Metastatic papillary thyroid carcinoma of the mandible: Case report and literature review

Neda Kardouni Khoozestani¹, Farzaneh Mosavat², Mohammad Shirkhoda³, Azin Sedaghati²

Departments of ¹Oral and Maxillofacial Pathology and ²Oral and Maxillofacial Radiology, School of Dentistry, Tehran University of Medical Science, ³Department of Oncosurgery, Cancer Institute, Tehran University of Medical Sciences, Tehran, Iran

Abstract The oral cavity is not a usual site for metastasis including about one percent of all oral tumors. Metastatic papillary thyroid carcinoma (PTC) is uncommon and usually occurs in regional lymph nodes of neck, so mandibular metastasis is a rare event. We present a case with swelling in the right mandible that has been diagnosed metastatic tumor of PTC. The clinical features, radiographic aspect and treatment plan are discussed. Furthermore, a thorough review of literature revealed 77 published cases of metastatic thyroid carcinoma to the oral cavity with their summarized features.

Keywords: Mandible, metastasis, papillary thyroid carcinoma

Address for correspondence: Dr. Azin Sedaghati, Department of Oral and Maxillofacial Radiology, Tehran Dental University of Medical Sciences, North Kargar Street, Tehran, Iran. E-mail: azin.sedaghati@yahoo.com

Received: 13.05.2018, Accepted: 27.11.2018

INTRODUCTION

Malignancy of the thyroid gland is the most common in the endocrine system and has been identified into different subtypes depending on the cells or growth pattern.^[1,2] Papillary thyroid carcinoma (PTC), a well-differentiated tumor, is the most common histological subtype including 80%–90% of all thyroid cancer.^[3,4]

It is generally an asymptomatic, slow-growing nodule with desirable prognosis.^[2,4] PTC almost has a lymphatic spreading to regional lymph nodes. Distant metastasis with bloodstream dissemination are less frequent^[2,3] and they have poor prognosis and decreased survival rate.^[4]

Distant metastasis more frequently involves lungs and bones such as sternum, vertebrae, pelvis, ribs and femur.^[2,3,5] Mandibular metastasis is rare, and it counts about 1% of

| Access this article online | | | | | | |
|----------------------------|------------------------------------|--|--|--|--|--|
| Quick Response Code: | Mahaitai | | | | | |
| | www.jomfp.in | | | | | |
| | DOI: 10.4103/jomfp.JOMFP_106_18 | | | | | |

oral malignancies.^[1,6] This tumor almost presents in the premolar-molar and angle-ramus region of the mandible.^[3,7]

We present a rare case with the mandibular metastasis from PTC, and a systematic review of the literature is performed.

CASE REPORT

A 68-year-old man was referred to the cancer institute of Tehran University of medical sciences with a history of painless swelling in the right side of the mandible [Figure 1] and a medical history of thyroidectomy 3 years ago because of PTC. After that, he received suppression therapy with levothyroxine. One year later, follow-up in the whole body scan with I-131 showed no evidence of residual thyroid tissue in thyroid bed and negative for local and distant metastasis. On the second year after thyroidectomy, he complained of swelling in the mandible then admitted. Laboratory tests were

For reprints contact: reprints@medknow.com

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

How to cite this article: Khoozestani NK, Mosavat F, Shirkhoda M, Sedaghati A. Metastatic papillary thyroid carcinoma of the mandible: Case report and literature review. J Oral Maxillofac Pathol 2019;23:S97-105.

normal except elevated thyroid-stimulating hormone level that showed subclinical hypothyroid. Abdominal-computed tomography (CT) scan with and without contrast was normal although lung CT scan shows sparse hypodense nodules in bilateral hemithorax that suspect metastasis. Neck sonography revealed disruption of the mandibular cortex and a hypoechoic lesion in the mandibular bone that posed to bony mass with a pathological fracture. Furthermore, hyperechoic of related soft tissue and multiple lymph nodes was observed. A hypoechoic region closed to the cortex probability proposed hematoma. For the second time, the whole body scan with TC-99 shows hyperactive hotspots in the right side of the mandible and degenerative changes in the body joints [Figure 2]. No other markable abnormality in other skeletal bones were reported. Panoramic radiography shows an invasive ill-defined radiolucent lesion in the body, angle and ascending ramus of the right side of the mandible. The destruction of the lower cortex and pathological fracture of medial border of ramus was also observed. The cortical border of the inferior alveolar canal was not detected [Figure 3]. CT scan without contrast from maxillofacial region shows an expansile lesion that measured about 50 mm × 32 mm which laterally extends and destructs the mandibular body and causes thinning buccal plate, medially extension to the mylohyoid muscle is seen. The lesion also involved the mandibular canal and mental foramen [Figure 4a-d]. Perforation of the lingual plate and inferior border of the mandible were also observed [Figure 5]. Hematologic malignancies or minor salivary gland tumors with same features can be suggested. A thick cortex 15 mm × 13 mm level I LAPD is observed on the right side.

Then, the incisional biopsy was performed. Microscopic examination shows bone trabeculae infiltrated by a malignant epithelial neoplasm composed of papillary structures which are characterized by distinctive nuclear features. The papillae are formed by fibrovascular cores, which are covered by cuboidal to columnar neoplastic epithelial cells. The crowded cells show round-to-ovoid nuclei with frequent indentations and typical clefted or grooved appearance. Furthermore, some nuclei have an empty or clear appearance. The cell cytoplasm is typically smooth and eosinophilic. The fibrous stroma shows lymphoplasmacytic infiltration. Perineural and lymphovascular invasion are identified [Figure 6a-d]. Due to the history of malignancy of thyroid and microscopic appearance, the diagnosis was metastatic carcinoma. The mentioned diagnosis was confirmed immunohistochemically by thyroid transcription factor 1 and thyroglobulin markers [Figure 7a and b].



Figure 1: Clinical view of the patient showing expansion in the right side of the mandible



Figure 2: Bone scan shows hot spot at the right side of the mandible



Figure 3: Panoramic view reveals an ill-defined radiolucency with interruption integrity of the right side mandibular cortex

The patient undergoes hemimandibulectomy and the right neck dissection level I-V. The mandible was reconstructed with a costochondral rib graft [Figure 8]. The pathology showed one reactive lymph node in level I. It was reported metastatic papillary thyroid carcinoma.



Figure 4: Computed tomography without contrast. Oblique coronal view shows perforation of inferior and lingual mandibular border (a and b). Axial view shows buccal and lingual cortex expansion (c and d)



Figure 6: Microscopic examination reveals papillary thyroid carcinoma with multiple branching and true papillae (a). Papillary thyroid carcinoma with bone trabeculae, filtration and destruction (b). Branching papillae lined by cuboidal cells with overlapping nuclei and finely dispersed ground glass chromatin "Orphan Annie nuclei" (c). Cuboidal cells with nuclear longitudinal grooves micronucleoli and eosinophilic intranuclear inclusions (d)

DISCUSSION

Metastatic tumor to the oral region is a rare event that comprises 1% of all oral malignancies.^[3,8] The primary malignancy that is giving more frequently metastasis to oral cavity differed between genders. These are breast cancer for a woman and lung cancer for men.^[9-11]



Figure 5: Three-dimensional reformatted computed tomography shows expansile destructive lesion



Figure 7: Thyroglobulin immunostaining reveals thyroid differentiation of the tumor cells (a). Thyroid transcription factor immunostaining shows strong nuclear expression in all cancer cells (b)

Other common primary sites are colon, kidney, prostate, bone, liver, adrenal gland and female's genital organs.^[3,9,12] Metastatic thyroid carcinoma to oral tissue is not very frequent. Metastatic PTC tumors to jaw include 4%–6.5% in all metastatic tumors to the jaw.^[3,9]

Most metastasis bones from PTC are being sternum, vertebrae, skull, pelvis, ribs and femur.^[5,10,13]

A review literature of available published cases for metastatic thyroid carcinoma to oral tissues revealed 77 cases with their summarized features in Table 1.

Patients' age with thyroid cancer is usually from 25 to 65 years, and especially in younger patients, the tumor tends to grow very slowly.^[2] As regards, the mean age for well-differentiated thyroid cancer is 40 years for papillary type and 50 years for follicular type; so, it spends the time to metastasize to other regions.^[3] The metastatic tumors commonly occur in 5th to 7th decade of life.^[14,15]

In our review, based on available information, among 73 cases with a wide age range (13–87), the mean age of patients was 56.6 years. The mean age was 64.1 years for males and 54 years for females. Most patients (45 cases)



Figure 8: Reconstruction of segmental mandibulectomy

were in 6th decade (24 cases) and 7th decade (21 cases), only 6 cases were younger than 40 years.

In this review, among 74 patients, 57 cases (77%) were females and 17 cases (23%) were males. Female predilection can be attributed to the more incidence of thyroid cancer in women.^[16,17]

Distant metastasis from PTC is very rare because it usually remains intraglandular or metastasize to regional cervical lymph nodes through the lymphatic system although follicular thyroid carcinoma with hematogenous spread causes more frequent distant metastasis.^[1,18,19] Metastasis to the jaw bone develops through blood vessels, that is the most common route.^[14]

Table 1: Summarizes of published case reports with metastatic thyroid carcinoma to the oral cavity (77 cases)

| Authors | Age | Gender | Location | Histological diagnosis | Time to metastasis | Treatment to metastasis | Other metastatic sites | Outcome (length of follow-up) |
|--|-----|--------|--------------------------------|---|------------------------|--|---|-------------------------------------|
| Meyer and Shklar, 1965 ^[3] | 51 | Female | Mandible | Adenocarcinoma | NA | NA | None | NA |
| Soumar <i>et al.</i> , 1970 ^[16] | 54 | Female | Mandible | NA | NA | NA | NA | NED (36 months) |
| McDaniel <i>et al</i> ., 1971 ^[3] | 77 | Female | Right mandible | Follicular thyroid Ca | First manifestation | Hemimandibulectomy and parotidectomy | None | NED (4 years) |
| | 48 | Female | Left mandible | Follicular thyroid Ca | First manifestation | NA | NA | NA |
| Al Ani 1973 ^[3] | 60 | Female | Right mandible | Follicular thyroid Ca | First manifestation | NA | NA | NA |
| Ripp <i>et al</i> ., 1977 ^[3] | 61 | Female | Right mandible | Thyroid Ca | First manifestation | Cobalt teletherapy | Widespread | DOD (1 year) |
| Draper <i>et al</i> ., 1979 ^[3] | NA | Female | Mandible | Follicular thyroid Ca | NA | Radiotherapy | skull | NA |
| Stypulkowska | NA | NA | Mandible | Adenocarcinoma | 1 year | symptomatic | NA | DOD (≤ 1 year) |
| <i>et al.</i> , 1979 ^[16] | NA | NA | Mandible | Adenocarcinoma | First manifestation | symptomatic | NA | DOD (\leq 1 year) |
| Osguthorpe and Bratton, 1982 ^[3] | 53 | Male | Right mandible | Follicular thyroid Ca | First manifestation | Partial mandibulectomy | None | NED (3 years) |
| Nishimura <i>et al.</i> , 1982 ^[1] | 74 | Female | Right Mandible | Follicular thyroid Ca | First manifestation | Chemotherapy | Vertebra | Alive (4 months) |
| | 51 | Female | Left mandible | Papillary thyroid Ca (follicular variant) | First manifestation | Chemotherapy, radiotherapy Hemimandibulectomy | Ribs , Lung | Alive with disease (4.5 year) |
| Parichatikanond et al.,1982 ^[3] | 42 | Female | Left mandible | Follicular thyroid Ca | First manifestation | Hemimandibulectomy | NA | NA |
| Tovi <i>et al.</i> , 1984 ^[3] | 33 | Male | Left mandible | Follicular thyroid Ca | First manifestation | Radioactive lodine therapy | Vertebra, mediastinum Parietal bone, femur | Died of thyroid Crisis (17 days) |
| Markitziu <i>et al.</i> , 1986 ^[3] | 69 | Female | Left mandible and parotid | Papillary thyroid Ca (follicular variant) | First manifestation | Radiotherapy and chemotherapy | NA | NED (1.5 years) |
| Kahn and McCord 1989 ^[3] | 82 | Female | Anterior mandible | Follicular thyroid Ca | 32 years | Radiotherapy, radioactive lodine therapy, resection | Mediastinum, tibia, lungs, pelvis | Died, NED (1.5 years) |
| Whitaker <i>et al.</i> , 1993 ^[3] | 87 | Male | Dorsal tongue and lower lip | Follicular thyroid Ca | First manifestation | Surgical removal, Radioactive lodine therapy | Lung | Alive with disease (4 months) |
| Hefer, 1998 ^[16] | 58 | Male | Maxilla | Follicular thyroid Ca | NA | Resection | NA | NED (2 years) |

Khoozestani, et al.: Metastatic papillary thyroid carcinoma of the mandible

| Table 1: Contd. | •• | | | | | | | |
|--|----------|------------------|--|--|------------------------|---|-----------------------------|------------------------------------|
| Authors | Age | Gender | Location | Histological diagnosis | Time to metastasis | Treatment to metastasis | Other metastatic sites | Outcome (length of follow-up) |
| Vural and Hanna 1998 ^[3] | 64 | Female | Right Mandible infratemporal fossa | Follicular thyroid Ca | First manifestation | Radical resection, radioactive iodine therapy | Multiple skeletal | Alive (6 weeks) |
| Agarwal <i>et al</i> ., 1998 ^[3] | 45 | Female | Left mandible | Follicular thyroid Ca | First manifestation | Hemimandibulectomy | NA | Alive (2 weeks) |
| Erdag <i>et al.</i> , 1999 ^[3] | 53 | Female | Right mandible | Papillary thyroid Ca (follicular variant) | 4 years | Radical Resection, Radioactive lodine therapy | Vertebra, Humerus | Alive with disease (2.5 years) |
| Anil <i>et al.,</i> 1999 ^[3] | 61 | Female | Right Mandible | Follicular thyroid Ca | 8 years | NA | NA | NA |
| Piatelli <i>et al.</i> , 2000 ^[3] | 54 | Female | Right Maxillary Gingiva | Medullary thyroid Ca | 4 months | Surgical removal | Cervical lymph nodes | NED (4 years) |
| Thomas <i>et al.</i> , 2001 ^[13] | 61 | Female | Mandibular gingiva | Follicular thyroid Ca | First manifestation | Radio lodine ablation | None | NED (5 years) |
| Bhansali <i>et al.</i> , 2003 ^[5] | 60 | Female | Mandible, maxilla | Papillary thyroid Ca (follicular variant) | NA | NA | Rib | NA |
| Colella <i>et al.</i> , 2003 ^[3] | 50 | Female | Right mandible | Papillary thyroid Ca | 5 years | NA | NA | NA |
| Ostrosky <i>et al.</i> , 2003 ^[3] | 72 | Male | Anterior mandible | Follicular thyroid Ca | First manifestation | Resection | None | NA |
| Bonder <i>et al.</i> , 2006 ^[3] | 65 | Male | Right mandible | Hurtle cell thyroid Ca | NA | Supportive | Widespread | DOD (2 months) |
| Liu <i>et al.</i> , 2007 ^[26] | 66 | Male | Masticator space, mandible | Papillary thyroid Ca (follicular variant) | 22 years | Radical Resection, radioactive iodine therapy | Scapula | NED (3 years) |
| Kaveri <i>et al</i> ., 2007 ^[14] | 65 | Male | Left mandible | Follicular thyroid Ca | First manifestation | NA | Ribs | NA |
| Tamiolakis <i>et al</i> ., | 69 | Female | Mandible | Papillary thyroid Ca | NA | Inoperable | NA | DOD (1 month) |
| 2007 ^[3] | NA 13 | NA Female | Mandible | Thyroid Ca Papillary | NA | Resection | NA | NA |
| Antunes 2008 ^[3] | 15 | Temale | Waxilla | Adenocarcinoma | NA | NA | NA | NA |
| Araki <i>et al.</i> , 2008 ^[16] | 55 | Female | Left mandible | Follicular thyroid Ca | NA | NA | NA | NA |
| Ismail <i>et al</i> ., 2009 ^[3] | 70 | Female | Left mandible | Follicular thyroid Ca | First manifestation | NA | Hilar lymph nodes | NA |
| Algahtani <i>et al.</i> , 2009 ^[3] | 66 | Female | Left mandible | Follicular thyroid Ca | First manifestation | Segmental mandibulectomy | Cervical lymph nodes | NA |
| Seoane et al., | 58 | Female | Tongue | Papillary thyroid Ca | NA | NA | NA | NA |
| $2009^{[10]}$ | 69 02 | Female | Mandible | Papillary thyroid Ca | NA | NA | NA | NA |
| 2010 ^[3] | 03 | remale | mandible | differentiated | z years | None | Lung | months) |
| Kumar <i>et al</i> ., 2010 ^[3] | 58 | Female | Left mandible | Follicular thyroid Ca | First manifestation | Segmental mandibulectomy | None | NED (2 years) |
| Yokoe <i>et al.</i> , 2010 ^[27] | 71 | Female | Mandible | Follicular thyroid Ca | First manifestation | Segmental mandibulectomy | NA | NED (48 months) |
| Chaturvedy <i>et al.</i> , 2010 ^[28] | 68 | Male | Upper lip | Follicular thyroid Ca | 4 years | Surgical excision Radioactive lodine therapy | None | NA |
| Daley and Darling 2011 ^[3] | 57 | Male | Anterior mandible gingiya | NA | NA | NA | NA | NA |
| Kim <i>et al.</i> , 2011 ^[18] | 46 | Female | Bilateral mandible | Follicular thyroid Ca | First manifestation | Hemimandibulectomy Radioactive lodine therapy | Lumbar spine, Femur neck | NED (12 months) |
| Muttagi <i>et al.</i> , 2011 ^[15] | 60 | Male | Mandible | Papillary thyroid Ca (follicular variant) | NA | NA | NA | NA |
| ··· | 63 | Female | Mandible | Papillary thyroid Ca (follicular variant) | NA | NA | NA | NA |
| | 51 | Female | Mandible | Papillary thyroid Ca (follicular variant) | NA | NA | NA | NA |
| | 44 35 | Female Female | Mandible Mandible | Papillary thyroid Ca Papillary thyroid Ca | NA NA | NA NA | NA NA | NA NA |

Khoozestani, et al.: Metastatic papillary thyroid carcinoma of the mandible

| Table 1: Contd. | ••• | | | | | | | |
|--|-----|--------|---|--|------------------------|--|--|----------------------------------|
| Authors | Age | Gender | Location | Histological diagnosis | Time to metastasis | Treatment to metastasis | Other metastatic sites | Outcome (length of follow-up) |
| Rohilla <i>et al</i> ., 2011 ^[24] | 55 | Female | Edentulous mandible | Follicular thyroid Ca | 2 years | Radiotherapy | Skull, ileum, Pelvis, vertebrae | Alive (6 month) |
| Narain and Batra, 2011 ^[29] | 62 | Female | Right maxilla | Follicular thyroid Ca | 15 years | NA | lymph nodes | NA |
| Nikitakis <i>et al.</i> , 2011 ^[3] | 63 | Male | Right posterior Maxilla, maxillary sinus | Papillary thyroid Ca | 2 years | Radiotherapy Chemotherapy | Sternum, ribs, left tibia | Alive with disease (2 years) |
| Slim <i>et al.</i> , 2012 ^[11] | 67 | Female | Maxilla (malar region) | Papillary thyroid Ca (follicular variant) | First manifestation | Radioactive iodine | Facial region, lung, neck | NA |
| Bhadage <i>et al.</i> , 2012 ^[21] | 40 | Female | Left mandible | Follicular thyroid Ca | First manifestation | NA | Submandibular | NA |
| Shabestari <i>et al.</i> , 2012 ^[17] | 21 | Female | Anterior left maxilla | Medullary thyroid Ca | 7 years | Chemotherapy | NA | NA |
| Pasupula <i>et al.</i> , 2012 ^[30] | 40 | Female | Left ramus of mandible | Follicular thyroid Ca | First manifestation | Excision | Rib | NA |
| Kotina <i>et al.</i> , 2013 ^[6] | 55 | Female | Left mandible | Follicular thyroid Ca | 15 years | NA | NA | NA |
| Kumar <i>et al.</i> , 2013 ^[31] | 31 | Female | Right maxilla | Follicular thyroid Ca | First manifestation | Radioactive iodine therapy Palliative chemotherapy | Right leg | NED (7 years) |
| Vishveshwaraiah et al., 2013 ^[26] | 56 | Female | Right mandible | Follicular thyroid Ca | NA | NA | None | NA |
| Pingel <i>et al.</i> , 2013 ^[16] | 76 | Male | Posterior mandible | Papillary thyroid Ca (tall cell variant) | 6 years | Radiotherapy | Rib Left clavicle | DOD (11 months) |
| Vazifeh mostaan <i>et al.</i> , 2013 ^[32] | 58 | Female | Right mandible | Follicular thyroid Ca | 12 years | Segmental mandibulectomy | None | NA |
| Das <i>et al.</i> , 2014 ^[22] | 55 | Female | Right mandible | Papillary thyroid Ca (follicular variant) | First manifestation | Segmental mandibulectomy Radioactive lodine therapy | None | NED (2 years) |
| Lavanya <i>et al</i> ., 2014 ^[7] | 76 | Male | Left Mandible | Follicular thyroid Ca | First manifestation | NA | NA | NA |
| Siddique <i>et al.,</i> 2015 ^[8] | 71 | Male | Labial gingiva of lower right lateral incisor | Papillary thyroid Ca | 8 weeks | Radioactive iodine therapy | Lymph nodes | NA |
| Kori <i>et al</i> ., 2015 ^[19] | 50 | Female | Left mandible | Follicular thyroid Ca | First manifestation | Segmental mandibulectomy Radioactive lodine therapy | None | NED (1 years) |
| | 40 | Female | Left mandible ramus, maxilla | Follicular thyroid Ca | First manifestation | Radioactive iodine therapy | None | NA |
| Bingol <i>et al.</i> , 2015 ^[2] | 33 | Female | Right mandible | Papillary thyroid Ca (follicular variant) | First manifestation | Hemimandibulectomy | Pelvic, central nervous system | DOD (5 years) |
| Hartinie <i>et al.</i> , 2015 ^[33] | 41 | Female | Right mandible | Follicular thyroid Ca | First manifestation | Segmental mandibulectomy, Radiotherapy | Anterior mediastinal nodes | NED (6 months) |
| Fatahzadeh <i>et al.</i> , 2015 ^[34] | 43 | Female | Right Maxilla, Maxillary sinus | Papillary thyroid Ca (follicular variant) | NA | Palliative radiotherapy | Left leg, scalp Right shoulder, Bilateral hips | NA |
| Krishnamurthy et al., 2016 ^[23] | 52 | Male | Left mandible | Follicular thyroid Ca | First manifestation | Hemimandibulectomy Radioactive iodine | None | NED (14 months) |
| Cai <i>et al</i> ., 2016 ^[25] | 59 | Female | Ascending Ramos of mandible | Thyroid adenocarcinoma | NA | Segmental mandibulectomy | NA | Alive (27 months) |
| Arepen and Mohamad <i>et al.</i> , 2016 ^[1] | 68 | Female | Bilateral mandible | Papillary thyroid Ca (follicular variant) | 4 years | Palliative radiotherapy | None | NA |
| Dholam <i>et al.</i> , 2017 ^[10] | 58 | Female | Left mandible | Papillary thyroid Ca (follicular variant) | First manifestation | Palliative radiotherapy | Sacral, lumbar | Alive (6 months) |
| Anajar <i>et al</i> ., 2017 ^[4] | 52 | Female | Mandible | Papillary thyroid Ca | First manifestation | Segmental mandibulectomy | None | NED (6 months) |

| Authors | Age | Gender | Location | Histological diagnosis | Time to metastasis | Treatment to metastasis | Other metastatic sites | Outcome (length of follow-up) |
|--|-----|--------|----------------|---------------------------|------------------------|--|--------------------------------|----------------------------------|
| Loureiro <i>et al.</i> , 2017 ^[12] | 54 | Female | Left mandible | Follicular thyroid Ca | First manifestation | None | NA | DOD |
| Varadarajan 2017 ^[9] | 73 | Female | Left mandible | Follicular thyroid Ca | First manifestation | Segmental mandibulectomy Radioactive iodine therany | Tongue, soft tissue of neck | NED (18 months) |
| Present case, 2017 | 68 | Male | Right mandible | Papillary thyroid Ca | 3 years | Hemimandibulectomy | None | Alive (12 months) |

NA: Not available, DOD: Died of disease, NED: No evidence of disease

Table 1. Contd

The most frequent subtype of metastatic thyroid carcinoma is the follicular type (39 out of 75 with available data: 52%). Its more predilection can be attributed to the bloodstream dissemination.^[3,11] PTC along with follicular variant (19 cases) and tall cell variant (1 case) accounted for 25 cases (33%) including present reported case. Although 11 cases were reported with medullary thyroid carcinoma, hurtle cell carcinoma, poorly differentiated thyroid carcinoma and adenocarcinoma.

Hirshberg *et al.*^[20] reported that the first sign of distant undiscovered malignancy in 23% of cases was oral metastasis. Oral metastasis in one-third of patients may be the first manifestation of its primary tumor.^[20]

Mandible, maxilla and oral soft tissue are more affected sites by metastatic cancer with predilection for mandible and gingiva.^[4,7] Almost 41% of facial bones metastasis from thyroid cancer occurs in the mandible.^[9,21] Maxillary metastasis is rare and less than one-fifth of all jaw metastatic tumors.^[3]

Oral soft-tissue involvement is less frequent than the jaw bones.^[2,16] Ramus and angle of the mandible due to rich blood circulation in the medullary cavity are most locations to metastasis^[3,4,22] and in radiography has been shown with poorly defined osteolytic lesion with ragged border.^[1]

In review of cases with available data, oral metastasis was the first manifestation of thyroid cancer in 37 patients (66%). In 19 patients including the present case, there is a time to diagnosis metastasis after discover primary thyroid cancer. This time has widely ranged from 8 weeks to 32 years.

The most location of oral metastasis was the mandible (80%), including our case and only 10% was in the maxilla. Soft-tissue metastasis in gingiva, tongue, lower and upper lip were seen in 7 cases.

The common symptoms of metastatic tumor to jaws are pain, swelling, tooth mobility, premature loss of teeth, paresthesia, cervical lymphadenopathy and rarely pathologic fractures.^[16,17,23] Because of rarity of oral metastasis, the first diagnosis can be squamous cell carcinoma, that is a most common malignant lesion in jaws with same clinical features.^[24] The presence of pain and chin paresthesia can be due to rapid progression of intraoral and extraoral expansion.^[9] A granulation-like mass in oral tissue and mucosa may appear and result in bleeding, infection, dysphagia and disturbance in mastication.^[9,16]

In the present case, painless swelling was a noticeable clinical sign and extra examination also showed pathologic fracture.

Oral metastasis is similar to an inflammatory and reactive conditions such as periapical lesions, periodontitis, osteomyelitis and pericoronitis.^[2,3]

The treatment modalities of oral metastasis from thyroid cancer have been varied from palliative to various combination management of surgical interventions, radioactive iodine ablation, radiotherapy, chemotherapy and hormone therapy.^[7,16,25]

For better survival, it has recommended total thyroidectomy (if not performed in the past) with surgical resection of metastatic turner, followed radioactive iodine treatment or radiotherapy.^[3,4] In younger patients with small metastasis lesion, I 131 seems to be a more effective treatment.^[16]

The treatment decision depends on spreading metastatic lesion, symptoms and fracture risk.^[3,4] In widespread metastasis disease, usually palliative treatment has been carried out; on the other hand, for the solitary and accessible lesion, surgical intervention has been recommended.^[3,4]

Free fibular flap for mandible reconstruction is a gold standard.^[4] In the present case, thyroidectomy was performed 3 years earlier before, and then, hemimandibulectomy with radical neck dissection of the metastatic tumor was done.

The prognosis depends on the age at diagnosis of metastasis tumor and the number of involved bones^[4] although patient prognosis with distant metastasis of thyroid cancer is generally poor and the survival rate is 4 years on average 40% of patients after discovering metastatic lesion.^[3,25] Metastatic thyroid carcinoma may be present adjacent to facial structures such as orbit, paranasal sinus and salivary gland.^[3,9]

The available follow-up information in our review revealed in 40 out of 77 cases, length of follow-up varied from 17 days to 7 years. About 25 patients were reported as alive (14 of them being free of disease) and 15 patients died (13 of them due to disease).

The review shows in 32 out of 48 cases, oral metastasis was accompanied by metastasis in other sites such as femur, tibia, lung, ribs, lymph nodes, skull and pelvis, that they are usually skeletal.

CONCLUSION

Oral metastasis from PTC is a very rare event that usually accompanies with a poor prognosis. Mandible metastasis revealed various symptoms and can be similar to the other conditions if it is the first manifestation. Therefore, thorough diagnostic workup for detection of the primary and metastatic sites is necessary. A proper evaluation can help to decide the best treatment. Surgical-based treatment with reconstruction can improve surgical outcome.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Arepen SA, Mohamad I. Massive bilateral mandibular metastasis from papillary thyroid carcinoma. Egypt J Ear Nose Throat Allied Sci 2016;18:167-8.
- Bingol UA, Cinar C, Cetinkale O. Mandibular metastases of papillary thyroid carcinoma treated by hemimandibulectomy and costochondral rib graft. Plast Reconstr Surg Glob Open 2015;3:e305.
- Nikitakis NG, Polymeri A, Polymeris A, Sklavounou A. Metastatic papillary thyroid carcinoma to the maxilla: Case report and literature review. Head Neck Pathol 2012;6:216-23.
- Anajar S, Hassnaoui J, Rouadi S, Abada R, Roubal M, Mahtar M. Mandibular metastasis revealed papillary thyroid carcinoma: Rare case. Int J Surg Case Rep 2017;37:130-3.

- Bhansali A, Kataria RN, Subrahmanyam KA, Radotra BD, Mathur SK. Maxillary mass as the presenting manifestation of papillary thyroid carcinoma. Indian J Cancer 2003;40:80-1.
- Kotina S, Kumar K, Raghunath V. Metastatic thyroid carcinoma of the mandible-mimicking paragangliom. Oral Maxillofac Pathol J 2013;4:390-3.
- Lavanya C, Ranganathan K, Veerabahu M. Mandibular metastasis of thyroid carcinoma: A case report. J Clin Diagn Res 2014;8:ZD15-6.
- Siddique I, Chengot P, Frewer J, Walker D. A rare case of gingival metastases from papillary thyroid carcinoma. Int J Surg Case Rep 2015;7C: 82-4.
- 9. Varadarajan VV, Pace EK, Patel V, Sawhney R, Amdur RJ, Dziegielewski PT, *et al.* Follicular thyroid carcinoma metastasis to the facial skeleton: A systematic review. BMC Cancer 2017;17:225.
- Dholam KP, Singh GP, Gurav SV, Rekhi BB. Rare late mandibular metastasis in follicular variant of papillary carcinoma thyroid: 'Resurgence of the sleeping tumour'. Arch Otolaryngol Rhinol 2017;3:1-5.
- Slim I, Mhiri A, Meddeb I, Goucha A, Gritli S, Ben Slimene MF. Malar bone metastasis revealing a papillary thyroid carcinoma. Case Rep Otolaryngol 2012;2012:795686.
- Loureiro AC, Figueiredo C, Matos JDM, Lima RV, Vasconcelos RBA, Nunes GA, *et al.* Metastasis of thyroid adenocarcinoma in mandible. Int J Dent Oral Sci 2017;4:439-43.
- Thomas G, Sreelatha KT, Pradeep VM, Pandey M. Follicular carcinoma of the thyroid gland with initial presentation as an oral soft tissue mass: Report of a case. J Oral Maxillofac Surg 2001;59:819-22.
- Kaveri H, Punnya VA, Tayaar AS. Metastatic thyroid carcinoma to the mandible. J Oral Maxillofac Pathol 2007;11:32-4.
- Muttagi SS, Chaturvedi P, D'Cruz A, Kane S, Chaukar D, Pai P, et al. Metastatic tumors to the jaw bones: Retrospective analysis from an Indian tertiary referral center. Indian J Cancer 2011;48:234-9.
- Pingel K, Kalogirou E, Koutlas I, Tosios K. Mandibular metastasis of a papillary thyroid carcinoma, tall cell variant. Case report and review of the literature. Hell Arch Oral Maxillofac Surg 2013;14:143-52.
- Shabestari SB, Shirinbak I, Agha-Hosseini F. Maxillary metastasis of a medullary thyroid carcinoma in a 21-year-old woman 7 years after thyroidectomy. J Oral Maxillofac Surg 2012;70:1495-9.
- Kim DW, Hah JH, An SY, Chang H, Kim KH. Follicular thyroid carcinoma presenting as bilateral cheek masses. Clin Exp Otorhinolaryngol 2013;6:52-5.
- Kori CH, Vishnoi JR, Rajan SH, Malhotra KP, Gupta S, Kumar V. Mandibular metastasis in patients of follicular thyroid carcinoma: A rare entity; report of two cases. Int J Health Sci Res 2015;5:581-5.
- 20. Hirshberg A, Leibovich P, Buchner A. Metastatic tumors to the jawbones: Analysis of 390 cases. J Oral Pathol Med 1994;23:337-41.
- Bhadage CJ, Vaishampayan S, Umarji H. Mandibular metastasis in a patient with follicular carcinoma of thyroid. Contemp Clin Dent 2012;3:212-4.
- Das R, Kumar M, Sharma JD, Krishnatreya M, Chakraborty PS, Kataki AC. Papillary thyroid carcinoma presenting with mandibular metastasis: An unusual presentation. Clin Cancer Investig J 2014;3:426-8.
- Krishnamurthy A, Deen S, Ramshankar V, Majhi U. Metastatic follicular carcinoma thyroid masquerading as a primary jaw tumor. J Maxillofac Oral Surg 2016;15:266-9.
- Rohilla K, Ramesh V, Singh V, Sriram K. Thyroid carcinoma metastasising in the mandible: A case report. J Int Med Sci Acad 2011;24:189-90.
- Cai Z, Zhu C, Wang L, Zhu L, Zhang Z, Zhu H, *et al.* A retrospective study of six patients with mandibular metastatic carcinoma. Oncol Lett 2016;11:3650-4.
- Vishveshwaraiah PM, Mukunda A, Laxminarayana KK, Kasim K. Metastatic follicular thyroid carcinoma to the body of the mandible mimicking an odontogenic tumor. J Cancer Res Ther 2013;9:320-3.
- Yokoe H, Kasamatsu A, Ogoshi K, Ogawara K, Endo-Sakamoto Y, Ono K, *et al.* Mandibular metastasis from thyroid follicular carcinoma: A case report. Asian J Oral Maxillofac Surg 2010;22:208-11.
- 28. Chaturvedy G, Kumar R, Sikka K, Karthikeyan CV, Mathur SR. An

unusual site of distant metastasis in carcinoma of the thyroid. Indian J Cancer 2010;47:479-80.

- Narain S, Batra H. Metastatic carcinoma of maxilla secondary to primary follicular carcinoma of thyroid gland – A case report. Indian J Dent 2011;2:30-2.
- Pasupula AP, Dorankula SP, Thokala MR, Kumar MP. Metastatic follicular thyroid carcinoma to the mandible. Indian J Dent Res 2012;23:843.
- 31. Kumar CS, Shanmugam D, Venkatapathy R, Munshi MA. Metastatic follicular carcinoma of thyroid in maxilla. Dent Res J (Isfahan)

2013;10:817-9.

- Vazifeh Mostaan L, Irani S, Rajati M, Memar B. Mandibular metastasis from follicular thyroid carcinoma: A rare case after twelve years. Arch Iran Med 2013;16:557-9.
- Hartinie M, Rosli YM, Muhd HA, Rusdi Abd R, Shaifulizan R. Metastatic thyroid follicular carcinoma to the mandible: A clinical note. Int Med J 2015;22:327-9.
- 34. Fatahzadeh M, Subramanian G, Singer SR. Metstatic papillary thyroid carcinoma to maxilla: A rare case. Quintessence Int 2015;46:431-5.