

# Intraosseous inflammatory pseudotumor of the maxilla: A case report

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Ali Al Khader<sup>1</sup> , Hadeel Odeh<sup>1</sup>, Esra Nsour<sup>2</sup>   
and Abdel-Ghani Ibrahim Al Awamleh<sup>3</sup>

## Abstract

Inflammatory pseudotumors are benign lesions that are rarely reported in the head and neck. Moreover, intraosseous inflammatory pseudotumors of the maxilla are extremely rare, with less than 10 cases reported. A 52-year-old woman presented with recurrent dental infections; computed tomography scan of the osteolytic maxillary bone lesion and incisional biopsy were performed. Histopathological examination revealed that the lesion was composed of fascicles of fibroblasts and myofibroblasts, in addition to sheets of plasma cells, lymphocytes, and occasional other inflammatory cells. An infiltrative growth pattern was observed. Immunohistochemical staining confirmed an inflammatory pseudotumor. A partial maxillectomy was performed. There was no evidence of recurrence during the 4-month follow-up period. Inflammatory pseudotumors should be considered when treating destructive maxillary lesions. Immunohistochemical staining was performed to confirm polyclonal plasma cell proliferation.

## Keywords

Inflammatory myofibroblastic tumor, jaw, maxilla, plasma cell granuloma

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## Introduction

Inflammatory pseudotumors (IPTs) can occur at any age. However, they mainly affect children and young adults (primarily in the first three decades of life), with a mean age of 10 years and median of 9 years. These tumors affect females slightly more than males.<sup>1</sup> Macroscopically, these tumors are typically well circumscribed and nodular with a whorled, tan, myxoid, or fleshy cut surface. Calcification, necrosis, or hemorrhage can be seen.<sup>1</sup>

An IPT is a benign lesion mainly composed of myofibroblastic/fibroblastic cells admixed with variable proportions of plasma cells, lymphocytes, and eosinophils, with a morphology that can be regarded on a spectrum from prominent spindle cell to plasmacytoma-like proliferative lesion.<sup>1</sup> Different names have been given to this curious entity, such as plasma cell granuloma (PCG), inflammatory myofibroblastic tumor, benign myofibroblastoma, xanthomatous granuloma, histiocytoma, spindle cell pseudotumor, pseudosarcomatous myofibroblastic proliferation, or even inflammatory fibrosarcoma.<sup>2</sup>

IPTs can occur at any site in the body. However, they typically arise in the abdominopelvic soft tissues, followed by the lungs. They have rarely been reported in the head or neck

region. Moreover, intraosseous IPTs of the maxilla are extremely rare, with less than 10 cases reported.<sup>3</sup> IPT tumor is considered of an intermediate grade with a risk for recurrence. This case involved a 52-year-old woman with an intraosseous maxillary IPT, which mimicked a malignant tumor at radiological and histopathological levels, representing a diagnostic pitfall.

## Case report

This case involved a 52-year-old woman with a history of recurrent dental infections with transient improvement by antibiotics. However, the symptoms became refractory to antibiotic treatment, and the patient underwent further

<sup>1</sup>Faculty of Medicine, Al-Balqa Applied University, Al-Salt, Jordan

<sup>2</sup>Department of Pathology, Al Hussein Salt Hospital, Ministry of Health, Al-Salt, Jordan

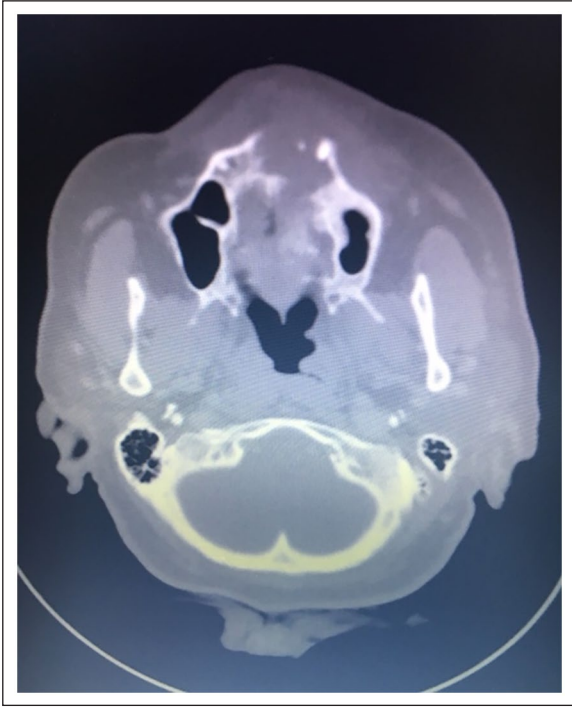
<sup>3</sup>Department of Maxillofacial Surgery, Al Hussein Salt Hospital, Ministry of Health, Al-Salt, Jordan

## Corresponding Author:

Ali Al Khader, Faculty of Medicine, Al-Balqa Applied University, Al-Salt, Jordan.

Email: ali.alkhader@bau.edu.jo



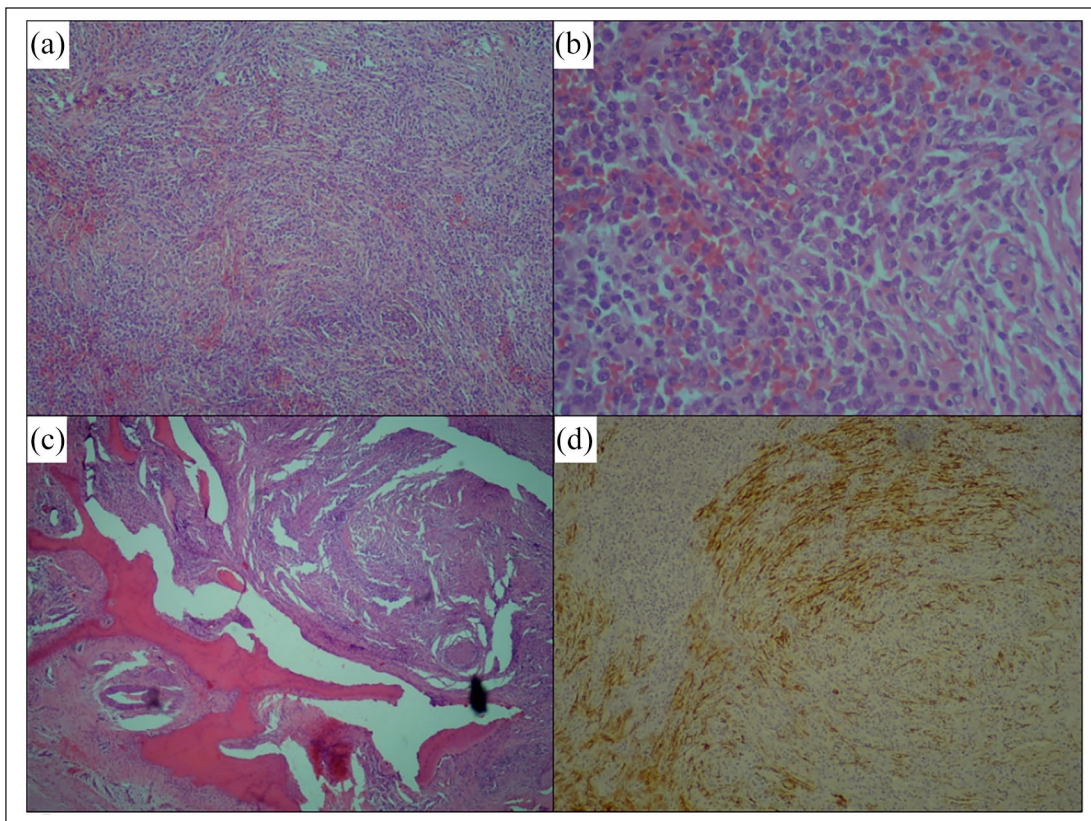


**Figure 1.** Computed tomography (CT) scan showing an osteolytic maxillary lesion located anterior to the inferior aspect of the left maxillary sinus.

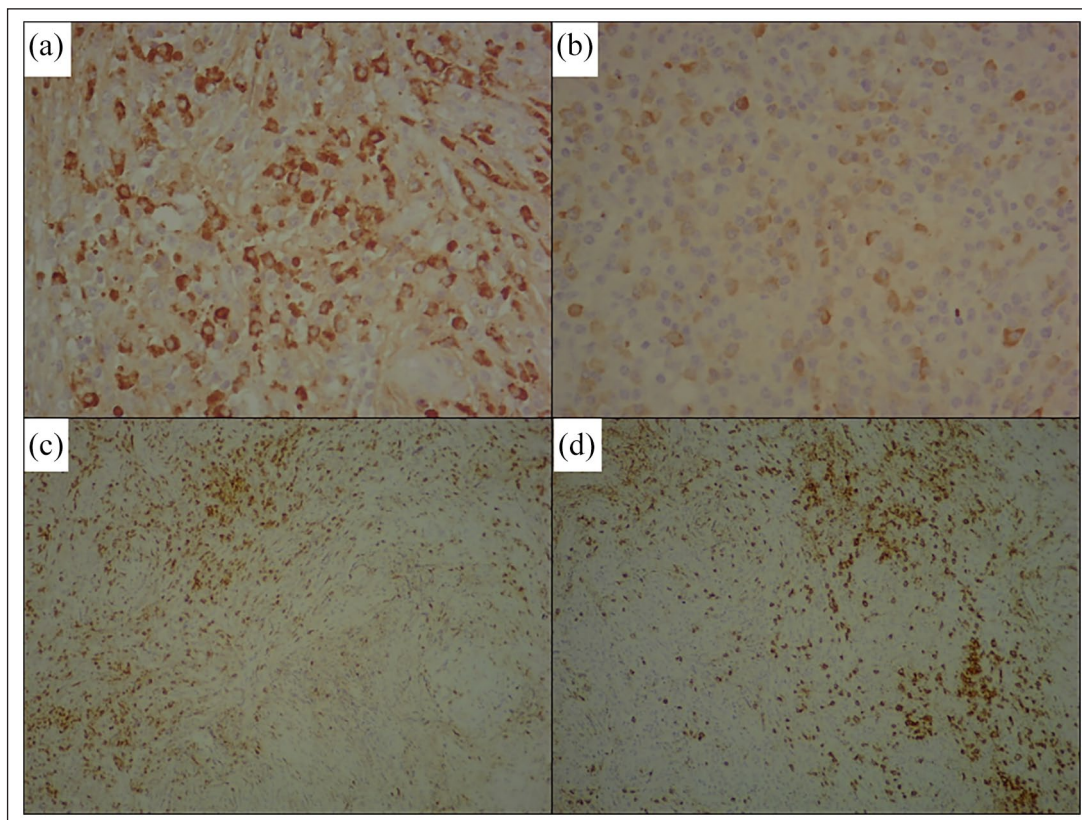
investigation. Computed tomography (CT) scan revealed a 3-cm-sized osteolytic maxillary lesion located anterior to the inferior aspect of the left maxillary sinus (Figure 1). An incisional biopsy was performed, and histopathological examination revealed a cellular lesion comprising spindle cell fascicles in a background of fibrotic stroma and areas akin to a storiform pattern. Sheets of plasma cells and lymphocytes were identified. Infiltration between the bone trabeculae was also identified. Immunohistochemical staining revealed that the spindle cells were positive for smooth muscle actin (Figure 2). Polyclonal proliferation of plasma cells was confirmed by kappa and lambda staining. CD3 and CD20 stainings revealed a mixed-lymphocyte population (Figure 3). In addition to the infiltrative pattern identified microscopically, CT and intraoperative findings also showed infiltrative tumor behavior. The patient underwent partial maxillectomy (Figure 4), and there was no evidence of recurrence during the 4-month follow-up period.

## Discussion

Based on histomorphological appearance, IPTs have been given different names, one of which is PCG.<sup>1</sup> This tumor is rarely reported in the head, with less than 5% of the cases reported at this site.<sup>2</sup> It has been reported in areas such as the sinuses, parapharyngeal area, and orbit, in addition to



**Figure 2.** Cellular lesion with vague storiform pattern, hematoxylin and eosin, 100 $\times$  (a). Sheets of plasma cells are characteristic, hematoxylin and eosin, 400 $\times$  (b). The tumor infiltrates in between the bone trabeculae, hematoxylin and eosin, 100 $\times$  (c). The spindle cells are positive for SMA immunohistochemical stain (d).



**Figure 3.** Polyclonal kappa-positive (a) and lambda-positive (b) plasma cell population. The lymphocytes are of mixed CD3-positive (c) and CD20-positive (d) cell population.



**Figure 4.** The partial maxillectomy specimen.

salivary and thyroid glands. IPTs have been reported in the maxillary sinus.<sup>4,5</sup> However, the tumor in this case was found in the maxillary bone, abutting but not involving the maxillary sinus. As a soft tissue tumor, IPTs can cause bone infiltration and destruction with a behavior highly suspicious for a malignant neoplasm.<sup>2,4</sup> The tumor usually occurs in young adults.<sup>5</sup> Morphologically, the sheeting of plasma cells, in

addition to the infiltrative pattern of growth, mimics plasmacytoma. Moreover, the highly reactive myofibroblasts also required pancytokeratin in our case to exclude carcinomas with single-cell patterns such as lobular carcinoma metastasizing to the bone, especially in view of the patient's sex and the reactive fibrotic background.

Microscopically, IPTs can be categorized into plasma cell-rich PCG, spindle cell-rich (inflammatory myofibroblastic tumor) type, and macrophage/xanthomatous cell-rich (fibrohistiocytic) type.<sup>6</sup> Accordingly, the semi-equal proportions of plasma cells and spindle cells in the tumor in this case made IPT the preferred term for reporting the case.

Intraoral and periodontal IPT cases have rarely been reported. Such lesions may mimic pyogenic granuloma clinically.<sup>7</sup> In addition, relatively few cases of IPTs have been reported in the temporal and frontal bones and skull base.<sup>8,9</sup> The presentation as a bony lesion has further complicated the diagnosis and made IPT a radiologically and histopathologically serious pitfall. IPT is considered a reactive lesion with no potential for malignancy.<sup>10</sup> There is no clear pathogenetic mechanism for IPT, and chronic antigen/allergen exposure has been proposed. The orbit is the most common involvement site in the head.<sup>7</sup>

Intraosseous IPT of the jaw is an extremely rare condition. In a recent report by Neronov et al., 25 cases of intraosseous

IPT of the jaw were reported, with the mandible being the most affected. After an extensive literature search, less than 10 cases of maxillary IPT were found,<sup>3,11,12</sup> all of which were osteolytic. Two cases were on the right side, and seven were on the left. Seven of the patients were female, with an age range of 11 to 75 years. The size of the lesion was known in four cases, ranging from 1 to 7 cm. As almost all reported cases of IPT of the jaw presented as central unilocular expansile radiolucent lesions on radiological examination, the differential diagnosis included odontogenic cysts, odontogenic tumors (e.g. unicystic ameloblastoma, odontogenic keratocyst, and ameloblastic fibroma), giant cell granulomas, and, sometimes especially with aggressive behavior, malignancy.<sup>3</sup> The histomorphological appearances mimicked different benign and malignant fibrous lesions, such as nodular fasciitis, desmoplastic fibroma, myofibroma, low-grade fibromyxoid sarcoma, myxofibrosarcoma, myofibrosarcoma, follicular dendritic cell sarcoma, and plasmacytoma.<sup>3</sup> Surgery was the treatment of choice for all cases, and steroids were used in one case. For aggressive cases, radiotherapy can be applied, and the follow-up period ranges from 6 to 72 months with no evidence of recurrence. This pointed to the benign nature of this lesion despite being a serious pitfall with regard to its possible radiological and morphological mimicry with malignant tumors.

## Conclusion

This report adds to the very small number of documented intraosseous IPTs of the jaw, especially the maxilla. The tumor in this case showed radiological and histomorphological mimicry with malignant tumors, similar to that of most documented cases, representing a diagnostic pitfall. Immunohistochemical staining to confirm polyclonal plasma cell proliferation is required in this scenario.

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## Ethical approval


Our institution does not require ethical approval for reporting individual cases or case series.

## Informed consent

Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

## ORCID iDs

Ali Al Khader  <https://orcid.org/0000-0001-8453-3629>

Esra Nsour  <https://orcid.org/0000-0002-7559-5032>

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