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Primary intraosseous carcinoma of the mandible

KEYWORDS
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Primary intraosseous carcinoma (PIOC) of the jaw is a rare malignant tumor with poor prognosis. PIOC is assumed to derive from the odontogenic epithelium and does not have connection with overlying tissues.[1]

A 67-year-old man was referred from a local dental clinic to our clinic for treatment of a swelling mass at the right mandibular second molar extraction wound. Based on the past dental history, the patient had a trauma to the right mandibular posterior teeth. The right mandibular second molar became hypermobile, and thus, he extracted the tooth by himself two weeks ago. Intraoral examination revealed an unhealed, reddish-white wound measuring 10×15 mm at the extraction socket wound of the right mandibular second molar. Extraoral examination showed a mild swelling over the right posterior cheek without local heat. Panoramic radiography revealed a unilocular radiolucency with scalloped borders extending from the distal side of the mandibular second premolar to the ascending ramus including the horizontal impacted right mandibular third molar (Fig. 1A). En bloc resection including the right mandibular segmental resection and right supraomohyoid neck dissection was performed. A reconstruction plate was placed to reconstruct the resected mandibular body. Because of the highly malignant potential of the tumor, postoperative radiotherapy and chemotherapy were performed. Microscopic examination revealed an epithelial neoplasm with squamous differentiation and extensive intrabony destruction. The tumor cells were arranged in trabeculae and islands and showed nuclear hyperchromatism and pleomorphism, abnormal mitotic figures, and an increased nuclear to cytoplasmic ratio. The multifocal areas of necrosis were also noted (Fig. 1B and C). However, there was no keratinization of the tumor cells. The surface epithelium was normal, and no transition of the surface epithelium to the underlying cancer cells was noted. The cancer cells were positive for CK (AE1/AE3) (Fig. 1D), indicating the tumor was a carcinoma. Because the tumor was a poorly-differentiated carcinoma, several immunostains using a panel of different antibodies were performed for further exploration of the origin of the tumor cells. We found that the cancer cells were positive for CK7 (a marker of primary lung carcinoma) (Fig. 1E), CK19 (a marker of odontogenic epithelium) (Fig. 1F), and p40 (p40 is highly specific for squamous and basal cells and is superior to p63 for diagnosing lung squamous cell carcinoma) (Fig. 1G), focally positive for CK20 (a marker of colonic adenocarcinoma) (Fig. 1H), and negative for CD56 (a marker of natural killer cell or Merkel cell carcinoma, and other cells including alpha beta T cells, gamma delta T cells, dendritic cells, and monocytes) (Fig. 1I) and thyroid transcription factor-1 (TTF-1, a marker of lung adenocarcinoma and small-cell lung cancer) (Fig. 1J). A high-grade central mucoepidermoid carcinoma was also included in our differential diagnosis for this tumor. However, no intracytoplasmic mucin was identified by mucicarmine stain and the fluorescence in situ hybridization (FISH) analysis for Mastermind-like 2 (MAML2) gene was negative, indicating that the tumor was not a mucoepidermoid carcinoma. Based on aforementioned immunostaining findings, a PIOC not otherwise specified (NOS) was diagnosed.

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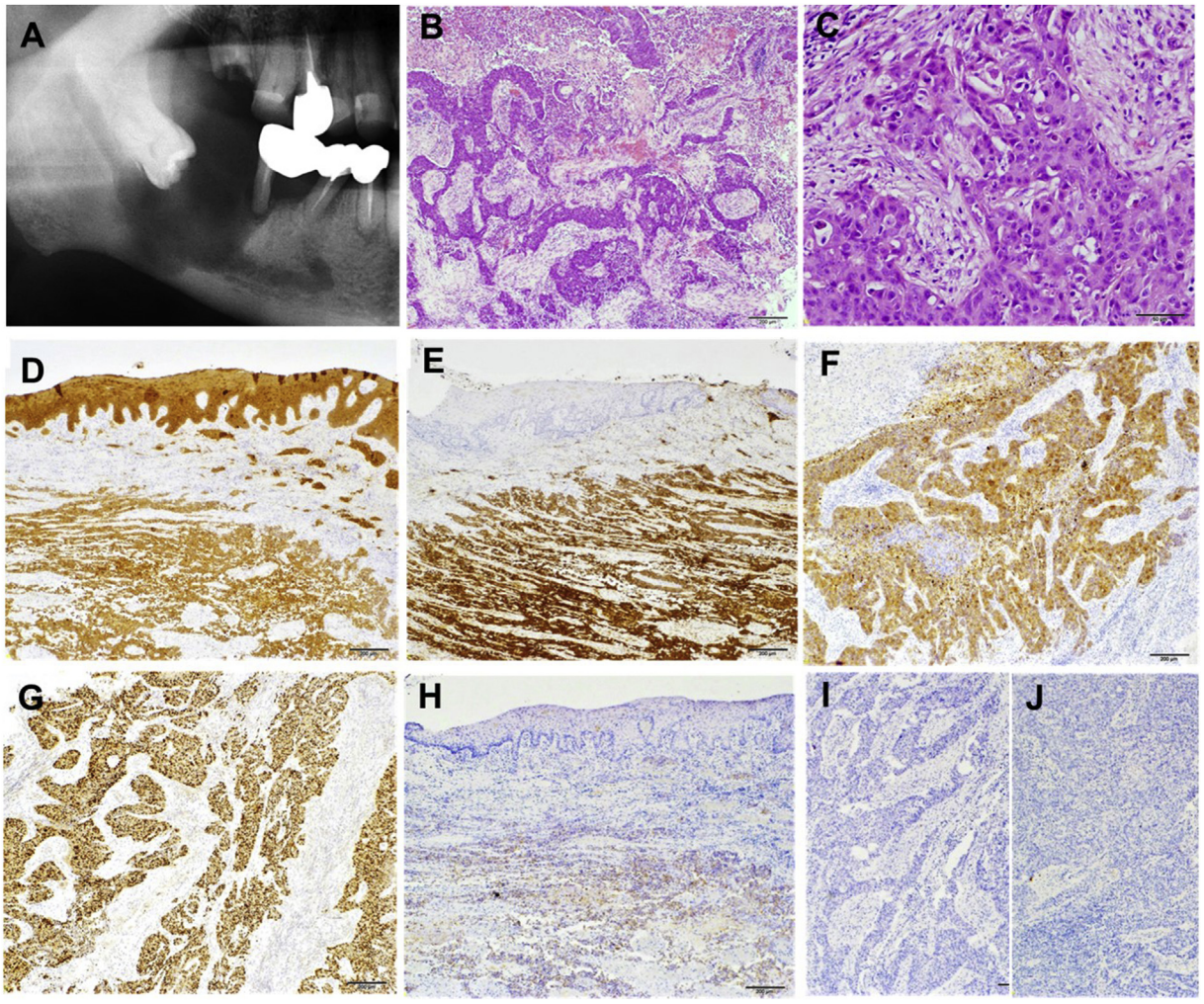

Figure 1 Radiographic photograph as well as histological and immunostained microphotographs of our case of intraosseous carcinoma. (A) Cropped panoramic radiograph showing a poorly-defined and non-corticated radiolucent lesion from the distal side of the right mandibular second premolar to the ascending ramus. (B) The tumor cells were arranged in trabeculae and islands infiltrating in the fibrous stroma with multifocal areas of tumoral necrosis (hematoxylin and eosin stain, original magnification 4×). (C) The tumor cells revealed nuclear hyperchromatism and pleomorphism, abnormal mitotic figures, and an increased nuclear to cytoplasmic ratio (hematoxylin and eosin stain, original magnification 20×). Immunohistochemical stains showed that the tumor cells were positive for CK (AE1/AE3) (D), CK7 (E), CK19 (F), and p40 (G); focally positive for CK20 (H); and negative for CD56 (I) and TTF-1 (J) (D to J, immunohistochemical stain; D to J, original magnification 4×).

PIOC is an aggressive malignant tumor, and its etiology remains unclear. PIOC is found more frequently in men than in women as well as in the posterior mandibular body and ascending ramus than in the maxilla.[1] Because the PIOC is a poorly-differentiated carcinoma. It usually needs the help of immunostains for confirmation of the tumor cell origin.[2–5] Therefore, immunostains using a panel of different antibodies were used to identify the tumor cell origin. The tumor cells of our case of PIOC were positive for CK7, CK19, and p40, focally positive for CK20, and negative for CD56 and TTF-1. Because a combination of TTF-1+/CK7+/CK20− was highly associated with a primary adenocarcinoma of lung, a combination of TTF-1-/CK7-/CK20 + was highly associated with an adenocarcinoma of gastrointestinal origin, and our tumor was positive for CK19 which was a marker of odontogenic epithelium, finally our tumor was diagnosed as a PIOC rather than a metastatic lung adenocarcinoma or a metastatic gastrointestinal adenocarcinoma.[5] The 5-year survival rate of PIOC was less than 40%. Due to the high recurrence rate and mortality, PIOC should be aggressively treated by radical surgery. In addition, radiotherapy, chemotherapy, and concurrent chemoradiation therapy are highly suggested for treatment of PIOC after surgical resection of the tumor.

Declaration of Interest

We have no conflicts of interest.

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