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Concealed gingival cancer mimicking necrotizing fasciitis in the head and neck region: A case report and review of diagnostic challenges

KEYWORDS
Malignancy;
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Cellulitis;
Infection

Severe head and neck infections frequently occur in the oral and maxillofacial regions, and malignancy can serve as a potential underlying cause. Therefore, severe infection symptoms can mask the clinical manifestations of underlying malignant tumors and are poorly defined, presenting a challenging scenario for subsequent tumor management.

We present a rare case in which the symptoms of necrotizing fasciitis concealed gingival cancer to remind clinicians to include malignant tumors in the differential diagnosis consistently.

A 56-year-old male presented with significant erythema and swelling encompassing the infra-orbital, buccal, submandibular, sublingual, and preauricular spaces on the left side and concomitant fever and trismus. We checked CT scan (Fig. 1A and B) and an incision and drainage procedure was performed under general anesthesia.

Samples for bacterial cultures and tissue analysis were collected. The microscopic evaluation (Fig. 1C) revealed acute suppurative inflammation. Improvement of infection was noted, however, we still found redness and induration in the left posterior buccal space and upper neck. Therefore, we arranged the second CT scan (Fig. 1D and E). We then proceeded with the second surgery (Fig. 1J). The pathological diagnosis was identical to those of the first operation (Fig. 1K).

In the week after the second surgery, the swelling had significantly improved. We then arranged for a third surgery to close the open wound of debridement. We accidently discovered verrucous lesions near the floor of the left mouth and lower gingiva during the surgery (Fig. 1H). The pathological results (Fig. 1I, J, and K) revealed a moderately differentiated squamous cell carcinoma at the left lower gingival and floor of mouth.

Subsequently, a contrast-enhanced head and neck CT scan (Fig. 1L, M, N and O) indicated soft tissue densities in the left buccal region and floor of mouth, along with evident destruction of the left anterior mandible. Regrettably, the patient declined the recommended surgical intervention and was subsequently lost to follow-up.

Studies suggest that many severe head and neck infections with unknown origins may reveal underlying malignancies. Some tumors can either cause infections directly or result from the inability of the patient to

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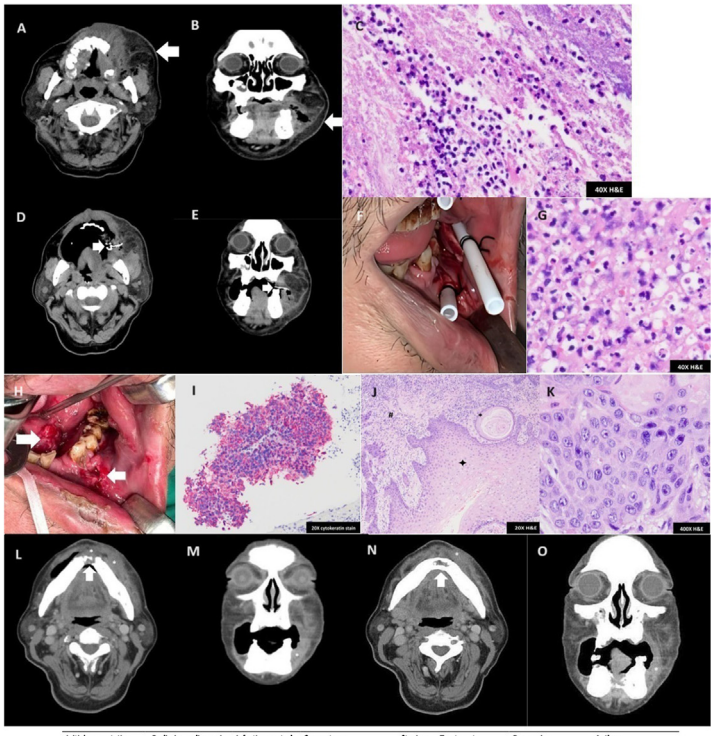

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Initial presentation	Preliminary diagnosis	Infection control	Cancer type	Staging	Treatment	Prognosis	Author
Sore throat, cough and neck swelling	Infected branchial cleft cyst	I&D	Maxillary sinus SCC	$T_4N_2M_1$	RND + RT	Died of disease	Wang et al. 2006
Painful swelling of neck, fever	Deep neck infection	I&D	Nasopharyngeal carcinoma	$T_4N_3M_1$	CCRT	Died of disease	Wang et al. 2006
-	Deep neck infection	Needle aspiration	Nasopharyngeal carcinoma	T_4N_3*	CCRT	Alive and well	Wang et al. 2006
Painful swelling of neck, fever	Deep neck infection	I&D	Parotid gland undifferentiated large cell carcinoma	$T_4N_3M_0$	Chemotherapy	Died of disease	Wang et al. 2006
-	Deep neck infection	Needle aspiration	Hypopharynx SCC	T_4N_3*	Chemotherapy	Died of disease	Wang et al. 2006
-	Deep neck infection	Needle aspiration	Tonsil SCC	T_4N_3*	-	Lost to follow up	Wang et al. 2006
-	Deep neck infection	Needle aspiration	Tongue SCC	T_4N_3*	-	Died of disease	Wang et al. 2006
Painful swelling of neck, fever	Deep neck abscess	I&D	Hypopharynx and the base of the Tongue SCC	$T_2N_{2b}M_0$	Resection+RND+RT	Alive and well	Ridder et al. 2000
Hoarseness, sore throat and dysphagia	Prelaryngeal abscess	I&D	Transglottic SCC	-	Laryngectomy+RT	No recurrence	Op de beeck et al. 2001
Hoarseness, sore throat and dysphagia	Prelaryngeal abscess	I&D	Laryngeal SCC	-	Laryngectomy+RT	Die of metastasized lung cancer	Op de beeck et al. 2001
Painful swelling in supraclavicular fossa	-	I&D	Supraglottic SCC	$T_2N_2M_0$	RT	No residual disease	Lee et al. 1996
Neck swelling	-	Needle aspiration	Supraglottic SCC	$T_1N_2M_0$	RT	-	Lee et al. 1996

Figure 1 Radiographic, pathologic, and clinical photographs of our case. (A and B) The CT scan revealed left facial swelling with gas formation in the left buccal, submasseteric, and pterygomandibular spaces, suggesting the presence of necrotizing fasciitis. (C) Microscopic findings showed diffuse infiltration of acute and chronic inflammatory cells in the fibromuscular tissue and subcutaneous fat, accompanied by focal areas of necrosis. Thus, the pathological diagnosis was acute suppurative inflammation. (D and E) A Penrose drain was placed in the left buccal area (white arrow). The swelling had somewhat decreased, but there was still gas and an abscess in the buccal and submasseteric space that had not been dissected. (F) During the second surgery, we made a more profound and broader incision from the original wound site to drain and debride the area and placed three Nelaton tubes for further drainage. (G) The microscopic findings showed diffuse infiltration of acute and chronic inflammatory cells in the fibromuscular tissue and subcutaneous fat. (H) In the third surgery for wound closure, we found verrucous lesions between the left tongue, floor of mouth, and left buccal mucosa (white arrows). (I) A small piece of mucosal tissue showing cells with marked hyperchromatic and pleomorphic nuclei highlighted by cytokeratin stain is found. Carcinoma is favoured. (J) Fragments from the left floor of mouth and tongue tissue. Nests of neoplastic cells arose from the surface epithelium with connective tissue invasion. (* indicates nests of neoplastic tissue; # indicates connective tissue; ✦ indicates surface epithelium) (K) Under 400× magnification, the neoplastic cells revealed epithelial differentiation with 1—2 % individual cell dyskeratosis and 10 % keratin pearl formation, showed moderately differentiated squamous cells. (L, M, N, and O) The tumor survey CT scan revealed that the previously infected lesions had almost disappeared. Soft tissue densities over the left buccal region and left lower gingiva (label * in L, M, N and O). Moreover, anterior mandible bone destruction was noted (white arrow in L—N, and O) (P) Researchers retrospectively examined 301 patients with deep neck infections (Wang et al., 2006), identifying seven cases where primary head and neck cancers initially manifested as deep neck infections. These patients were aged 40 to 74, with a median age of 64, and were notably older than those with typical pyogenic deep neck infections. The study also highlighted squamous cell carcinomas predominantly at T3 or T4 stages, often with lymph node involvement. There are some isolated cases, such as those published in 2001 by Katya Op de beeck et al., reporting two cases of prelaryngeal neck abscess finally diagnosed as squamous cell carcinoma. Furthermore, Lee et al. and Ridder et al. reported cases of similar conditions, including two cases of supraglottic squamous cell carcinomas and one case of hypopharynx and tongue squamous cell carcinomas, respectively. (-Indicated not recorded in the paper; *Indicated the metastasis stage not mentioned in the paper; SCC: Squamous cell carcinoma; CCRT: concurrent chemoradiotherapy; RND: Radical Neck Dissection; RT: Radiotherapy).

maintain proper oral hygiene due to the tumor, leading to the secondary dental infections, which can even progress to severe diseases such as Ludwig's angina and necrotizing fasciitis. Due to various complex conditions, the exact number of primary head and neck cancers initially presenting as infections remains poorly defined.[1]

A case report[2] published in 1985 described a 60-year-old woman who experienced breathing difficulties and progressive swelling in the neck and tongue, resembling Ludwig's angina. A tongue base mass was discovered during intubation and was subsequently confirmed as invasive squamous cell carcinoma on biopsy.

In another study, researchers retrospectively examined 301 patients with deep neck infections,[1] identifying seven cases where the primary head and neck cancers initially manifest as deep neck infections. There are some isolated cases, such as Katya Op de beeck et al. published in 2001, reporting two cases of prelaryngeal neck abscess finally diagnosed as squamous cell carcinoma.[3] Furthermore, Lee et al. and Ridder et al. reported cases of similar conditions,[4,5] including two cases of supraglottic squamous cell carcinomas and one case of hypopharynx and tongue squamous cell carcinomas, respectively. We have compiled detailed information about these cases in Fig. 1P.

Hence, patients with tumors who undergo incision and drainage due to severe inflammation or who have had a biopsy or needle aspiration must be treated aggressively as if they have positive-margin tumors. In this case, even when tumors are removed with clear margins and there is no metastasis to the neck lymph nodes, it is recommended to consider adjuvant radiotherapy or concurrent chemotherapy due to possible margin involvement by incision and drainage. Establishing a careful follow-up procedure after the initial treatment is essential and recommended.

In conclusion, malignant tumors should be included in the differential diagnosis for cases with severe infection symptoms. In such cases, aggressive treatment approaches, including wider safe margins, neck dissection, and adjuvant therapy, may be necessary.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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