict the characteristics of BMS patients with cancer phobia, chronic fatigue, and recurrent GIT problems. Patients presenting with BMS and who were cancerphobic were 4.3 times more likely to take hypnotics and 2.1 times more likely to have recurrent GIT problems (Table V). Patients presenting with BMS who had chronic fatigue were 3 times more likely to have recurrent GIT problems and 2.3 times more likely to have a disturbed sleep pattern (Table 5). Patients presenting with BMS who had recurrent GIT problems were 2.4 times more likely to have had a mother who suffered depression in their childhood and 2.2 times more likely to have suffered a recent bereavement (Table V).

DISCUSSION

Health differences were shown to exist between patients presenting with BMS and the control subjects. Patients with BMS stated that their overall health was poorer, complained of more illnesses and gastrointestinal

Table V. Characteristics of BMS patients with cancer phobia, chronic fatigue, and recurrent GIT problems

<i>BMS patient characteristics with</i>	<i>Relative odds (exp B)</i>	<i>(± 95% CL)</i>
Cancer phobia		
Hypnotics	4.33	(1.54, 12.12)
Recurrent GIT problems	2.70	(1.00, 4.49)
Chronic fatigue		
Recurrent GIT problems	3.14	(1.42, 6.90)
Disturbed sleep pattern	2.37	(1.04, 5.39)
Recurrent GIT problems		
Maternal depression	2.40	(1.00, 5.74)
Recent bereavement	2.21	(1.03, 4.76)

problems,²⁴ felt chronically fatigued,²⁵ had disturbed sleep patterns, and felt more anxious and/or depressed compared with the control subjects. The differences in ill-health perception were reflected in the types of prescribed medication (e.g., tranquilizers, hypnotics) and over-the-counter medicines (such as laxatives) taken by BMS patients compared with the control subjects. Although the sample was matched for age and gender more of the control subjects compared with the BMS female patients were still menstruating as a greater proportions of the BMS female patients had had a hysterectomy. This suggested the likelihood that the BMS women had experienced a premature menopause. It is possible to propose from these findings that in terms of physical and emotional functioning the BMS subjects were in some way different to those in the control group.

Although no differences could be identified with regard to their intimate lives, the life experiences of the BMS patient group compared with controls were dissimilar. The BMS respondents had mothers who were depressed, had greater experience of difficult confinements and deliveries of their children, and had experienced a recent bereavement. In view of these results it seemed appropriate to suggest that people with BMS also comprised a different population with regard to their life experiences.

It seems reasonable to propose that perceptions of ill health may be related to their presenting symptomatology and that their life experiences are associated with a vulnerability to experience emotional difficulties as physical symptoms (Fig 2). Support for this proposition may be found in the findings of the logistic regression analysis. Patients presenting with BMS were characterized by gastrointestinal problems, chronic fatigue, and cancer phobia. It seemed that gastrointestinal problems, chronic fatigue, and cancer phobia represented a constellation associated with the presenting symptomatology of BMS and as such could be considered as being associated with seeking professional help.²⁶

The probability of chronic fatigue and cancer phobia were associated with a disturbed sleep pattern and the use

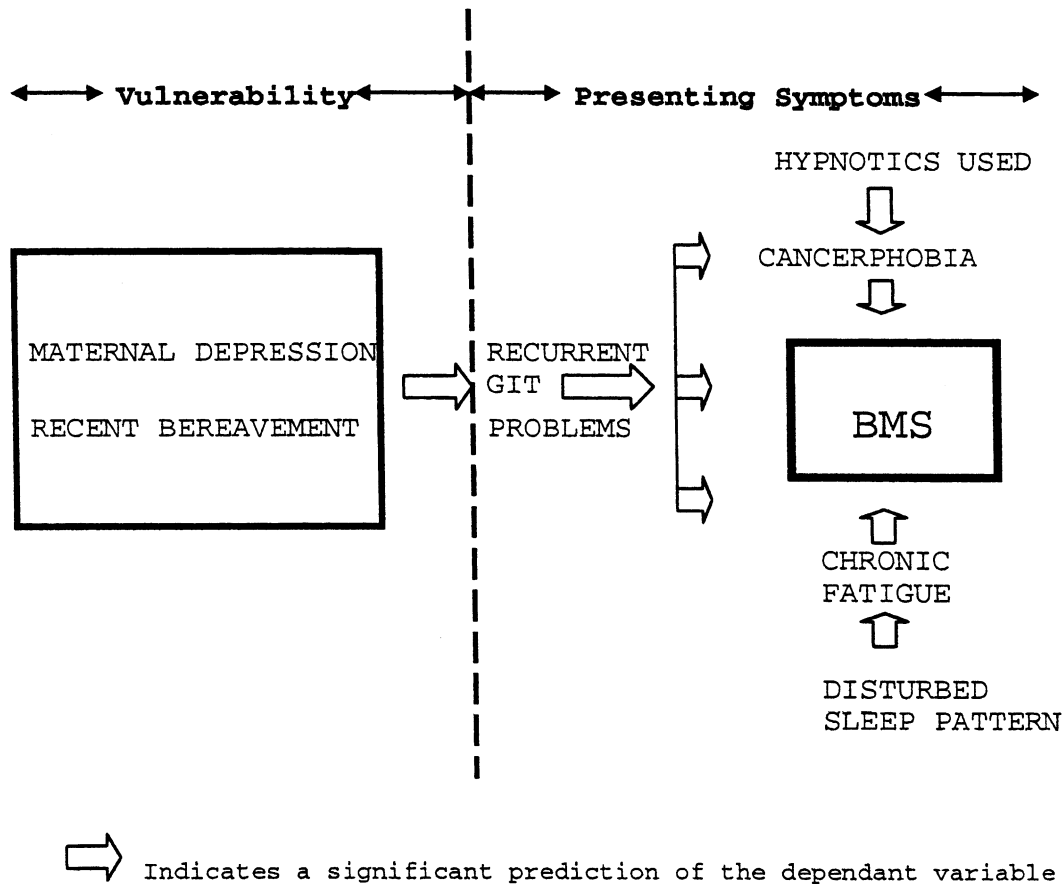


Fig 2. A predictive model for patients presenting with BMS.

of hypnotics, whereas the probability of gastrointestinal problems was related to their life experience of maternal depression and recent bereavements. It seems that these predictors compared with those for chronic fatigue or cancer phobia represented a vulnerability associated with life experiences which may have sensitized the individual to react with physical symptomatology to current stressful life events as suggested by Essex et al.¹⁹ The onset and duration of BMS would tend to support this hypothesis because the BMS respondents presented with physical (gastrointestinal problems) and emotional concomitants (cancer phobia, chronic fatigue, disturbed sleep) of anxiety. Furthermore, the onset was related to physical symptoms and surgical intervention.

The findings of this investigation are supported by Hammaren and Hugoson,²⁷ who have shown that BMS patients have experienced “catastrophes in their lives in the form of stillborn children or children born with various handicaps, children injured in accidents or prolonged social problems.” Korszun²⁸ has proposed that such “adverse life events” together with “the interaction between underlying predisposition and environmental stress” are needed if vulnerable individuals are to

experience life stresses as chronic pain. Therefore, as Bergdahl and Bergdahl²⁰ insist, clinicians must appreciate that BMS may be indicative of “distress,” thus providing a rationale and support for considering underlying (vulnerability) factors as potential markers for those who experience emotional distress as bodily illness. In the present study, patients with BMS had significantly higher mean scores for both anxiety and depression HAD subscales,^{11,23} hence supporting Bergdahl and Bergdahl’s²⁰ proposition that BMS may be indicative of emotional distress.

One consideration of the present study is that although a verifiable psychological inventory²³ was used to measure anxiety and depression the remaining questionnaire items were developed from in-depth work with a previous group of BMS patients. This may call into question the generalizability of the findings. In contrast, however, one of the strengths of the study relates to the fact that the subjects were sampled from 3 geographically separate institutions and as such this improves the strength and generalizability of the findings with regard to vulnerability in patients with BMS.

Therefore, while suggestions of a more cognitive²⁹⁻³¹ framework or a physical causation associated with trigeminal neuropathy³² may be proposed, what is suggested here relies on the idea of a psychogenic-physical (mind-body) continuum in which unbearable thoughts and feelings are experienced as bodily symptoms.^{18,33} The life experiences of people with BMS provides the emotional backdrop (vulnerability) for the affects associated with loss and disappointments to be expressed physically as the presenting symptomatology. The role of life experiences and the presenting symptoms must be considered as complementary factors and be included in the initial assessment of BMS patients.²⁰ The issue of vulnerability factors as concomitant to the onset and presentation of BMS remains an area for further research.

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