



Case Report: Brucella Parotitis with Abscess Formation and Paradoxical Reaction on Therapy

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Abstract: In this report we describe a patient with sub-acute bacteremic brucellosis complicated by Brucella parotitis with abscess formation as a paradoxical reaction on therapy. A 69 years old male known to have Warthin's tumor of parotid glands two years before his presentation. He presented with generalized ill-health, body-aches and feeling of further enlargement of his previous swellings at jaw angles for a few weeks. He admitted unpasteurized camels' milk ingestion. Brucellosis was suspected and proven by a positive blood culture. 10 days after starting anti-brucellosis therapy, he reported back with bilateral parotid abscesses. Patient was diagnosed as sub-acute bacteremic brucellosis with *brucella* parotitis and abscess formation. The interval enlargement of parotid swellings and abscess formation 10 days after initiation of therapy is explained by paradoxical inflammatory reaction following response to therapy.

Keywords: *Brucella* Parotitis, Paradoxical Reaction, *Brucella* Abscess

1. Introduction

Brucellosis is a zoonotic infectious disease with multisystem involvement. Human brucellosis has worldwide distribution. Saudi Arabia is considered as an endemic area where the prevalence of the disease is 15-19% [1].

Human brucellosis presents with various manifestations, onset might be acute, insidious (sub-acute) or chronic. Clinical presentation might be systemic or localized. Skeletal, gastrointestinal and hematologic complications occur commonly. Involvement of the neck was rarely reported, particularly the parotid glands [2].

Paradoxical reaction, defined as inflammatory response following initial improvement with appropriate antimicrobial treatment manifested as worsening of clinical or radiological findings in the absence of evidence of disease relapse or presence of other diagnosis, is well documented with human immunodeficiency virus/ tuberculosis co-infection [3]. It is rarely documented with infections other than tuberculosis in immunocompetent host [4-6]. In this report we describe brucellosis involving the parotid glands causing parotid abscess as a paradoxical reaction on therapy.

2. The Case

A 69 years old gentleman who presented to the emergency room complaining of generalized ill-health, weakness and body-aches for few weeks. He felt some enlargement of his bilateral angle of the jaw swellings that he had since 2 years before presentation. He denied any other symptoms e.g. fever, sweating, cough, anorexia, weight loss. He denied contact with sick people or animals but admitted unpasteurized camels' milk ingestion.

Past medical history: Bilateral parotid enlargement since two years before presentation, for which fine needle aspiration (FNA) revealed Warthin's tumor. Treated conservatively without surgical removal of the benign tumor. Patient denied previous brucellosis infection.

Social history: He resides at Riyadh capital of Saudi Arabia, used to work an office job, now retired, no recent travel.

Patient's examination was unremarkable apart from bilateral parotid swellings, without tenderness, lymphadenopathy or ear discharge, normal temperature.

Laboratory results showed: Normal complete blood count and hepatic profile, ESR 102 mm/hour CRP 52.3 mg/L. Blood culture positive *Brucella melitensis* (Requested in keeping with history of unpasteurized milk ingestion).

Brucella total Ab 1:20480 (Method used is SAT. Serum Agglutination Technique)

Brucella IgG Ab 1:20480

Patient was diagnosed as sub-acute bacteremic brucellosis, started on treatment (doxycycline 100 mg orally twice daily, trimethoprim-sulphamethoxazole 800/180 mg orally twice daily). Rifampicin was avoided in therapy to minimize resistance and save it for tuberculosis treatment since we are in endemic area of tuberculosis. Aminoglycosides were avoided to simplify the regimen and avoid bringing the patient frequently to hospital for injection.

10 days later patient presented with appreciation of interval increase of his jaw angles swellings more on the right side. Physical examination revealed bilateral parotid swellings: Right parotid 6x4 cm size hard, tender, non-fluctuant, intact skin. Left parotid 4x3 cm size hard swelling, no discharge at ear canal. Oral examination revealed no signs of inflammation at duct openings. Small enlargement of the right posterior cervical lymph nodes. (See figure 1)



Figure 1. Before Treatment.

Patient was arranged for computed tomography and fine needle aspiration (FNA) of parotids (See figure 2). It showed Interval development of multi-loculated collection at the

region of the right parotid extending medially, the largest transverse diameter is 5.5 cm as well as interval development of smaller collection on the left side with the largest transverse diameter of 1.2 cm, as compared to previous CT (see arrows).

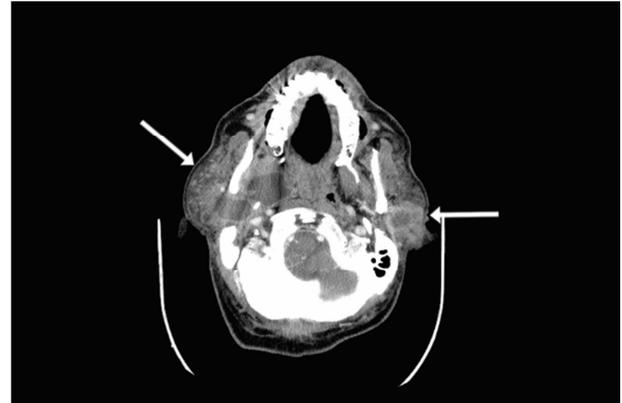


Figure 2. CT Neck.

FNA right parotid result showed negative stain and culture for bacterial, fungal and tuberculosis organisms, histology showed acute inflammation with macrophage and necrotic material consistent with abscess, no granuloma, no malignant cells.

Patient continued on anti-brucellosis therapy, showed improvement and excellent response. (See figure 3).



Figure 3. After Treatment.

Patient completed 3 months medical therapy, his symptoms resolved with clinical and radiological regression of his parotid swellings as shown by figures and follow up CT parotids done 1 month later (See figure 4). Patient cleared his bacteremia, and brucella IgG Ab titer reduced from 1:20480 upon presentation to 1:1280 in 3 months duration. Patient's regular follow up until one year after presentation, confirmed no relapse.

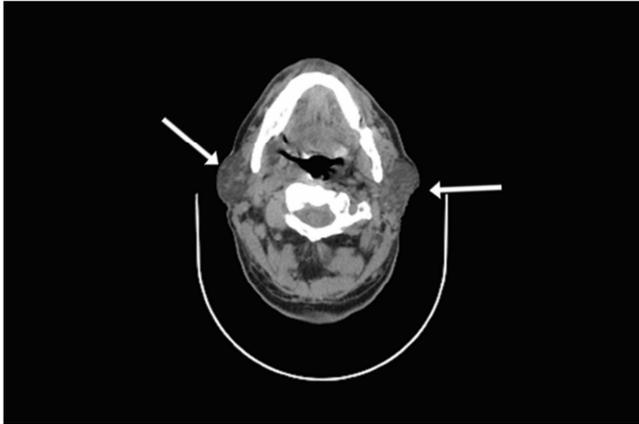


Figure 4. Follow-up CT Neck 1 month later.

3. Discussion

Brucellosis is well documented in the literature to affect multiple organs and tissues of the body e.g. neuro-brucellosis, meningoencephalitis, radiculitis [10-12]. Recurrent uveitis unresponsive to treatment with steroids found to have systemic brucellosis, *Brucella millitenses* isolated from paravertebral abscess, the condition responded to systemic antibiotics and completely recovered from her uveitis [13]. Epididymo-orchitis [14], cardiovascular: endo, myo and pericarditis [15, 16]. Peritonitis [17]. Ileitis [18], Hepatitis [19]. Cutaneous endophlebitis on the leg caused by brucella infection, maculopapular rash, panniculitis [20].

There are minimal reported cases of brucellosis involving head and neck, *Brucella millitenses* infection within Warthin's tumour of parotid gland [2], thyroid gland [7], cervical lymphadenopathy [8], and neck abscess [9].

In our patient, who is known of Warthin's tumor of the parotid, we presume hematogenous spread of his brucellosis bacteremia had led to development of parotitis. Excellent response to anti-brucellosis treatment proves this assumption. Surgical fine needle aspiration specimen didn't grow organism because our patient was already on treatment for 10 days.

Interval enlargement of parotid glands and abscess formation after 10 days of initiation of therapy, is explained as a paradoxical inflammatory reaction following response to therapy. Warthin's tumor (papillary cystadenoma lymphomatosum) is a common benign tumor of the parotid gland. It contributes to abscess formation by: 1. Compression caused by neoplasm may lead to stasis and this predisposes to infection. 2. The lymphoid element within Warthin's tumor may provide a site of tropism resembling normal lymphoid tissue where invading organisms can suppurate. 3. The cystic

nature of Warthin's tumor provides a fluid filled cavity which is a good milieu for abscess formation [2].

It faded away with continuation of therapy in our patient without addition of steroids. Paradoxical reaction has been well documented with tuberculosis in HIV-infected or non-infected cases, but more with HIV-infected cases [3]. Predictors of paradoxical reaction could be high bacterial host burden, low lymphocyte count, low haemoglobin, low albumin, C-reactive protein (CRP), erythrocyte sedimentation rate (ESR) but; are yet to be determined [3, 4]. Paradoxical reaction in immunocompetent host with infection other than tuberculosis was rarely described. A paradoxical reaction to treatment in two patients with severe acute paracoccidiomycosis treated with corticosteroids has been reported [4]. Paradoxical reaction during treatment of *Mycobacterium ulcerans* (Buruli ulcer) has been reported in 31 patients (19%) of 163 patients. Severe cases were treated successfully with steroids [5]. A paradoxical response preceding control of *Scedosporium apiospermum* mycetoma with posaconazole treatment without steroids has been reported [6]. The factors that trigger the paradoxical reaction are not yet known. It is presumed that these paradoxical reactions are a result of exaggerated response to persistent microbial antigens which manifest when disease related immunosuppressive mechanisms are blunted [21]. Paradoxical reactions, in infections other than tuberculosis in immunocompromised host, could provide pathway to informations on pathophysiology of the condition.

On reviewing the literature we couldn't find previous reporting of paradoxical reaction with brucellosis.

4. Conclusion

This is a case report of a patient with sub-acute bacteremic brucellosis complicated by Brucella parotitis with abscess formation as a paradoxical reaction on therapy.

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Conflict of Interest

The author has no conflicts of interest in the report, no support from any organization for the submitted work; no financial relationships with any organizations that might have an interest in the submitted work; no other relationships or activities that could appear to have influenced the submitted work.

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