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## CASE REPORT



# Nontyphoidal Salmonellosis Presented as a Parotid Abscess within a Warthin's Tumor: Case Report and Review of Literature

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Most suppurative parotid abscesses are caused by bacterial infection via the Stensen's duct. Formation of a unilateral parotid abscess secondary to *Salmonella* enterocolitis seldom occurs, and cases arising in the setting of a previous existing parotid tumor have rarely been described to date. This study presents a 71-year-old male patient having a parotid abscess. This was caused by *Salmonella enterica serovar Enteritidis* (*S. enteritidis*) in an apparently immunocompetent adult with an inherent parotid tumor. The pathology is consistent with a Warthin's tumor with infarction. By integrating surgical and antimicrobial drug therapy, the disease responded well to treatment. Despite an increasing incidence of extraintestinal salmonellosis in the past decades, a clinical practitioner must maintain a high index of suspicion for rare locations, such as the parotid gland, after uncomplicated bacteremia accompanying enterocolitis. Early diagnosis and prompt treatment are necessary for the prevention of lethal complications.

Key words: Parotid abscess, nontyphoidal Salmonella, Salmonella enteritidis, Warthin's tumor

#### INTRODUCTION

Among the major salivary glands, the parotid gland is much more susceptible, relative to the submandibular and sublingual glands, due to its serous secretion, which is devoid of antibodies, lysosomes, and mucins. The predisposing factors may include dehydration, debilitation, and sialoliths with obstruction, which are usually bacterial in nature and may be complicated with abscess formation. The common pathogens are Staphylococcus aureus, Klebsiella pneumoniae, Escherichia coli, Alpha-hemolytic streptococci, and anaerobic bacteria not otherwise specified.1 The efficient therapeutic strategies include surgical debridement plus adequate use of antibiotics. To the best of our knowledge, there are scanty reports about parotid nontyphoidal salmonellosis. Extraintestinal presentation within an inherent parotid tumor is extremely rare. Herein, a case is presented and the related English literature is reviewed.

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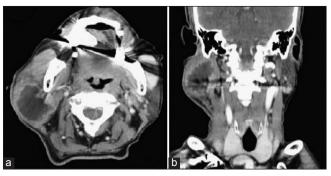
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## CASE REPORT

A 71-year-old male patient, working as a construction worker, had a history of a slow-growing right infra-auricular tumor mass, for 10 years. One week before admission he had an episode of non-bloody watery diarrhea, nausea, and vomiting that lasted for three days. Four days later, he complained of severe right cheek pain over an inherent tumor. His medical history is unremarkable for not taking corticosteroid, diabetes mellitus, autoimmune disease, malignancy, and a similar episode in this site. He had a recent ingestion of raw eggs. He denied contact history with birds, insects or reptiles. The physical examination showed protrusion of the right cheek, of size  $7 \times 6$  cm<sup>2</sup>, with central hyperemia and local heat. The oral cavity showed neither dilated papilla (soft tissue protuberance at the exit of the Stensen's duct) nor purulent substance from it. The facial nerve function was intact bilaterally.

The patient wanted a second opinion after taking oral antibiotics from an outside hospital. Fine-needle aspiration yielded 3 mL of purulent material. Laboratory tests demonstrated a white blood cell count of  $11650/\text{mm}^3$  (64% neutrophils, reference range  $4\times10^3/\text{mm}^3$  to  $8.5\times10^3/\text{mm}^3$ ) and C-reactive protein of 4.6 mg/L (normal reference less than 1 mg/L). Contrast-enhanced computed tomography [Figure 1] demonstrated an irregular ring-enhancing loculated lesion with a homogenous low-density substance inside, without evidence of sialolithiasis. Diagnosis was compatible with parotid

Parotid abscess due to Salmonella enteritidis in a patient with Warthin's tumor



**Figure 1**. Head and neck computed tomography (CT) with contrast, axial view (a), coronal view (b), disclosed an irregular ring-enhancing loculated lesion in the right retromandibular region, measuring  $5 \times 4 \times 4.5$  cm<sup>3</sup>, closely jostling the deep lobe of the right parotid gland

abscess with reactive neck lymphadenopathy. The radiological imaging differential showed a necrotic tumor.

Emergent operative drainage of the abscess was performed and the intraoperative specimen grew *Salmonella enterica serovar enteritidis* (*S. enteritidis*), serotype B. Histopathology illustrated Warthin's tumor with infarction [Figure 2].

The relevant serology investigation for human immunodeficiency virus (HIV), diabetes mellitus, and pertinent autoimmune antibodies were all negative. Stool culture also grew *S. enteritidis*, group B. Consultation was then sought with physicians from Infectious Diseases. The diagnosis was solitary parotid gland extraintestinal salmonellosis within Warthin's tumor, secondary to uncomplicated bacteremia accompanying enterocolitis. Ceftriaxone 2 g every eight hours was previously administered and was replaced with ciprofloxacin 400 mg twice daily for a total of 14 days. Second-stage debridement and wound closure was performed seven days later, with complete remission of symptoms. The postoperative course was smooth and the wound healed well without facial nerve injury, fistula formation or recurrent abscess formation.

#### DISCUSSION

Bacteria of the genus *Salmonella* are facultative anaerobic gram-negative bacilli, which can be divided into two broad categories according to the clinical syndromes in human subjects. One category consists of typhoid serotypes (*Salmonella typhi* and *Salmonella paratyphi*), which cause classic typhoid fever, and the other are made up of non-typhoid serotypes (non-typhoidal *Salmonella* or NTS) that can colonize the gastrointestinal tracts of a broad range of animals, and more than 200 non-typhoid serotypes are pathogenic to humans. The epidemiology of NTS has evolved over time. In the past 10 years, *S. Enteritidis* (65% of isolates), *S. Typhimurium* (12%), and *S. Newport* (4%)<sup>2</sup> are the most common serotypes worldwide.

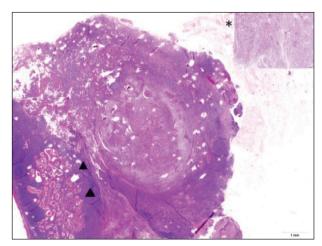


Figure 2. Histopathology shows a gross lesion containing an intact Warthin's tumor (▲) and localized necrotic debris (\*) (hematoxylin and eosin stain, magnification 2.5); inset, shows the architecture of preserved dead tissue composed of eosinophilic, anucleate cells (hematoxylin and eosin staining, magnification ×40)

Non-typhoidal *Salmonella* cause four clinical syndromes in humans including diarrhea disease, invasive bacteremic illness, focal suppurative infection, and asymptomatic carriage in the stool.<sup>3</sup> An individual may show several of these. The most common initial symptoms are self-limited nausea and non-bloody diarrhea, without associated intestinal perforation. In our patient, it is very likely that the infection was acquired seven days earlier after ingestion of raw eggs, accounting for gastrointestinal symptoms.

Although less than 5% of the infected patients developed disseminated bacteremia or localized suppurative foci, it was significantly more likely in a population of immunocompromised adults, including underlying severe disease, chronic granulomatous disease, defects or blockade of specific cytokines, and HIV. These patients have dramatically a more severe and invasive presentation, markedly increased mortality, and may suffer recurrence.<sup>4,5</sup> Common extraintestinal sites of infections include urinary tract, the endovascular structure, joints, long bone, and brain.5,6 Parotid abscess has been inadequately described in the past decades. However, the presence of a rapidly progressing infra-auricular firm mass, combined with the patient's ethnicity and contact history, should raise suspicion of a parotid NTS. The differential includes necrotic lymphadenopathy, infected salivary gland tumor or congenital cyst. Fluoroquinolones are a reasonable empiric antibiotic choice, while chloramphenicol, amoxicillin, and trimethoprim/sulfamethoxazole are reasonable alternatives. A 14-day course of therapy is likely adequate for otherwise healthy individuals, with uncomplicated bacteremia accompanying enterocolitis.7

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A search of the English literature published since 1950 revealed only eight cases of parotid salmonellosis [Table 1]. All the eight patients were male. The age rage was 15 to 79 years, with a mean of 55.9 years. All the cases had underlying diseases, except for one subject. <sup>14</sup> Three had HIV and one had an inherent parotid tumor.

Among the patients having HIV, there were two cases<sup>8,9,11</sup> of parotid abscess, complicated with HIV-associated lymphoepithelial cysts. The pathogenesis was attributed to the diffuse infiltrative CD8 lymphocytosis syndrome and salivary cystic degeneration. Thus, it was assumed that the most likely etiology was sialectasis, secondary to the prolonged history of a parotid tumor. In combination with high virulence and inoculums from the blood stream infection secondary to enterocolitis, *Salmonella spp.* elicited development of a devastating soft tissue. Subsequent exudates into the ductal parenchyma promoted further destruction and subsequent abscess formation.

Parotid salmonellosis is not necessarily rare, but easily neglected. The case is informative from a microbiological point of view, with regard to the parotid abscess. Thus clinicians, especially in the endemic regions, should consider salmonellosis as a potential contributing etiology for parotid abscess, which necessitates surgical intervention for both therapeutic drainage and diagnostic purpose.<sup>8-15</sup>

Table 1. Published literatures regarding parotid salmonellosis

References	Subtypes	Predisposing factors
J Assoc Physicians India 1984 <sup>8</sup>	Salmonella typhi	HIV, enteric fever
Otolaryngol Head Neck Surg 1992 <sup>9</sup>	Salmonella schwarzengrund	HIV, enterocolitis, parotid cystic lymphoepithelial disease
Clin Infect Dis. 1994 <sup>10</sup>	Salmonella enteritidis	Acute promyelocytic Leukemia/Staph parotitis
Clin Infect Dis. 1997 <sup>11</sup>	Salmonella heidelberg	HIV, parotid cystic lymphoepithelial disease
Am J Otolaryngol. 2002 <sup>12</sup>	Salmonella enteritidis	Parotid basal cell adenoma
Acta Cytol. 2006 <sup>13</sup>	Salmonella enteritidis	Chronic alcohol consumption, hematogenous spread
Int J Med Microbiol. 2007 <sup>14</sup>	Salmonella enteritidis	Immunocompetent
Am J Med Sci. 2008 <sup>15</sup>	Salmonella enteritidis	Diabetes mellitus, sepsis

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