ARC Journal of Dental Science
Volume 6, Issue 2, PP 6-9
ISSN (Online) 2456-0030
https://doi.org/10.20431/2456-0030.0602002
www.arcjournals.org

A Rare Case Report of Cholangiocarcinoma Metastasising in the Parotid Gland

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Abstract: *Cholangiocarcinoma is a rare cancer that forms in the bile duct and is often discovered in the later stages of the disease. There have been several reports of this cancer metastasising, with the most common site being the liver and other sites being the lining of the abdominal cavity, the lungs, bone and brain. However, there are very few documented cases of the presence of cholangiocarcinoma metastasis in the head and neck and fewer still that note infiltration of the parotid gland. For this reason, the case of a 69-year-old female with a confirmed diagnosis of cholangiocarcinoma was of particular interest to us as she later presented with a right sided facial swelling and a buccal fistula, which was confirmed to be cholangiocarcinoma metastasis, an extremely rare occurrence. Therefore, the aim of this report is to raise awareness of the signs and symptoms of oral cavity and parotid gland metastasis by exploring the history of this case.*

Keywords: *Cholangiocarcinoma, Parotid gland, metastasis, buccal fistula, oral cavity, mandible*

1. Introduction

There are very few documented cases of the presence of cholangiocarcoma metastasis in the head and neck, and fewer still, that note infiltration of the parotid gland. Cholangiocarcoma is a malignancy that arises from cholangiocytes, the epithelial cells that line the biliary tree[1]. It forms in the bile duct and commonly occurs in adults over the age of 50. This cancer can be subdivided into several categories and common signs and symptoms can include jaundice, itchiness of the skin, pale stools, fatigue, abdominal pain in the right side, unplanned weight loss, fever, night sweats and dark urine.

Cholangiocarcinoma is often diagnosed in the later stages with the possibility of metastasing to other structures. The only curative therapy currently is radical surgery however, due to the often-delayed presentation, the prognosis is often poor[2]. Such was the tragic case of a 69-year-old female who first presented with the diagnosis of cholangiocarcinoma and later developed a buccal mucosal fistula on the right side of the oral cavity.

2. Case Report

A 69-year-old female was referred to our service following the discovery of a right sided facial swelling and swelling on the right side of the neck. She was initially diagnosed with cholangiocarcinoma metastatic stage IV IHC following a liver biopsy which was conducted by the gastroenterology team, and following discussions with the oncology team, was advised to undergo palliative chemotherapy. Medically, she was hypertensive for which she was taking Amlodipine, was a previous smoker, and had a family history of her father having suffered from lung cancer and her sister having suffered from pancreatic cancer. A CT scan of the abdomen, chest and pelvis showed the liver was enlarged and contained a large lobular mass measuring approximately 16cm in maximal diameter with hepatic, nodal and bone metastases.

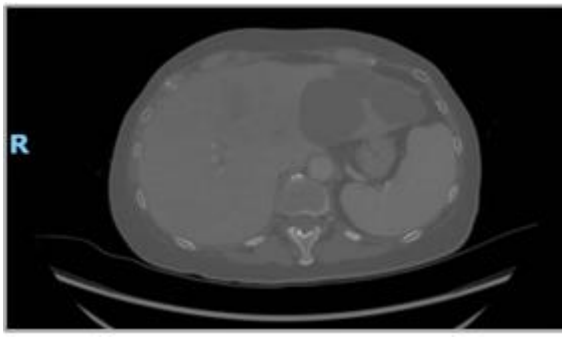

Figure 1.*CT chest/abdo/pelvis with contrast: The liver is enlarged and contains a large lobular heterogeneously enhancing mass (image taken 28-01-22)*

Two months after the initial diagnosis, the patient was admitted following attendance in the emergency department due to raised calcium levels. The patient had been experiencing fatigue, dizziness and shortness of breath and had appeared to have deteriorated rapidly, raising concerns with her oncologist. On assessment during admission, a one-week history of a facial swelling on the right side was noted for the first time, giving the initial impression of right sided parotitis. Amongst the investigations carried out, a CT head scan, originally taken for investigation of the new symptoms of dizziness and high calcium levels, revealed a large enhancing mass deep in the right parotid gland. This subsequently led to an assessment by the Ears, Nose and Throat team of the parotid gland where a detailed history of this individual swelling was obtained.

During their time as an inpatient, the patient revealed that they had experienced an episode of parotitis a few months prior to this assessment. This had been treated with Clarithromycin at the time and had improved however some residual swelling had been noted. The most recent swelling,which had been noted during this admission,had been assessed by the patient's general practitioner three days before admission and a course of Clarithromycin and Metronidazole had been commenced. Another new symptom of note was difficulty with mouth opening due to pain, which was first reported at this assessment. Jugular digastrics lymphadeno- pathy was also noted on the right side of the neck.

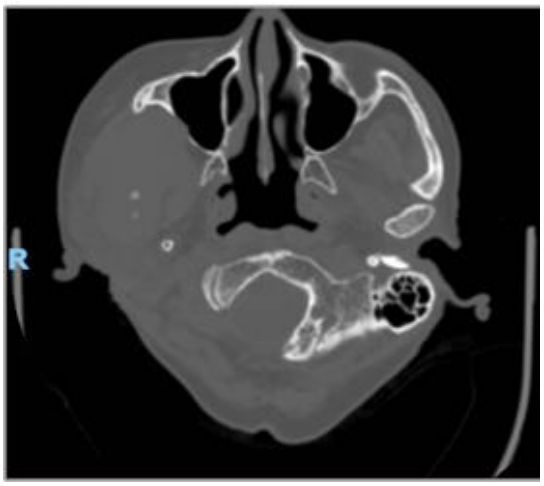

Figure 2._*CT Head with contrast: 52mm x 32mm enhancing mass deep to the right parotid gland and filling the entire right infratemporal fossa. A well circumscribed mass containing coarse calcifications. (Image taken 14-04-22)*

Following this assessment, the patient was reviewed by the oral and maxillofacial team one month later and further information was detailed. This painful right sided facial swelling had been ongoing for approximately four months according to the patient and clinical examination confirmed a right sided fixed facial swelling overlying the right gland area with extension of the mass into the posterior oral cavity with mucosal dehiscence and necrosis. An ultrasound scan with guided fine needle aspiration was carried out which revealed malignant cells.

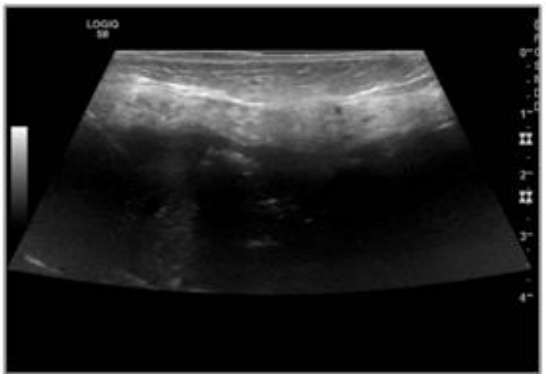

Figure 3._*USS with FNA of neck: The right cheek mass is arising from the right hemimandible(Image taken 16-05-2022)*

This mass was soon identified as a metastasis of the cholangiocarcinoma that was not amenable for surgical resection, but rather would be treated palliatively with the planned chemotherapy that was due to treat the primary mass in the liver. The oncology team confirmed this to be suitable and a plan was made for the patient to be reviewed by the OMFS team in sixmonths.

It was noted that the most recent assessment revealed that the parotid gland lump had reduced considerably and the buccal mucosa fistula on the right side of the oral cavity had closed following chemotherapy and the intra-oral pain had become more manageable. At present, the patient is being seen by the OMFS team on a six-monthly basis for review of the right sided parotid mass and the buccal fistula continues to remain closed.

3. DISCUSSION

Tumours that metastasise from distant regions to the oral and maxillofacial region are noted to be uncommon, comprising of only 1-2% of all malignancies. With regards to cholangiocarcinoma, this type of cancer is difficult to diagnose and has a poor prognosis with five-year survival rates of <5% and a tendency to progress while remaining unseen. [3]

In the cases of the oral region, it is an uncommon site for metastatic spread, whereby if there are findings such as a mass of increasing size, this usually indicates a widespread disease. Approximately 25% of oral metastases have been found to be the first sign of a metastatic spread with 23% being the first indication of an undiscovered malignancy at a distant site. [4]

There are very few reported cases that discuss oral findings of cholangiocarcinoma metastatic spread. Within these cases, the individual sites where any findings such as masses are noted tend to vary. Current case reports discuss bony findings in the mandible and condyles, and soft tissue findings in the floor of the mouth and parotid gland. A recent case report by Mago et al [5] in 2022 discusses a 33-year-old female with diagnosed cholangiocarcinoma who presented with trismus, dysphagia, pain, necrosis and drainage in the mental region in the later stages. Similarly, Watts et al [6] spoke of cholangiocarcinoma in the right sub condyle in a 39-year-old and Harding-Kaba et al [7] discussed a 64-year-old female who presented with painful cheek swelling and an ulcerated lesion in the lower jaw. McClure et al [8] also spoke of single case of left mandibular condyle metastasis, while Tae Min You et al [3] discussed a 54-year-old female who presented with painful swelling in the right mandible and a needle aspiration biopsy and CT scan revealed metastatic cholangiocarcinoma.Of the seven case reports found, these five detail bony lesions in the mandible and condyle.

Two other reports mention soft tissue findings. Nakanishi et al [9] reported a case involving a 77-year-old female, where a metastasis of cholangiocarcinoma was found in the right side of the floor of the mouth, contiguous with the lingual gingival mucosa. This patient noticed a growing mass in the mouth after the initial diagnosis of cholangiocarcinoma was made. Patrocinio et al [10] discussed a 29-year-old female who had a history of trismus for two months prior to admission and after an upper left third molar extraction. Interestingly this case also found a parotid tumour which presented as a metastasis of cholangiocarcinoma and was the first documented case at the time (2008), where the initial presentation was trismus. While cases such as this are rare, perhaps further consideration should be given to long standing episodes of trismus as this symptom was also noted to have been present for our patient in this case. She had described her trismus as being present for four months and this was before the buccal-mucosal fistula had developed.

There are too few reported cases to determine a clear pattern of where oral findings are likely to manifest with cholangiocarcinoma, however in studies conducted where all metastases are considered, majority of oral malignancy is found in the jaw bones, particularly the mandible. Given that such findings of cholangiocarcinoma are rare, it is important that we, as the oral and maxillofacial team take time to consider these potential findings and raise awareness of the importance of assessing the oral cavity throughout treatment for these patients in order to optimise our care.

REFERENCES

[1] Tania M. Welzel and others, Impact of Classification of Hilar Cholangiocarcinomas (KlatskinTumors) on the Incidence of Intra- and Extrahepatic Cholangiocarcinoma in the United States, *JNCI: Journal of the National Cancer Institute*, Volume 98, Issue 12, 21 June 2006, Pages 873–875

[2] Gatto M, Alvaro D. New insights on cholangiocarcinoma. *World J Gastrointest Oncol* 2010; 2(3): 136-145 [PMID: 21160821 DOI: 10.4251/wjgo.v2.i3.136]

[3] You TM, Kim KD, Jeong HG, Park W. **Mandibular metastasis of cholangiocarcinoma: A case report**. Imaging Sci Dent. 2015 Dec;45(4):247-251

[4] Abraham Hirshberg, Anna Shnaiderman-Shapiro, Ilana Kaplan, Rannan Berger, Metastatic tumours to the oral cavity – Pathogenesis and analysis of 673 cases, Oral Oncology, Volume 44, Issue 8, 2008, Pages 743-752, ISSN 1368-8375, 2007.09.012.

[5] Alessandra Kuhn Dall'Magro, Letícia Copatti Dogenski, Patrícia Bade, Larissa Cunha Cé, Eduardo Dall'Magro, João Paulo De Carli, Mandibular metastasis of primary extrahepatic biliary carcinoma: Case report, International Journal of Surgery Case Reports, Volume 98, 2022, 107498, ISSN 2210-2612

[6] Watts, P. Secondary cholangiocarcinoma in the mandible. *Br Dent J* **146**, 385–386 (1979)

[7] Harding-Kaba MB, Delaval C, Lakouichmi M, Jammet P, Goudot P, Yachouh J. Métastasemaxillofaciale d'un cholangiocarcinome [Maxillofacial metastasis of a cholangiocarcinoma]. Rev Stomatol Chir Maxillofac. 2008 Feb;109(1):51-2. French. doi:10.1016/j.stomax.2007.09.005.Epub 2008 Jan 4. PMID: 18177909.

[8] McClure, Shawn A. et al, **Maxillofacial Metastases: A Retrospective Review of One Institution's 15-Year Experience,** Journal of Oral and Maxillofacial Surgery, Volume 71, Issue 1, 178 – 188

[9] Dobashi, Yoh, Nakanishi, Yukihiro, Xu, Bo, LeVea, Charles,(2014), 2014/05/07, Oral Floor and Gingival Metastasis of Cholangiocarcinoma: A Case Report and Review of the Literature, 2090-6781, 10.11 55/2014/712912

[10] Patrocinio LG, Patrocinio TG, Pacheco LF, Patrocinio JA. Trismus as the first manifestation of cholangiocarcinoma. Med Oral Patol Oral Cir Bucal. 2008 Sep 1;13 (9):E573-5. PMID: 18758402.

Citation: *Neena Nayyar & Michael Amin. "A Rare Case Report of Cholangiocarcinoma Metastasising in the Parotid Gland "ARC Journal of Dental Science, vol 6, no. 2, 2023, pp. 6-9. DOI: https://doi.org/10.20431/2456-0030.602002.*