ABSTRACT

Incidences of metastatic tumor occurrences in the oral cavity are very rare and their participation is less than 1% of all the jaw tumors. About two-third of the total occurrences (65%) are derived from the primary lesions of breast, lungs, liver, kidney, pancreas and one-third oral metastases (35%) are derived from testicles, uterus, thyroid, colon, rectum, stomach and prostate. The commonly affected oral sites for metastases are gingiva, buccal mucosa, soft palate, tongue and jaw bones. Their occurrences are more common in mandible than in maxilla and are more predominantly seen in females. Among all oral metastases a metastatic lesion from a germinomatous or a nongerminomatous germ cell tumor is very unusual. Commonly these types of tumors are benign in nature, but occasionally they turned into carcinomatous lesions. An unusual case of oral manifestation of an extremely rare extragonadal nongerminomatous primary choriocarcinoma of pancreas, with secondaries in liver and lung, is described in this article.

Keywords: Oral metastasis of choriocarcinoma, Early oral manifestation of choriocarcinoma, Pancreatic primary choriocarcinoma, Primary extragonadal choriocarcinoma of pancreas.


Source of support: Nil

Conflict of interest: None declared

INTRODUCTION

The metastases from a primary tumor may occur anywhere in the body and the manifestation mainly depends upon the route of spread which may either be through the lymphatic system or through the bloodstream. But metastases in the oral cavity are rarely seen and they usually spread from the primaries of breast, lungs, kidney, prostate, thyroid, etc. and their occurrences in the oral cavity are less than 1% of all the jaw tumors.1-4 Germ cell tumors are rarely found in people and the metastases in the oral cavity from a germ cell tumor is extremely rare. Germ cell tumors are classified into germinomatous (gonadal origin) and nongerminomatous (extragonadal origin). Tumors of extragonadal origin must be viewed seriously as they exhibit serious signs including metastasis rather than tumors develop from gonads. Oral metastases from germinomatous (gonadal origin) germ cell neoplasms have been seldom reported. But metastases into the oral cavity from an extragonadal germ cell tumor are very rarely reported. ET Adeyabo and SO Ajike in 2004 reported six metastatic tumors of the oral cavity. Out of six cases only one metastasis was reported as originated from the primary germ cell tumor of gonads and four have chosen the mandible as the site for metastases.2 NJ D’Silva et al in 2006 in a retrospective study with 114 cases concluded the breast take prior position than lung, kidney and prostate for metastasizing to the oral cavity.1-3 They also reported that the mandible was the common site for metastases and females are more prone to suffer twice as that of males.3 If the oral cavity metastases is the first manifestation of the disease the diagnosis can be difficult since the symptoms mimic the odontogenic infection.2-5 Majority of the oral metastases are derived from tumors of epithelial and connective tissues only. The germ cell malignancy is serious life-threatening one and has high tendency to metastasize with bad prognosis, if neoplasm arises from the placental tissue or develops extragonadally.6 Oral metastases from gonadal origin such as testicular and prostate choriocarcinomas have been reported in literatures.7,8 Of all the oral metastases, metastases from an extragonadal choriocarcinoma is extremely rare7,8 and very few reports are seen in literatures. Generally the prognosis is worse if, choriocarcinoma is found in the liver, pancreas, brain,9 the primary tumor developed outside the gonads and the serum human chorionic gonadotropin hormone level is elevated in blood. In this case report we described about the unusual oral metastasis of an extragonadal primary pancreatic choriocarcinoma where the primary was identified only after the oral lesion was diagnosed. Only very few cases of oral metastases from primary choriocarcinoma of pancreas are reported so far.10

MATERIALS AND METHODS

A 24-year-old female attended with the complaints of swelling and pain in the left lower buccal and lingual gingival region in relation to 34, 35 and 36 since 3 months. On examination the growth was about 3 × 2 cm and bleed on probing (Fig. 1A). Appearance of the lesion was more looking like inflammatory, hyperplastic, granulomatous, or fibromatous. Radiograph revealed bone resorption below the teeth 34, 35 and 36 (Fig. 1B). The lesion was completely
removed along with shaky teeth and sent for biopsy. The biopsy report explained the presence of highly malignant pleomorphic tumor cells with the features of necrotic, granulation tissue spots. Immunohistochemical studies revealed elevated level of beta-human chorionic gonadotropin hormone (hCG), positive reports for cytokeratin, cytokeratin 7 and CD 10 (Figs 2A to D), which suggested that the metastatic lesion of the oral cavity is purely related to the features of germ cells and also gave the clue that the oral lesion might have metastasized from a primary germ cell tumor, existing elsewhere in the body. Subsequently the patient was referred to the relevant specialists where necessary investigations were performed.

The CT abdomen and chest revealed the presence of pseudocysts in pancreas involving the neck, body and tail regions (Fig. 3), multiple abscesses in the liver (Fig. 4) and a hypodense lobulated pulmonary mass present in the superior segment of the right upper lobe of the lung (Figs 5A and B). Histopathological and immunohistochemical examination of the CT-guided FNAC (Fig. 6) specimen of pancreas explained the presence of enormous amount of tumor cells and suggested the possibilities for germ cell or pleomorphic carcinoma of pancreas. Investigations like serum study showed an elevated level of beta-hCG up to 98,097 mIU/ml and a normal level of alpha-fetoprotein (AFP) to 1.31 mg/ml. The blood biochemistry also showed an elevated level of lipase 86,830 u/l and amylase 13,720 u/l which suggested the fluid collection around the pancreatic cysts. The elevated level of beta-hCG reading confirmed the presence of choriocarcinoma of pancreas. The treatment, radiotherapy and chemotherapy, given were not successful because of the extensive spread and the advanced condition of the malignancy.

DISCUSSION

In males lung tumor take an important role in metastasizing to the oral cavity where as in females breast tumor take a
malignant growth may occur anywhere in the body and the site of occurrence is mainly determined by the route of spread, which may be through lymphatic or bloodstream. The choriocarcinoma from the placenta or uterus may involve the retroperitoneal nodes first and subsequently reach the lungs, liver and the brain. Tamiolakis et al in the year 2007 explained that the red bone marrow is believed to be necessary for the establishment and proliferation of metastases and they also reported that the metastatic occurrence in the mandible bone is not common as the red marrow is scanty in the matured mandible. Scolozzi et al in 2006 reported that the increased blood flow in the inflamed gingival region may serve as the fertile place for the proliferation of the engulfed tumor cells. A remarkable drawback exist during histopathological examination of the metastasized tumor as the specimen contains only poorly differentiated tumor cells, which is challenging to determine the type and location of the primary tumor. Particular attention and knowledge, to diagnose and differentiate the oral lesion before arriving to a conclusion, is mandatory to every clinician. It has commonly happened in majority of cases that primary tumor is left undiagnosed at the time of metastasized jaw tumor is presented and diagnosed. Regarding this case, the primary carcinoma was not developed either in the placenta or in the uterus (germinomatous). Though the primary tumor developed in the pancreas (nongerminomatous) with secondaries in liver and lung the patient had no knowing about the existence of primary and its secondaries since they were symptom free at that time. The primary tumor and secondaries were diagnosed and treated only after the oral lesion was diagnosed. They first developed in the pancreas as the nongerminomatous primary, and metastasized to the oral cavity with secondaries in the lungs and liver which is an extremely rare entity and only two cases were reported in the literatures so far.

lead. Germ cell tumor is rarely seen in the population. In females it may commonly arise from the placenta or uterus following molar pregnancy. But in this case the patient had never been pregnant for the past 5 years. Usually the tumors are benign in nature and may occasionally change into carcinomatous lesion. Reason for the development of choriocarcinoma is unknown but protein deficiency and malnutrition are said to be a cause for molar pregnancy which in turn is responsible for the development of choriocarcinoma. Distant metastases from a primary
SUMMARY

Metastases into the oral cavity from a primary malignancy are rare which contributes only 1% of all the jaw tumors. Organs like breast, lung, kidney, thyroid, prostate play important role to produce metastases to the oral cavity. The choriocarcinoma from the placenta or uterus may readily involve the retroperitoneal nodes, and ultimately spread to the lungs, liver and the brain as the secondary. In our case the choriocarcinoma developed first in the pancreas as the extragonadal growth rather than developing from the placenta or from the uterus and metastasized to the oral cavity which is a very rare incident. At the time when the oral lesion was diagnosed the primary choriocarcinoma and the secondaries were left undiagnosed. The dental clinician’s responsibility is very important and he should be skilled enough to diagnose the disease properly in right time without making further delay.

CONCLUSION

Metastases of germ cell tumors occurrence in the oral cavity seldom happened. Primary extragonadal choriocarcinoma occurred in pancreas with secondaries in liver and lung is a very rare and highly malignant entity. In addition to the rarity, the metastases found in the oral cavity from primary choriocarcinoma of pancreas are extremely rare. A metastatic choriocarcinoma found in the oral cavity was initially diagnosed by the dental surgeon, which helped the general clinician to detect the extragonadal primary choriocarcinoma of pancreas and the secondaries in liver and lung. This incident shows that the responsibility of the dental surgeon is very essential for proper diagnosis of the oral lesion since all the lesions of the oral cavity mimic the odontogenic infection. Although the treatment, chemotherapy and radiotherapy, rendered to her promptly in time, the effort taken to save the patient end in failure.

REFERENCES


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