

# Total Aplasia of the Paranasal Sinuses: A Rare Presentation

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## ABSTRACT

Paranasal sinuses are air-filled cavities located within the bones around the nasal cavity. Abnormalities and anatomical variations of the paranasal sinuses are commonly seen. Various degrees and combinations of paranasal sinus aplasia and hypoplasia cases have been reported. However, very few cases of total paranasal sinus aplasia have been reported to date. We herein describe a total paranasal sinus aplasia case detected incidentally in a 22-year-old male patient without any disease affecting the skeletal system.

Key words: Paranasal sinus, aplasia, hypoplasia, cone-beam computed tomography

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## INTRODUCTION

Paranasal sinuses are air-filled cavities located within the bones of the face and skull around the nasal cavity. They consist of the maxillary, frontal, and sphenoid sinuses, and the ethmoid air cells [1,2]. Paranasal sinuses develop as invaginations from the nasal fossae during the third and fourth fetal months. They undergo their major expansion after birth according to the development of the facial cranium and the teeth [3]. Although a variety of paranasal sinus anomalies have been reported in the literature, only few cases of total paranasal sinus aplasia have ever been reported [4-6].

We present a case of total paranasal sinus aplasia without any craniofacial anomaly or disease affecting the skeletal system, and the first ever case reported using cone-beam computed tomography (CBCT).

### **CASE REPORT**

A 22-year-old healthy male patient was referred from

orthodontics clinic for 3D cephalometric analysis and 3D evaluation of impacted teeth. He had no previous history of either facial trauma or systemic diseases affecting the skeletal system. The patient's previous panoramic and cephalometric radiographs were examined, and multiple impacted teeth were detected in both jaws. Moreover, there was a total lack of pneumatization in maxillary and frontal sinuses (Figures 1 and Figure 2). A CBCT (NewTom 5G; QR, Verona, Italy) scan was performed to determine the exact location of the impacted teeth and to confirm the aplasia of maxillary and frontal sinuses. Detailed examination of the CBCT images revealed that there were multiple impacted permanent teeth localized in different positions in the jaws. Further, there was a total lack of development or pneumatization of all paranasal sinuses (Figures 3-5). The patient was referred



Figure 1: Panoramic image shows total lack of pneumatization of bilateral maxillary sinuses.

to medical genetics, endocrinology, and hematology clinics to investigate if he had any associated diseases or syndromic anomalies. As a result of laboratory



Figure 2: Lateral cephalometric image shows the lack of pneumatization of the maxillary and frontal sinuses.



Figure 5: Sagittal sections of CBCT images show the lack of pneumatization of maxillary, sphenoid, and frontal sinuses.

examinations, no skeletal, systemic, or hematological abnormalities were detected in the patient.

#### DISCUSSION

Development of the paranasal sinuses begins as an evagination of the nasal mucosa in 3rd to 4th month of intrauterine life and their expansion continues after birth throughout early adulthood [3,7]. At birth maxillary sinus and ethmoid air cells are present, while the frontal and sphenoid sinuses develop later in childhood. The maxillary sinus is the first paranasal sinus to develop during human fetal life [8]. By the age of 4 the maxillary sinus extends laterally past the infraorbital canal and by the age of 9 it reaches the maxillary bone. At birth, ethmoid cells are more developed anteriorly and pneumatization progresses in a posterior direction. Their growth lasts until late puberty. From as early as 2 years old pneumatization of the sphenoidal sinuses can be detected and by the age of 14 they reach their final size. After the age of 2, the frontal sinus which is the last paranasal sinus that undergoes major expansion, starts growing and reaches its final size after puberty [9].

Paranasal sinus pathologies and anatomic variations can be evaluated using different imaging modalities. CBCT is an exciting technology in the field of oral and maxillofacial radiology but is generally accepted as one of the pioneering imaging techniques for paranasal sinus evaluation by dentists, maxillofacial radiologists, and otolaryngologists [10,11]. CBCT image evaluation for the nasal cavity and paranasal sinuses provides many advantages, such as low radiation doses compared with computed tomography and low cost compared with magnetic resonance imaging [11].

Having the largest volume within the paranasal sinuses, the maxillary sinuses have a critical importance in dentistry because of their close proximity to the maxillary premolar and molar teeth [12]. Especially during the



Figure 3: Axial sections of CBCT images show the lack of pneumatization of maxillary, sphenoid, and frontal sinuses.



Figure 4: Coronal sections of CBCT images show the lack of pneumatization of maxillary, frontal, and sphenoid sinuses and ethmoid air cells.

maxillofacial surgical procedures, osteotomies are performed on the lateral wall of the maxillary sinus in surgical interventions such as the Caldwell-Luc, Le Fort I osteotomy, and more often, in sinus floor elevation with the lateral window approach [13]. The maxillary sinuses are also prone to odontogenic infection, cyst and tumors of odontogenic origin [14].

Anatomical variations are commonly seen in the paranasal sinuses. Sphenoid sinus aplasia is a rare entity. The frequency of unilateral aplasia of frontal sinus was reported to be 15% and bilateral aplasia in 5% of adults [9]. While maxillary sinus hypoplasia is a well-known clinical entity, maxillary sinus aplasia is quite rare. In the literature, the prevalence of maxillary sinus hypoplasia was reported from 2.1% to 10.4% [15]. Total aplasia of paranasal sinuses is an extremely rare condition [6].

Although the causes of paranasal sinus anomalies are not fully understood, several other clinical syndromes are found to be associated with paranasal sinus hypoplasia and aplasia like Down's syndrome, Crouzon syndrome, Treacher-Collins syndrome, Apert syndrome, osteodysplasia (Melnick-Needles), craniosynostosis, Paget's disease, thalassemia, cystic fibrosis, and ciliary dyskinesia [4,16]. Pifferi et al. [17] reported a high prevalence of frontal and sphenoidal sinus aplasia or hypoplasia in patients with cystic fibrosis or primary ciliary dyskinesia. Therefore, in patients with paranasal sinus hypoplasia and aplasia, primary ciliary dyskinesia and cystic fibrosis should be considered and excluded [5]. The developmental pathologic abnormalities of the paranasal sinuses may be misdiagnosed as sinusitis or neoplasm. So, CT should be performed in suspicious cases [5,18].

There are few cases reported of total paranasal sinus aplasia in the literature. In a case presented by Celebi et al. [4] in 2012, total paranasal sinus aplasia was reported in a 35-year-old man whose complaints were fullness on the face and nasal obstruction. Korkmaz, et al. [5] reported a case of total paranasal sinus aplasia in a 57-year-old woman with complaints of nasal fullness and chronic headaches. Joshi et al. [6] reported a case of total paranasal sinus aplasia in a 15-¬year-¬old male with complaints of chronic frequent bilateral headache associated with facial fullness and frequent throat clearing habit. A case of total aplasia of the paranasal sinuses and nasal cavity in a 26-year-old male with complaints of nasal obstruction and mouth breathing since birth was presented by Hasan et al. [19]. Takaichi et al. [20] reported a case of total paranasal sinus aplasia with impacted teeth and no sinonasal symptoms. In our case, the patient was asymptomatic and total paranasal sinus aplasia was diagnosed incidentally as in the case presented by Takaichi et al. [20].

In conclusion, we present an extremely rare case with total paranasal sinus aplasia using CBCT. Awareness of the paranasal sinus anomalies is essential for an accurate surgical planning and to avoid complications during the procedure. Although CT is accepted as the gold standard in the evaluation of paranasal sinus pathologies and anatomical variations, CBCT can be preferred to CT for these purposes due to higher spatial resolution, lower patient dose, and lower cost.

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