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Original

Angina bullosa hemorrhagica of the soft palate: a clinical study of 16 cases

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Abstract: Angina bullosa hemorrhagica (ABH) is an oral mucosal blood blister that develops without blood dyscrasia or vesiculobullous disorder. Although a minor mucosal trauma has been suggested as a triggering factor for ABH, its etiopathogenesis, especially the causative role of systemic conditions, is largely unknown. We investigated the presence or absence of local factors as well as systemic background disease in 16 patients with ABH arising in the soft palate. All the lesions were solitary, and 75% of them (n = 12) appeared during the ingestion of hard or crispy food. With regard to underlying systemic conditions, hypertension was the most common (n = 6), and asthma, insomnia, diabetes mellitus, rheumatoid arthritis, gastrointestinal disorder and hyperuricemia were also recorded (n = 1 each). Five patients had no significant background disease. There were no recalcitrant or recurrent cases. In conclusion, the present study has revealed that scratching of the oral mucosa during eating plays an important role in the formation of ABH. Hypertension appears to be the most frequent background condition, but its pathogenic relationship with ABH remains speculative, as hypertension is fairly common in adults. (J. Oral Sci. 50, 33-36, 2008)

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Introduction

In 1967, Badham (1) advocated the term angina bullosa hemorrhagica (ABH) for an oral mucosal blood blister that occurs with no evidence of blood dyscrasia or vesiculobullous disorder. Clinically, ABH is a solitary and often painful lesion, and primarily affects the soft palate of middle-aged adults. The buccal mucosa and the lateral border of the tongue are also affected, although infrequently (2-4). Multiple lesions are exceptional, and repeated occurrences have been reported in some patients (1-9). ABH soon heals spontaneously, but for advanced lesions with suspected airway obstruction, active treatment is recommended (3,4). Although the diagnosis is non-problematic in most cases, it is important to critically distinguish ABH from more serious conditions including various hemorrhagic disorders and bullous diseases. As to the etiopathogenesis of ABH, both local mucosal trauma and the long-term use of steroid inhalers have been suggested in the literature (2-4,9-13). However, the predisposing systemic diseases have not been studied in detail.

In the present study, we retrospectively investigated the presence or absence of local factors and systemic conditions in 16 patients with ABH. To our knowledge, this is the largest Japanese case studies to have appeared in the literature.

Materials and Methods

From the medical records filed in the Oral Surgery Clinic of Saitama Medical Center during the last 6 years (2001-2006), 16 cases of ABH were retrieved. All of the patients underwent serum examination (blood count and baseline coagulation tests). Previously reported cases of ABH were also thoroughly reviewed for comparison (2-4,6-9).

Results

As shown in Table 1, the mean age of the 16 patients was 50.9 years, ranging from 26 to 74 years. The male to female ratio was 1.7:1. The size of the lesion ranged from 1.0 to 3.5 cm, with a mean of 2.0 cm. All the cases were solitary and appeared during or just after eating. Twelve patients were recorded as having eaten hard or crispy food of various types. The results of serum examination were within normal limit in all patients. The most frequent underlying systemic condition was hypertension (n = 6), and asthma (steroid-based inhaler user), insomnia, diabetes mellitus, rheumatoid arthritis, gastrointestinal disorder and hyperuricemia were also found (n = 1 each). Five patients underwent aspiration therapy with antibiotic medication, and three were given medication only. Two patients were given antihypertensive therapy, and one of them was admitted because of possible airway obstruction. No active treatment was given to the remaining six patients. None of the cases recurred.

The published data on local and systemic factors possibly involved in the pathogenesis of ABH are summarized in Tables 2 and 3. Local factors were found in 24.1-100% of cases, and eating accounted for 77.8-100% (Table 2). In the previous reports (3,4), dental treatments (n = 2) and coughing and sneezing (n = 1) were also listed (Table 2). As shown in Table 3, various systemic conditions were recorded in more than 44.4% of the patients. Hypertension (n = 9) was the most frequent disease, followed by diabetes mellitus (n = 7), ischemic heart failure (n = 5) and liver disease (n = 3). However, there was no significant background disease in the formation of ABH.

Table 1 Clinical summary of 16 cases of angina bullosa hemorrhagica

Case	Gender	Age	Size (cm)	Ingestion of hard or crispy food	Systemic conditions	Treatments
1	F	45	3.5	+ (coarsely-ground pepper)	Insomnia	AB
2	M	43	2.0	-	-	AB
3	M	70	3.2	-	Diabetes mellitus	AB
4	F	31	1.2	+	-	AS, AB
5	M	46	2.8	+	-	-
6	M	51	2.0	+ (rice cake)	Hypertension	AB, AH
7	F	66	1.5	-	Hypertension	AS, AB
8	M	63	UK	-	Hyperuricemia, Gastrointestinal disorder	AS, AB
9	M	63	UK	+ (french bread)	Rheumatoid arthritis	AS, AB
10	M	33	1.5	+ (pork cutlet)	Hypertension	-
11	F	34	1.0	+	Asthma (steroid inhaler)	-
12	M	64	3.3	+	Hypertension	AB, AH
13	M	67	1.5	+	Hypertension	-
14	F	74	1.3	+ (fried fish)	Hypertension	-
15	M	39	1.0	+ (fried prawn)	-	AS, AB
16	F	26	1.5	+ (fried oyster)	-	-

AB, antibiotics; AH, antihypertensive; AS, aspiration; UK, unknown.

Table 2 Reported local factors

Authors	Total number of cases	Eating	Dental treatment	Cough and sneeze	Total
Hopkins and Walker (2)	9	7	1	1	9
Stephenson et al (3)	30	15	1		16
Deblauwe and van der Waal (6)	9	5			5
Jinbu et al (7)	6	3			3
Grinspan et al (8)	54				13*
Giuliani et al (4)	8	7			7
Yamamoto et al (9)	11	7			7
Horie et al	16	16			16

*Detailed data were not provided.

Table 3 Reported systemic conditions

Authors	Total number of cases	GI	AN	DM	RT	HT	LD	AT	CRF	IHD	NF	IS	HU	RA	KG	Total
Deblauwe and van der Waal (6)	9			1						3						4
Giuliani et al (4)	8		1	1	1										1	4
Yamamoto et al (9)	11			4		3	3		1	2	1					14
Horie et al	16	1		1		6		1				1	1	1		12

GI, gastrointestinal disorder; AN, anemia; DM, diabetes mellitus; RT, radiotherapy; HT, hypertension; LD, liver disease; AT, asthma; CRF, chronic renal failure; IHD, ischemic heart disease; NF, nephritic syndrome; IS, Insomnia; HU, hyperuricemia; RA, rheumatoid arthritis; KG, kidney graft.

Discussion

Traditionally, ABH has been considered as an idiopathic condition (1). However, accumulated experience has shown that minor mucosal insults may be involved in the pathogenesis of ABH (2,3,14). A search of the literature revealed that food ingestion was the most common cause, accounting for 50-100% of cases (2-4,6,7,9). It is interesting to note that mucosal irritation due to food ingestion is the primary cause of esophageal hematoma (15).

The soft palate, the most common site for ABH, is covered by a thin, friable squamous epithelium of the non-keratinized type (3,16). Considering the fragility of the soft palate mucosa, it is easy to speculate that submucosal hemorrhage may be elicited even by subclinical trauma (1). It is also noteworthy that mastication significantly increases the blood flow rate in the soft palate via parasympathetic reflex vasodilatation (17). Collectively, the soft palate is easily injured during mastication of hard or crispy food, and is prone to ABH (5,13).

It is now clear that the use of steroid-based inhalers for the treatment of asthma is an additional pathogenic factor (10-14). The long-term (> 5 years) use of such inhalers reportedly induces mucosal atrophy and a decrease of submucosal elastic fibers (4,10). These sequential changes may evoke weakness and breakdown of capillaries, resulting in the formation of ABH. In middle-aged adults, similar morphologic alterations can be observed as senile atrophy. This matches well with the fact that ABH invariably affects older individuals (12).

In the present series, the most common systemic condition was hypertension (37.5%), but four of our six cases were well medicated. None of the lesions were multiple or recurrent, indicating the possible participation of hypertension in the onset of ABH. Since hypertension is fairly common in adults, it is difficult to definitively explain its causal relationship with ABH. The sudden onset of a solitary blood blister on the soft palate is diagnostic of ABH, and biopsy is not necessary for most lesions. In general, no active treatment is needed, but large blisters should be broken to prevent airway obstruction (4). The prognosis of ABH is uneventful, and spontaneous healing can be expected within 7-10 days (1,4,9).

In summary, it is apparent that various mechanical stimuli, especially the ingestion of hard or crispy food, may be a direct initiating factor for ABH. To clarify the presence or absence of susceptibility to ABH, further case studies are recommended.

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