

How Rare Is an Oral Presentation of Myeloid Sarcoma in the Infant?

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Myeloid sarcoma, or chloroma, is a malignancy consisting of immature, extramedullary leukemic cells of the granulocytic lineage that was first described by Burns¹ in 1811. In 1853, King² coined the term “chloroma,” because the lesion commonly, but not always, develops a characteristic green color when exposed to air. Dock³ was the first to associate this tumor with the presence of acute leukemia. The tumor is composed of granulocytic immature cells, resembling sarcoma, and it infiltrates into the adjacent tissue and effaces the architecture. As such, myeloid sarcoma is a more appropriate term than chloroma.³⁻⁵ Myeloid sarcoma can present in virtually any organ.⁶ Approximately 50% of the cases will present in the skin and subcutaneous tissue.⁷ Less commonly, these lesions present in bone, with the incidence in the skeleton estimated at 10%.⁷ The initial presentation of myeloid sarcoma in the oral cavity is exceedingly rare, although a number of cases have been reported (Table 1).⁸⁻⁷⁰ The more recent reports and case reviews of Pau et al⁵² in 2010, Zhou et al⁵⁸ in 2013, and Kumar et al⁷⁰ in 2017 estimated that 88 previous cases of oral MS have been reported. To the best of our knowledge, our case is the 89th case reported and the youngest patient reported to date (Table 1).

Case Report

An 11-month-old female infant with a noncontributory medical history presented to the Seattle

Children’s Hospital oral and maxillofacial surgery (OMS) clinic in late March 2017 with acute right mid-facial swelling. The parents stated they had noted their child to display discomfort associated with her right maxillary tooth 8 days previously. She quickly developed an intraoral swelling, in addition to an infected right eye, with the result that her parents took her to their local emergency department. The working diagnosis was conjunctivitis, and a course of clindamycin was prescribed. Because the swelling continued, the parents returned to their pediatrician 3 days later. The pediatrician referred them back to the emergency department for attempted bedside aspiration of the swollen right cheek. A computed tomography (CT) scan was obtained, and the patient was referred to the OMS clinic. On further questioning, the parents reported their child was not feeding well and appeared to be in pain. The parents also reported a color change in the child’s right iris.

The clinical examination confirmed right midfacial swelling that had obliterated the nasolabial angle (Figs 1, 2). No lymphadenopathy was present. The overlying midfacial skin ecchymosis was attributed to the previous emergency department attempt at aspiration. Intraorally, vestibular swelling was present with no active drainage (Fig 3). The right maxillary deciduous canine was mobile.

CT imaging demonstrated a circular radiolucency within the right maxilla measuring 2.2 cm at its greatest

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Table 1. MYELOID SARCOMAS ASSOCIATED WITH ORAL CAVITY: PREVIOUSLY REPORTED CASES

Year	Investigators	Cases (n)
1980-1990	Neiman et al; Hansen et al; Conran et al; Takagi et al; Castella et al; Reichart et al; Welch et al; Dreizen et al; Saleh et al; Ficarra et al; Alessi et al; Barker and Sloan; De Vicente Rodriguez et al; Timmis et al; Cho et al	15
1991-2000	Eisenberg et al; Stack and Ridley; Ritter et al; Roth et al; Tuset et al; Wiernik and Serpik; Lynch et al; Menasce et al; Tong and Lam; Tomás Carmona et al	13
2001-2010	Lee et al; Jordan et al; Amin et al; Asna et al; Antmen et al; Stoopler et al; Goteri et al; Colella et al; Koudstaal et al; Yinjun et al; Yoon et al; Matsushita et al; Xie et al; Srinivasan et al; Mohamedbhai et al; Lu et al; King Kim et al; Tuntiwong et al; Osterne et al; da Silva-Santos et al; Fasanmade et al; Papamantinos et al; Pau et al	25
2011-2017	Dym and Movahed; Ahmad J et al; Colovic et al; Seema et al; Mei et al; Zhou et al; Guastafierro et al; Niscola et al; Kurdoglu et al; Moshref et al; Yap et al; Wang et al; Sharma et al; Ponnamm et al; Yuhong et al; Dineshkumar et al; Jin et al; Kumar P et al	35

The patient characteristics of the cases reported included age range 1 to 89 years; both male and female subjects; tumor locations included palate, maxilla, mandible, lips, tongue, gingiva, cheek, tooth roots, and implant sites. Among the articles reviewed, most reported only 1 case.

A PubMed search was performed using “granulocytic sarcoma,” “chloroma,” and “myeloid sarcoma” located in the oral region; studies in the English language or translated to English were included; abstracts and selected studies in their entirety were manually searched for pertinent patient descriptive variables of interest. A total of 88 other cases were reported specific to the oral cavity; these cases did not include extraoral head and neck sites, such as the nasopharynx and tonsil, which were included in other case reports, such as by those Zhou et al and Cheng et al.

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dimension, distorted facial structures, maxillary sinus involvement, and displaced dentition (Figs 4, 5). The referring hospital’s radiology report gave the differential



FIGURE 1. Extraoral photograph during clinical examination showing right midfacial swelling and flattening of the right nasolabial angle. The skin ecchymosis was attributed to the emergency department aspiration performed by the referring institution.

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diagnosis as odontogenic and nonodontogenic lesions, including, but not limited to, aneurysmal bone cyst, odontogenic keratocyst, cystic ameloblastoma, or, less likely, a dentigerous cyst. At the child’s evaluation at Seattle Children’s Hospital, however, the acute nature of the presentation and radiographic findings, including extensive bone destruction and dental follicle damage, were consistent, suggesting a more malignant process, including lymphoma and rhabdomyosarcoma. The next day, in the operating room, the surgeon performed a right maxillary intraoral biopsy and extraction of the maxillary right deciduous canine. The specimen was gelatinous, had green components, and measured 2 cm in the greatest dimension (Fig 6). The wound was packed with an absorbable gelatin sponge and closed with polyglactin suture, and the patient was admitted to the hospital for observation.



FIGURE 2. Extraoral photograph, bird’s eye view, showing severe right midfacial asymmetry, swelling, and distortion of the right alar base.

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