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CASE REPORT

Oral Health Care Management in Atypical Oral and Cutaneous Bullous Pemphigoid

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ABSTRACT

Background: *Bullous Pemphigoid (BP) is the most common subepidermal bullous autoimmune disease, typically presenting as tense bullae. Atypical presentations of bullous pemphigoid often mimic TEN or impetigo, creating diagnostic ambiguity. Effective management in these cases requires integrated strategies to address systemic health factors and extensive oral mucosal involvement.* ***Objectives:*** *This report highlights the Oral Health Care Management with atypical oral and cutaneous manifestations in Bullous Pemphigoid.* ***Case:*** *A 58-year-old female with a history of Diabetes Mellitus and heart disease was referred with extensive bullae, erosions, and "honey-like" crusts involving over 30% of her body surface area. Her condition worsened despite two weeks of treatment with acyclovir for suspected herpes. The presentation was highly suggestive of TEN and impetigo.* ***Case Management:*** *Management of the oral and perioral lesions focused on infection prevention and pain relief. Debridement was performed using sterile gauze with normal saline and 0.2% chlorhexidine. This was followed by the application of Aloevera extract gel/spray. Comprehensive systemic therapy, including corticosteroids and immunosuppressants, was coordinated by a multidisciplinary team to control the autoimmune disease and comorbidities.* ***Conclusion:*** *The presence of comorbidities, such as diabetes, further complicates the systemic management and heightens the risk of secondary infections, requiring meticulous wound care. A multidisciplinary diagnostic algorithm, supported by supplementary examinations, is crucial for differentiating atypical BP from TEN and impetigo infection. Adequate management of the associated oral and perioral manifestations is an integral component of comprehensive patient care.*

Keywords: *Oral Health Care, Bullous Pemphigoid, Toxic Epidermal Necrolysis, Impetigo Multidisciplinary Approaches.*

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Denta Jurnal Kedokteran Gigi, Februari 2026; Vol.20 No.1; Hal 1-9
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INTRODUCTION

Bullous Pemphigoid (BP) is the most common subepidermal bullous autoimmune disease, accounting for up to 80% of subepidermal immune-bullous cases and primarily affecting elderly patients, generally between the ages of 60 and 80 [1,2]. Pathogenetically, BP is caused by IgG autoantibodies targeting hemidesmosome components in the Basement Membrane Zone (BMZ), especially the BP180 (collagen XVII) and BP230 antigens, leading to the separation of the epidermis from the dermis and the formation of tense bullae. The classic clinical manifestation is an eruption of tense bullae accompanied by severe pruritus on normal or erythematous skin, which significantly impacts the patient's quality of life and mortality [3,4].

Although the clinical presentation of BP is often characteristic, the diagnosis of BP can be difficult due to variable clinical phenotypes [2]. Atypical presentations can resemble much more severe and life-threatening acute bullous diseases, such as Toxic Epidermal Necrolysis (TEN), which histopathologically involves full epidermal keratinocyte necrosis, contrary to the subepidermal clefting in BP [5]. Furthermore, ruptured bullae often undergo secondary superinfection, which in this case results in yellowish crusts (honey-like appearance) similar to the characteristic features of Impetigo. The distinction between atypical BP, TEN, and Impetigo is critical because the management protocols and prognosis for these conditions differ significantly. Confirmation of the diagnosis cannot rely solely on clinical features [6].

A systematic diagnostic algorithm is necessary to overcome the diagnostic challenges caused by atypical and overlapping clinical manifestations. The definitive diagnosis of BP actually requires confirmation through supplementary examinations, including a biopsy for histopathology (showing a subepidermal cleft with a rich eosinophil infiltrate) and Direct Immunofluorescence (DIF) on perilesional skin (showing linear deposition of IgG and/or C3 at the BMZ), which is the gold standard for autoimmune bullous diseases [1,5]. Additionally, serological tests such as ELISA detecting anti-BP180/BP230 antibodies have high sensitivity and specificity [2]. The presence of systemic comorbidities, such as uncontrolled Diabetes Mellitus (DM) in this case, further complicates disease management and increases the risk of infectious complications, demanding an interprofessional approach [7].

Oral mucosal involvement, though less common than skin lesions, is reported in a number of BP patients. Oral lesions in the form of bullae, which inevitably can rupture into painful erosions, may lead to difficulty eating (dysphagia) and speaking, and they are a high potential source of infection. Therefore, for Oral Medicine specialists, the management of oral manifestations encompassing pain management, prevention of secondary infection, and supportive care is a crucial component of comprehensive treatment [6,8]. This case report aims to present a case of BP with highly atypical cutaneous and oral manifestations, highlight the challenge of distinguishing it from TEN and Impetigo, and specifically emphasize the strict diagnostic algorithm and outline the comprehensive management of oral manifestations as an essential contribution to the management of BP patients with complex clinical presentations.

CASE

A 58-year-old female was referred to the Emergency Department of Dr. Ramelan Naval Hospital with extensive bullae and crusting over her entire body. The patient had previously been treated with Acyclovir (5x800 mg) for two weeks from another healthcare facility for suspected herpes, but showed no improvement, leading to a worsening condition and referral to a higher-level facility for follow-up. The patient had a history of Diabetes

Denta Jurnal Kedokteran Gigi, Februari 2026; Vol.20 No.1; Hal 1-9
Available at http://journal-denta.hangtuah.ac.id/index.php/jurnal/issue/view/41
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Mellitus (routine therapy: vidagliptin 50 mg, Gliclazide 40 mg) and heart disease (bisoprolol 5 mg).

Vital sign examination showed a decreased level of consciousness (GCS 325), blood pressure 135/41 mmHg, pulse 99x/minute, body temperature 36.5°celcius, respiration rate 40 bpm, and oxygen saturation 96%. Extraoral clinical examination revealed bullae, erosions, and "honey-like appearance" crusts, yellowish-brown, tense, and painful, involving over 30% of the total body surface area, including the face, superior and inferior labial areas, perioral area, hands, and feet, resembling the features of TEN (Toxic Epidermal Necrolysis) and impetigo (Figure 1). Intraoral examination was limited because the patient had difficulty opening her mouth, but accessible areas (labial mucosa and gingiva) appeared normal. The initial working diagnosis was suspected Bullous Pemphigoid with differential diagnoses of impetigo and TEN.

Figure 1. Clinical manifestations of the patient at visit 1, (a) showing "honey-like appearance" crusts on the face, including the labial and perioral regions; (b) reddish-brown to blackish erosion on the foot; and (c) tense bullae on the fingers and wrist skin.

CASE MANAGEMENT

Supplementary examinations performed included a complete blood count, liver function, kidney function, random blood glucose, HbA1c, urine culture, blood culture (Table 1), and bacterial culture from an oral swab of the crusted lesions (Table 2).

Denta Jurnal Kedokteran Gigi, Februari 2026; Vol.20 No.1; Hal 1-9
Available at http://journal-denta.hangtuah.ac.id/index.php/jurnal/issue/view/41
DOI: https://doi.org/10.30649/denta.v20i1

Table 1. Results of the supportive examinations included a complete blood count, liver function, kidney function, random blood glucose, HbA1c, urine culture, blood cultur

Date	Tested Markers	Result	Unit	Reference Range	Comment
28 Sep 2025	Leukosit (WBC)	19.88	×10^3/µL	4.00-10.00	High
	Neutrofil%	90.6	%	50.0-70.0	High
	Limfosit%	4.5	%	20.0-40.0	Low
	IMG%	2.9	%	0.00-0.99	High
	Hemoglobin	10.6	g/dL	12-15	Low
	Hematokrit	32.4	%	37.0-47.0	Low
	Trombosit (Platelet)	516	×10^3/µL	150-450	High
	PCT (Plateletcrit)	4.7	×10^3/µL	0.108-0.282	High
	Albumin	2.21	mg/dL	3.50-5.20	Low
	Glukosa Darah Acak (GDA)	580	mg/dL	<200	Critically High
	Kreatinin	2.5	mg/dL	0.6-1.5	High
	BUN	90.2	mg/dL	10-24	High
	Na (Sodium) serum	126.6	mEq/L	135-147	Low
	K (Potassium) serum	7.7	mmol/L	3.0-5.0	Critically High
	PO2	185.1	mmHg	80.0-100.0	High
	Urine ALB	150	g/L	≤0.02	High
	Urine RBC (Eritrosit)	48.8	/hpf	0-3	High
	Urine WBC (Lekosit)	7.7	/hpf	0-5	High
	Urine CAST (Silinder)	11.1	/hpf	0-2	High
	Urine BACT (Bakteri)	7.9	/hpf	0-2	High
29 Sep 2025	Albumin	2.09	mg/dL	3.50-5.20	Low
	K (Potassium) serum	7.17	mmol/L	3.0-5.0	Critically High
	Cl (Chloride) serum	112	mEq/L	95-105	High
30 Sep 2025	Albumin	2.28	mg/dL	3.50-5.20	Low
08 Oct 2025	Leukosit (WBC)	15.19	×10^3/µL	4.00-10.00	High
	Neutrofil#	13.97	×10^3/µL	2.00-7.00	High
	Neutrofil%	92	%	50.0-70.0	High
	Limfosit%	6.6	%	20.0-40.0	Low
	Monosit%	1.3	%	3.0-12.0	Low
	Hemoglobin	10.6	g/dL	12-15	Low
	Hematokrit	33.2	%	37.0-47.0	Low
	MCHC	31.9	g/dL	32-36	Low
	Trombosit (Platelet)	97	×10^3/µL	150-450	Low
	MPV	13.3	fL	6.5-12.0	High
	P-LCR	51.1	%	11.0-45.0	High
	Albumin	2.17	mg/dL	3.50-5.20	Low
09 Oct 2025	Albumin	2.52	mg/dL	3.50-5.20	Low
10 Oct 2025	Albumin	3.4	mg/dL	3.50-5.20	Low

Denta Jurnal Kedokteran Gigi, Februari 2026; Vol.20 No.1; Hal 1-9
Available at http://journal-denta.hangtuah.ac.id/index.php/jurnal/issue/view/41
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Table 2. Bacterial culture results of the oral and perioral lesion swab specimen and antibiotic susceptibility test

Category	Parameter	Result	Interpretation/Clinical Note
Specimen Details	Specimen Type	Swab	
	Completion Date	October 5, 2025	
Bacterial Isolate	Organism Found	Serratia marcescens	Identified as potential causative agent of infection.
	Intrinsic Property	Intrinsically resistant	
Antibiotic Susceptibility	Sensitive (S)	Ciprofloxacin, Gentamicin, Meropenem, Trimethoprim-Sulfamethoxazole	Effective antibiotics for treatment.
	Intermediate (I)	Tigecycline	
		Amikacin, Amoxicillin, Ampicillin, Cefazolin, Cefepime, Cefotaxime, Ceftazidime, Ceftriaxone, Nitrofurantoin, Piperacillin-	Antibiotics deemed

The management of this case involved several disciplines, including internal medicine, dermato-venereology, and oral medicine. This case report highlights the management of oral and perioral lesions, which aimed to prevent secondary infection and reduce pain symptoms in the affected areas.

Debridement, which is the initial stage of Oral Health Care (OHC) action, was performed by compressing sterile gauze soaked in 0.9% NaCl (normal saline) until the crusts softened and could be peeled off naturally. This was followed by a sterile gauze compress soaked in 0.2% chlorhexidine gluconate solution. Subsequently, aloe vera extract gel was applied to the labial and perioral mucosa, and aloe vera extract spray was used for difficult-to-reach intraoral areas. This series of procedures was performed routinely 3 times a day and showed significant progress in the oral and perioral areas after 4 consecutive days of Oral Health Care (OHC) actions (Figure 2).

Topical therapy for areas other than the oral and perioral regions, as well as comprehensive systemic therapy consisting of antibiotics, corticosteroids, and anti-histamines, was coordinated by the internal medicine and Dermato-Venereology specialists to control the autoimmune disease and comorbidities. These medications included Albumin 25%, Fusidic Acid cream 20 mg/gram, Methylprednisolone 125 mg injection, Mebhydrolin napadisylate, Meropenem 1 gram, and Gentamicin sulfate 0.3% eye ointment.

Figure 2. Progress of the patient's oral and perioral lesions after 4 consecutive days of Oral Health Care (OHC) procedures.

DISCUSSION

Bullous Pemphigoid (BP) is the most common subepidermal bullous autoimmune disease, classically characterized by tense bullae and intense pruritus on the skin [1,2]. This

Denta Jurnal Kedokteran Gigi, Februari 2026; Vol.20 No.1; Hal 1-9
Available at http://journal-denta.hangtuah.ac.id/index.php/jurnal/issue/view/41
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case presents a significant diagnostic challenge due to its atypical clinical manifestations. The patient's presentation of extensive erosions and crusting, involving both the skin and oral mucosa, resembled epidermal sloughing. The widespread clinical features, combined with a history of prior treatment with Acyclovir (for suspected herpes simplex), made it visually very difficult to distinguish from Toxic Epidermal Necrolysis (TEN), a life-threatening drug hypersensitivity reaction, especially in the early phase.

The primary diagnostic challenge lies in differentiating BP from TEN. Clinically, TEN is characterized by a painful, dark erythematous rash followed by keratinocyte necrosis and total epidermal detachment [5]. Although atypical BP variants (BP-like TEN) can mimic this picture, a definite diagnosis cannot rely on clinical presentation alone. The definitive diagnosis is established through a combination of examinations. Several previous case reports have documented a similar phenomenon where Bullous Pemphigoid (BP) presents with massive epidermal sloughing resembling Toxic Epidermal Necrolysis (TEN). For comparison, a study by Patel et al.(2020) reported a case of BP that was initially diagnosed as TEN due to extensive mucosal involvement and a positive Nikolsky sign; however, the diagnosis of BP was only established after histopathological examination revealed a subepidermal cleft rather than the full-thickness necrosis characteristic of TEN. The similarities found in this case emphasize that while TEN is a primary medical emergency that must be ruled out, clinicians should not overlook the possibility of an autoimmune subepidermal bullous disease when the culture results or response to initial therapy do not align with the typical clinical course of TEN.

The gold standard supplementary examinations are histopathology-anatomy (HPA) and direct immunofluorescence (DIF). However, a tissue biopsy could not yet be performed on this patient because their haematological and albumin status had not reached the minimum acceptable level for the procedure, thus requiring albumin therapy and blood transfusion. If the patient's condition becomes suitable for a biopsy, the expected HPA result to strengthen the BP diagnosis is a subepidermal cleft with a rich eosinophil inflammatory cell infiltrate (typical of BP), which would rule out the full keratinocyte necrosis characteristic of TEN (4). Furthermore, the definitive finding expected from immunopathology is the linear deposition of IgG/C3 at the Basement Membrane Zone (BMZ) via Direct Immunofluorescence (DIF) and increased anti-BP180 autoantibody levels on ELISA, which reliably confirm the BP diagnosis [9].

This case is further complicated by clinical features resembling Bullous Impetigo. Bullous impetigo, caused by toxins from *Staphylococcus aureus*, produces superficial bullae that rupture and leave honey-colored crusts. The golden-yellow crusts observed in the patient's perioral and cutaneous areas were likely a manifestation of bacterial superinfection on the BP erosions. Therefore, urine, blood, and oral/perioral lesion swab cultures were also performed in this case [10,11].

Oral mucosal involvement in BP, although less common than in Pemphigus Vulgaris, is reported to occur in approximately 10–40% of cases. In this patient, the extensive and severe oral involvement caused trismus and significant pain, which is not only an indicator of disease severity but also a critical management challenge. This severe oral involvement significantly reduces the patient's quality of life. Comorbidity risk factors, such as Diabetes Mellitus (DM), also play an important role by increasing the patient's susceptibility to secondary infections and slowing down healing. Furthermore, the history of DM also necessitates evaluating for drug-induced BP, given that certain anti-diabetic medications (such as DPP-4 inhibitors) have been identified as significant BP triggers [2,8,12]. The role of Diabetes Mellitus as

Denta Jurnal Kedokteran Gigi, Februari 2026; Vol.20 No.1; Hal 1-9
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a comorbidity in this case warrants specific attention. Recent literature indicates a strong correlation between the use of dipeptidyl peptidase-4 inhibitors (DPP-4i) and an increased risk of BP. According to a meta-analysis by Kridin and Bergman (2019), patients with DM face a significant relative risk of developing drug-induced BP, which frequently manifests with more severe mucosal involvement compared to the classic variant. Comparison with the current case suggests that evaluating the patient's diabetic medication history is crucial, as discontinuing the triggering agent serves as a primary management step that often accelerates clinical remission more effectively than relying solely on immunosuppressants.

The use of systemic and topical medications in this atypical BP case was designed to achieve three main goals: controlling the autoimmune response, treating and preventing secondary infection, and providing vital patient support. Given the complexity of the clinical presentation, disease severity, and risk of infectious complications, this case justifies a comprehensive multidisciplinary management approach [12]. A medical team involving specialists in Dermato-Venereology, Internal Medicine, and Oral Medicine is highly necessary. Systemic management to control the autoimmune disease was coordinated by the Internal Medicine and Dermato-Venereology specialists. Methylprednisolone 125 mg injection was administered as a high-dose corticosteroid to suppress the abnormal immune response underlying BP. Corticosteroid use is the mainstay therapy for controlling autoimmune diseases [13]. Additionally, Albumin 25% was administered as supportive therapy. Albumin use was crucial because the patient's hematological and albumin status had not yet reached the minimum threshold for a biopsy, thus Albumin was given for stabilization and support before the definitive diagnostic procedure could be performed [14].

Infection control was a priority, considering the patient's clinical presentation resembled impetigo with golden-yellow (honey-like appearance) crusts in the perioral and cutaneous areas, suspected to be a bacterial superinfection of the BP erosions. Based on the bacterial culture results from the oral lesion swab, *Group-A Streptococcus (GAS)*, the etiology of impetigo, was not found. However, the culture test showed the presence of *Serratia marcescens*, a bacterium that is one of the causes of Ventilator-Associated Pneumonia (VAP). Meropenem is a broad-spectrum antibiotic used to treat severe systemic bacterial infections, including those causing pneumonia, which is a high risk in hospitalized patients with extensive skin lesions and comorbidities like Diabetes Mellitus. The choice of this antibiotic was reinforced by the patient's bacterial culture and antibiotic sensitivity test results. Meanwhile, Fusidic Acid cream 20 mg/gram served as a topical antibiotic for the treatment of local skin infections in areas other than the oral and perioral regions. The patient was given Mebhydrolin napadisylate and Gentamicin sulfate 0.3% eye ointment to manage symptoms and protect sensitive areas. Mebhydrolin napadisylate functions as an anti-histamine, part of the comprehensive systemic therapy to reduce the itching (pruritus) that usually accompanies BP.

Gentamicin sulfate 0.3% eye ointment, an antibiotic eye ointment, was used to prevent or manage potential infection and inflammation in the vulnerable eye area due to the widespread bullous disease. Integrated management between systemic therapy (autoimmune control) and local/symptomatic care (infection and pain control) is key to improving patient prognosis [15].

Specific oral management, including daily debridement and 0.2% Chlorhexidine gluconate irrigation for infection control, as well as the application of Aloe vera extract gel/spray for pain palliation and mucosal protection, is essential to support adequate nutritional intake. Aloe vera extract gel/spray forms a protective layer over the labial, perioral,

Denta Jurnal Kedokteran Gigi, Februari 2026; Vol.20 No.1; Hal 1-9
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and intraoral mucosal erosions and ulcerations. This protective layer serves to relieve pain and burning sensations. This combination of antiseptics and anti-inflammatory covering agents is key to creating an optimal healing environment and supporting the success of the primary systemic therapy for BP. Integrated management between systemic therapy (to control the autoimmune disease) and local care (to control infection and symptoms) is the key to improving patient prognosis [4,16].

Atypical Bullous Pemphigoid (BP) mimicking the features of TEN and impetigo poses a significant diagnostic challenge that cannot be overcome by clinical appearance alone. A multidisciplinary diagnostic algorithm supported by supplementary examinations is necessary to differentiate BP from its differential diagnoses. Therefore, integrated comprehensive management, involving specialists in Dermato-Venereology, Internal Medicine, and Oral Medicine, is crucial. The management of oral and perioral manifestations through OHC (Oral Health Care) procedures is a highly essential, integral component of the overall care to support patient recovery.

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Denta Jurnal Kedokteran Gigi, Februari 2026; Vol.20 No.1; Hal 1-9
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Denta Jurnal Kedokteran Gigi, Februari 2026; Vol.20 No.1; Hal 1-9
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