Gardner syndrome with odontogenic sinusitis: A case report

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Abstract

Gardner syndrome (GS) with odontogenic sinusitis is rare but should be suspected in patients with multiple osteomas of the skull and facial bones, excess teeth, impacted teeth, and odontomas. Early diagnosis of GS and treatment for familial adenomatous polyposis may improve prognosis.

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RUNNING TITLE

Gardner syndrome with sinusitis

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CONFLICTS OF INTEREST

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STATEMENT OF INSTITUTIONAL REVIEW BOARD APPROVAL

Ethical approval for this case report was obtained from the institutional review board of Tokai University Hospital (approval number 20R-352). The study was carried out in accordance with the Code of Ethics of the World Medical Association (Helsinki Declaration). The institutional review board took responsibility for the anonymization of the patients, and the requirement for informed consent was waived.

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KEYWORDS

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INTRODUCTION

Gardner syndrome (GS) is an autosomal dominant genetic disease that is a subtype of familial adenomatous polyposis (FAP) with soft tissue tumors and osteomas[1]. If untreated, colorectal cancer will occur; therefore, early diagnosis is important. Gardner syndrome with sinusitis is rare. We report a case of GS with odontogenic sinusitis.

CASE REPORT

A 29-year-old male presented with a one-month history of left cheek pain and a two-week history of left cheek swelling. He had a history of diabetes and was receiving hypoglycemic medication. His mother died of gastric cancer, and his aunt had a history of total colectomy. An endoscopic examination revealed an elevated anterior nasal floor and closed inferior meatus bilaterally (Fig. 1). Mucopurulent discharge was noted in the left middle meatus.

Intraoral examination revealed multiple caries and tooth defects (Fig. 2). Computed tomography showed nodular osteosclerosis of the maxilla and mandible (Fig. 3a, b) and multiple impacted maxillary teeth. Soft tissue shadows and bone erosion were observed around the upper left impacted tooth. Additionally, the soft tissue shadow around the impacted tooth was continuous with the left maxillary sinus (Fig. 3c, d). Multiple osteomas were found in the ethmoid sinus, frontal sinus, and skull (Fig. 4). We diagnosed the patient with left odontogenic maxillary sinusitis.

GS was suspected due to the positive family history, multiple osteomas of the skull and facial bone, and abnormal teeth. Multiple adenomas were found in the stomach and colon (Fig. 5) on gastrointestinal endoscopy. Furthermore, genetic testing revealed a mutation in the adenomatous polyposis coli (*APC*) gene, leading to a diagnosis of GS. Sinusitis improved following tooth extraction and macrolide therapy for three months. Prophylactic colectomy has been planned for FAP.

DISCUSSION

We encountered a case of Gardner syndrome with odontogenic sinusitis. Many patients with GS are asymptomatic, and diagnosis is based on incidental findings or family history. Patients with GS may exhibit tooth abnormalities such as excess teeth, impacted teeth (delayed tooth eruption), and odontoma, in addition to osteoma[2]. Odontoma is usually asymptomatic; however, reports of infected odontoma exist[3]. GS with odontogenic sinusitis is rare. As far as we are aware, there are no similar case reports. In our patient, two upper left maxillary teeth formed an odontoma, which got infected, leading to odontogenic sinusitis.

Osteoma occurs in 46%–93% of patients with GS, which is 4–20 times more frequent than in the general population. It most commonly occurs at the outer cortex of the skull, mandible, and frontal sinus[2]. While a single osteoma might be accidently identified in clinical practice settings, GS should be suspected if three or more osteomas are found[2]. Treatment is not indicated for osteoma, but surgery may be required for mandibular dyskinesia or cosmesis[2].

If left untreated, patients with FAP will develop colorectal cancer[4], which is the leading cause of death in this population[5]. Prophylactic colectomy is a reliable treatment; however, surgery is recommended for patients in their 20s. In GS, because of the mutation in *APC* , which is a tumor-suppressor gene, complications such as gastric cancer, duodenal cancer, thyroid cancer, and malignant dental tumors may occur, in addition to colon cancer. Desmoid tumors also greatly influence the prognosis[4,5]. Therefore, full-body examination and long-term follow-up are required.

An otolaryngologist may see a patient with GS presenting nasal and buccal symptoms such as sinusitis and a dental infection. GS should be suspected if characteristic findings—including multiple osteomas of the skull and facial bones, excess teeth, impacted teeth, and odontomas—are observed. Eliciting a family history and examining the whole body, including the colon, can contribute to early diagnosis and treatment, potentially improving the patient's prognosis.

CONCLUSION

GS with odontogenic sinusitis is rare but should be suspected in patients with multiple osteomas of the skull and facial bones, excess teeth, impacted teeth, and odontomas. Early diagnosis and treatment of GS may improve prognosis. Further case studies are needed to determine the optimal strategy for treating patients with GS and odontogenic sinusitis**.**

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FIGURE LEGENDS

Figure 1.

Initial endoscopic examination image shows an elevated anterior nasal floor (yellow triangles) and a closed inferior meatus (red arrows) bilaterally.

(IT: inferior turbinate, NS: nasal septum)

Figure 2.

Intraoral examination image shows multiple caries (purple arrows) and tooth defects (yellow arrow).

Figure 3.

Initial computed tomography image shows nodular osteosclerosis of the maxilla and mandible (a, b; red arrows), multiple impacted maxillary teeth (c, d; yellow arrows), soft tissue shadows (*), and bone erosion around the upper left impacted tooth. The soft tissue shadow (*) around the impacted tooth is continuous with the left maxillary sinus (c, d).

Figure 4.

Initial computed tomography image shows multiple osteomas in the ethmoid sinus (a), frontal sinus (b), and skull (c).

Figure 5.

Upper (a) and lower (b) gastrointestinal endoscopy images show multiple adenomas in the stomach and colon.