

## A Rare Case of Skull Base Osteomyelitis Caused By *E.Coli* and *Candida Albicans* in a Patient with Gapo Syndrome: A Case Report

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Received date: January 31 2020; Accepted date: February 19, 2020; published date: February 24, 2020

Citation: Abdurrahman Kaya, Sibel Yıldız Kaya, Sena Alkan, Mustafa Alkan (2020) A Rare Case of Skull Base Osteomyelitis Caused By *E.Coli* and *Candida Albicans* in A Patient with Gapo Syndrome: A Case Report. J Clinical Research and Reports, 3(1); DOI:10.31579/2690-1919/033

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### Abstract:

Skull base osteomyelitis is a very rare and life-threatening disease. It has significant mortality and morbidity though appropriate antibiotherapy. Here we report a case of atypical skull base osteomyelitis, which is rarely seen in infectious disease practice, caused by *E.coli* and *Candida albicans* in a patient with GAPO syndrome.

**Key words:** skull base; osteomyelitis; gapo syndrome

### Introduction

Skull base osteomyelitis (SBO) is a very rare and life-threatening disease. It has significant mortality and morbidity although appropriate antibiotherapy [1]. SBO can be presented in two clinical forms: typical and atypical [2]. Diabetes mellitus, immunosuppression and advanced age are among the risk factors for typical presentation and it is easy to identify infectious focus and etiological agent in this clinical form. Otitis externa is the most usually encountered underlying infectious focus for typical form [3]. Atypical form is not associated with clearly defined ear and paranasal sinus infections and clinical symptoms have slower course compared to typical form. Identification of the infectious focus and causative microorganism is also difficult [3]. Here we report a case of atypical skull base osteomyelitis caused by *Escherichia coli* and *Candida albicans* in a patient with GAPO (growth retardation, alopecia, pseudoanodontia, optic atrophy) syndrome.

### Case:

A 33 years-old male patient was admitted to our clinic with earache, fever and headache lasting for about 3 months. His history revealed that he has been followed-up for GAPO syndrome since he was 2 years-old. On his physical examination bilateral periorbital swelling was present unabling the patient to open his eyelids. Previously, the patient had received multiple antibiotics (amoxicillin clavulanate, ciprofloxacin, cefuroxime axetil). Multiple calcified dilated vascular veins were detected on his scalp. Focal neurologic deficit was not determined. Paranasal sinus computerized tomography (CT) was normal, rheumatologic markers were in normal range. Body temperature was 38°C, pulse rate 100/min and arterial blood pressure 120/80 mmHg. Laboratory studies were as follows: leukocyte 22800 /mm<sup>3</sup> ((86% neutrophils), C-reactive protein 125 mg/L, and erythrocyte sedimentation rate (ESR) 133 mm/h. Cranial CT scan showed large destructive and erosive involvement of the anterior and middle skull base which revealed findings compatible with osteomyelitis in the skull base (Figure 1).



**Fig.1:** Cranial CT scan showed large destructive and erosive involvement of the anterior and middle skull base.

Due to pre-diagnosis of SBO empirical antibiotherapy was initiated with piperacilin-tazobactam (4,5g three times /daily) after obtaining blood and ear swab cultures. The cultures of blood and ear swabs remained sterile. As no clinical and laboratory response was obtained with antibiotherapy transnasal biopsy was performed to determine the etiology. Many leukocytes were seen (with neutrophilia predominance) in gram staining of the biopsy and histopathology was consistent with osteomyelitis. Culture of the sample yielded extended-spectrum beta-lactamase producing (ESBL) *Escherichia coli* and *Candida albicans*. The antibiotherapy was shifted to meropenem 3gr/daily and fluconazole 400 mg/daily according to the susceptibility results. The periorbital swelling was regressed under this combination therapy allowing the patient to open his eyelids. Fever was subsided on the 4<sup>th</sup> day of treatment and the parenteral therapy was continued for four weeks. Maintenance therapy with oral amoxicilline-clavulanate and fluconazole was planned for four months. Whenever the findings observed in cranial imaging regressed completely and C - reactive protein, ESR decreased to normal levels the therapy was stopped. After six months of cessation of therapy the patient is doing well with no recurrence on his follow-ups.

### Discussion:

Skull-base osteomyelitis is a very rare disease; with a prevalence of 1.5% in all osteomyelitis [4] occurring secondary to pyogenic and fungal infections [5] Although it is a very rare disease, it has high mortality rate (20–40%) [6]. Therefore, the early diagnosis and appropriate antibiotherapy is crucial in decreasing morbidity and mortality rates [4]. Many risk factors such as increasing age, diabetes mellitus and microvascular disease were described in the etiology of SBO [7, 8]. However, the index patient had no diagnosis of diabetes mellitus and otitis externa. In addition, we did not detect any infectious foci and the identification of the etiological agent was difficult (by means of

biopsy). Therefore we have considered the patient has atypical form of SBO.

GAPO syndrome is an autosomal recessive disease, characterized by many disorders. Impairment of facial bones vascularity and upper airway infections are also reported in this syndrome. [9,10]. Many predisposing factors were described for SBO such as trauma, surgical interventions, bacteremia, diseases impairing bone vascularisation and host defences [6]. Craniofacial vasculopathies with dilated calcifications present in our patient as a sign of GAPO syndrome has been considered as a related risk factor for SBO. Up to our knowledge, SBO was not reported in any of the 38 articles involving GAPO syndrome in English language literature; this is the first case report of a patient with GAPO syndrome developing SBO.

SBO may be pyogenic or fungal. *P. aeruginosa* is the predominant causative agent in pyogenic SBO [5]. Although *E. coli* is not among frequently encountered etiological agents, it was identified in our case. Fungal SBO is usually presented with sinonasal pain, facial or periorbital swelling and nasal stuffiness/discharge [5]. Periorbital swelling; a sign more frequently with fungal SBO was also a predominant clinical sign in our case. Fungal and polymicrobial SBO is very rare. This is also a first case report of polymicrobial SBO due to *E. coli* and *C. albicans*.

As a conclusion, SBO is a rare and life-threatening disease. The diagnosis of SBO can be very difficult as there is no pathognomonic sign and symptom and a delay in antibiotherapy might lead to high mortality and morbidity. GAPO syndrome may be predisposing factor due to craniofacial vasculopathies. Although rare fungi and resistant bacteria should be kept in mind in case of nonresponse to antibiotherapy.

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DOI: [10.31579/2690-1919/033](https://doi.org/10.31579/2690-1919/033)

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