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Original Research Article

Angina bullosa hemorrhagica: a rare condition?

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Abstract

Introduction: Angina bullosa hemorrhagica is characterized by oral mucosal blood-filled vesicles or blisters. These lesions, however, are frequently asymptomatic and they are only observed when their content is spilled over the oral cavity. **Objective:** The aim of this study was to perform a retrospective evaluation of Angina Bullosa Hemorrhagica (ABH) cases diagnosed in patients referred to the Diagnosis Center of Oral Diseases of the Dentistry School of Federal University of Pelotas, over a 4-years period. **Material and methods:** All the clinical records from this period were reviewed and Angina Bullosa Hemorrhagica could be diagnosed in 47 patients. The following information pertaining to the patient and the lesion were collected from the dental record: gender, age, ABH site, size of the lesion, presence of symptoms, bleeding, likely predisposing factors, treatment and recurrence of the lesion. **Results:** The results revealed 47 patients with clinical features of ABH, 26 were female and 21 were male, with ages between 42 to 87 years. Thirty-six patients (76.6%) developed the lesion in the palate. Pain was reported by 17 patients (36.2%) and bleeding by 19 patients (40.4%). Seventeen patients (36.2%) reported the occurrence of the lesion following trauma. **Conclusion:** It is possible to conclude that although ABH has been traditionally described as a rare condition, the data obtained in this study suggest a fairly high occurrence of the disease.

Introduction

Angina bullosa hemorrhagica (ABH) was first described by Badham [1] in 1967, and it is characterized by oral mucosal blood-filled vesicles or blisters [9]. These blisters present a color ranging from dark red to purple and may cause some discomfort [4]. However, they are frequently asymptomatic and are only observed when their content is spilled over the oral cavity.

ABH may occur either as solitary or multiple lesions [8, 11]. As rule, they affect the soft palate, but these lesions can also occur in the anterior pillar of the fauces, epiglottis, arytenoids, pharyngeal wall and esophagus [4, 7, 11, 12, 13]. Both middle-aged and elderly individuals are more affected by ABH [9, 11].

ABH is an acute disease which is generally associated with trauma [4, 7, 9, 13, 14] mainly during food ingestion, and can eventually present frequent recurrences [11]. In addition to trauma, the use of steroid-based inhalers [3, 5, 6, 8, 9, 10, 11] and Diabetes Mellitus [3] have been reported as predisposing factors for ABH.

Treatment usually aims to relieve discomfort caused by blister disruption and to improve the healing of ulceration [12]. In patients who use steroid-based inhalers regularly, water gargling following medicine application can be an effective way to prevent ABH [3].

The aim of this study was to assess the prevalence of ABH at the Diagnosis Center of Oral Diseases of the Dentistry School from the Federal University of Pelotas in a 4-year period, evaluating clinical characteristics, likely predisposing factors and treatment.

Material and methods

The present study had been approved by the Ethical Committee of Federal University of Pelotas. Over a 4-year period, from 2005 to 2009, 10,244 patients were referred to the Diagnosis Center of Oral Diseases of the Dentistry School of Federal University of Pelotas. All the clinical records from this period were reviewed and Angina Bullosa Hemorrhagica could be diagnosed in 47 patients.

From the 47 patients presenting Angina Bullosa Hemorrhagica, 26 were female and 21 were male, the patients were between 42 and 87 years old.

The following information pertaining to the patient and the lesion were collected from the dental record: gender, age, site for ABH, size of the lesion, presence of symptoms, bleeding, likely predisposing factors, treatment and recurrence of the lesion.

Results

Palate was the most common site for ABH, with 36 patients (77%) of the 47 cases presenting the lesion in this area. The size of the lesion ranged from 0.5 cm to 2 cm, however, most lesions are not bigger than 1 cm in diameter.

Only 3 patients (6.4%) presented non-ruptured blisters. Twenty-three lesions (48.9%) were ulcer-like types at the time of diagnosis, while the remaining patients did not present active lesions at the time of dental appointment, and the diagnosis was established based on the disease history.

The information regarding the time for healing was available in only 20 clinical records. Lesion repair occurred within 10 days in 18 cases, and only 2 lesions took up to 10 days to heal. Local corticoid was prescribed in 16 cases (34%), with the aim of symptoms relief.

Seventeen patients (36.1%) reported painful symptomatology upon lesion occurrence. In 12 cases (25.5%) the pain was reported as mild, and in 5 (10.6%) cases as severe. Nineteen patients (40.4%) reported mild bleeding.

Diabetes and trauma appeared as likely predisposing factors in ABH occurrence. Seventeen patients (36.1%) had recognized the lesion following some trauma, 9 of whom (19.1%) specified that trauma had occurred during food ingestion. Two trauma-reporting individuals belonged to the diabetic patient group (n=4). Thus, two concomitant predisposing factors in ABH occurrence were revealed. With reference to other systemic alterations, 17 (36.1%) patients presented hypertension and 16 (34%) were hypertensive drugs users.

Thirteen patients (27.6%) showed lesion recurrence within a 12 months period in most cases. In these cases, the lesion area and duration of the recurred condition were similar to those in the first episode of the disease.

Discussion

ABH has been traditionally described as a relatively rare alteration [1, 9]. However, the data in this study suggest that the condition may have a higher occurrence, since 47 cases were diagnosed in 5 years. Also Slezák [12] and Yamamoto *et al.* [14] related that ABH has been considered to be more common that previously recognized. It is also worthy to mention that this number may be even higher, taking in account the fact that ABH often presents no symptoms and is not perceived by the patient, thus not diagnosed.

In relation to clinical characteristics, the results of this study have proved a typical ABH age distribution among middle-aged and elderly individuals, as well as a strongly higher prevalence in the palate (76.6%), which confirms other authors' reports [4, 8, 12].

In accordance with Giuliani *et al.* [4] and Slezák [12], a slightly higher female prevalence was found in the present research, in which 55.3% of the ABH-affected patients were female.

The ABH causing factors remain unclear. However, some predisposing factors such as steroid-based inhalers and Diabetes mellitus are mentioned [2, 5, 6, 10, 14]. We were unable to demonstrate this relationship in our sample, once only 4 patients were diabetic and none of them reported the use of corticosteroids. Trauma, mainly during food ingestion, remains the most likely cause for ABH occurrence [3, 4, 7, 13] and was reported by a number of patients (36.1%) in the study.

The high number of hypertensive patients in the sample (36.2%) is striking. However, this factor tends to be more closely associated with age, and possibly do not represents a likely predisposing factor for the ABH. Hypertension is a rather common symptomatic condition in adults, and the connection between this condition and ABH occurrence remains a mere speculation [8]. Further studies are required in order to reach a final conclusion.

ABH lesions should be differentiated from other kinds of subepithelial blisters, such as those observed in epidermolosis bullosa, bullous lichen planus, pemphigus vulgaris, linear IgA disease, amyloidosis and stomatitis herpetiformis [4, 11]. The characteristic clinical history allows diagnosis confirmation without the need of a biopsy. A detailed anamnesis is indispensible so that the dentist can reach the correct diagnosis, avoiding procedures which will cause the patient unnecessary strain.

The results obtained in this study, support that ABH is a fairly common disease. Thus, it is important to the dental professional to know its characteristics, to diagnose the lesions, and to clarify to the patient the nature of the lesion.

Conclusion

It is possible to conclude that, although ABH has been traditionally described as a rare condition, the data obtained in this study suggest a fairly high occurrence of the disease.

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