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CASE REPORT

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Malignant melanoma in submandibular gland—A rare diagnostic dilemma

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Abstract

Diagnostic surprises such as melanoma occurring within unusual sites such as submandibular salivary glands are possible. The role of a discerning pathologist with the aid of histochemistry and MDTs is crucial in such circumstances.

KEYWORDS

immunohistochemistry, melanoma, metastasis, submandibular gland

1 | CASE REPORT

Melanoma occurring de novo in salivary glands is very rare and those reported have been in the parotid, while none so in the submandibular glands. We present a case of melanoma within the submandibular gland which created a diagnostic dilemma regarding whether this was a primary submandibular gland melanoma or a metastatic deposit in an intraglandular lymph node.

A 63-year-old gentleman presented with left-sided neck lump, which he noticed 6 months prior to presentation, gradually increasing in size without any pain or fluctuations in size with meal.

On examination in otolaryngology clinic, a 2 × 1 cm lump could be palpated in the submandibular region. There were no other evident neck lumps. Oral cavity and oropharynx were unremarkable on examination and so was flexible endoscopy.

An ultrasound scan that was arranged showed a primarily hypoechoic left submandibular gland lesion with internal heterogeneous echogenicities and increased Doppler signal within the lesion (Figure 1). Subsequent fine-needle aspiration (FNA) was reported as pleomorphic adenoma.

Following this, a left submandibular gland excision was performed with an uneventful postoperative period. The initial histopathology report surprisingly showed features of spindle cell malignant melanoma. Plump spindle cells were arranged in fascicles with entrapped acini and ducts of the salivary gland (Figure 2). Nuclear pleomorphism with often eosinophilic nucleoli and mitotic figures was described. Lesion was well circumscribed and, in most areas bore fibrous capsule. Immunostains SOX-10 (Figure 3), S-100, MART-1, HMB-45, and microphthalmia transcription factor (MITF) requested to confirm the diagnosis came back positive. The adjacent lymph nodes which were excised with the specimen were clear of the disease.

We considered the possibility of either a primary submandibular gland melanoma or a metastatic deposit in an intraglandular node. A positron emission tomography (PET-CT) scan, which was arranged to look for disseminated disease or any occult primary, failed to identify any other lesion. This was then referred to a specialist melanoma multidisciplinary tumour board (MDT). The consensus among the melanoma MDT members was that we should consider this to be a metastatic intraglandular lymph node melanoma deposit overrun by

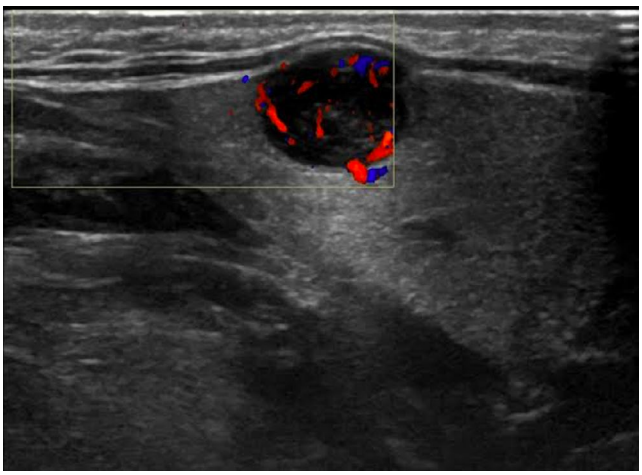

FIGURE 1 Ultrasound scan showing heterogeneous echogenicities in left submandibular gland lesion and increased Doppler signal within lesion

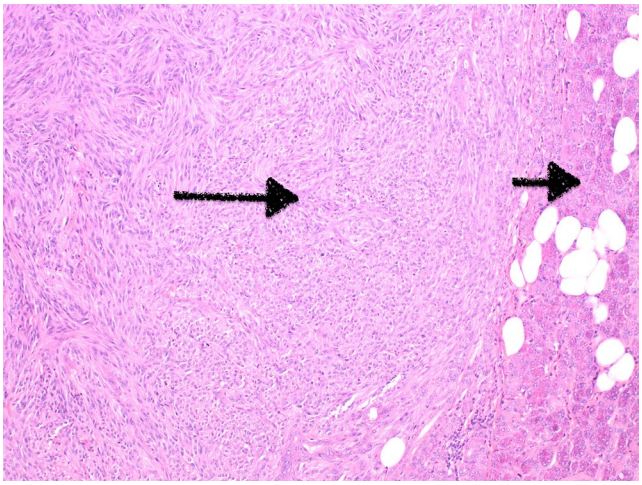

FIGURE 2 The plump spindle cells arranged in fascicles (long arrow) with entrapped acini and ducts of the salivary gland (short arrow) (hematoxylin and eosin, 10×)

malignant cells, since a primary melanoma in a submandibular gland was hitherto not reported and thus considered improbable. He is currently receiving systemic treatment for melanoma.

2 | DISCUSSION

Melanomas in salivary glands are of rare occurrence with all the cases reported involving the parotid, with most being metastatic secondaries. The majority of melanomas involving the parotid gland have been described to be associated with lymph node metastasis in and around the gland from a cutaneous primary in the head and neck region.[1] Furthermore, since melanocytes are not embryologically a part of the salivary glands, the diagnosis of primary melanoma in a salivary gland is quite debatable.

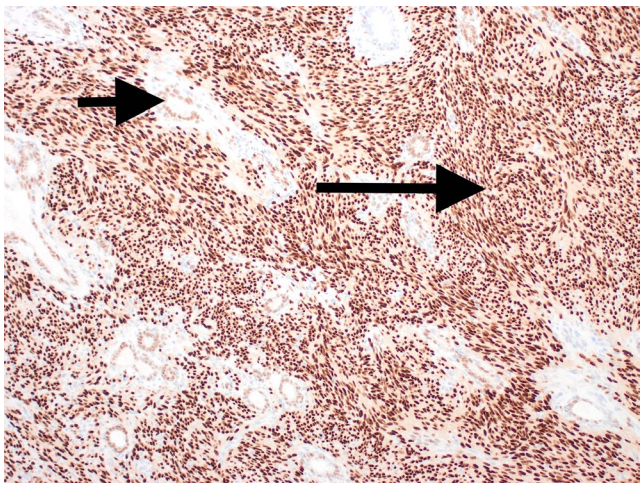

FIGURE 3 Immunostaining with SOX-10 showing normal acini and ducts of salivary gland (short arrow) and SOX-10 positivity in malignant spindle cells (long arrow)

Primary melanoma lesions in submandibular gland have hitherto not been reported. Anatomically, lymph nodes have not been described within the gland. Hence, identification of melanoma in the submandibular gland as in this instance presents a true diagnostic dilemma between a de novo primary and metastatic deposit from a regressed primary elsewhere. Though there are arguments such as the one put forth by Greene and Bernier[2] that melanoblasts can present themselves in the salivary glands due to their descent along the downgrowth of the oral epithelium into the salivary glands, which make the diagnosis of primary salivary gland melanoma theoretically possible, the probability of a metastatic deposit seems more plausible.

Metastatic deposits of melanoma from unknown primaries account for about 3% of all melanomas with two-thirds of these being in the lymph nodes.[3] These instances could be due to factors such as a spontaneously regressed melanoma primary[4] or a previously excised lesion with a missed diagnosis on histology among others. Regression may be seen in about 10%-35% of melanomas.[5] Hence, the role of a detailed history regarding previous skin and mucosal lesions coupled with a diligent examination of the head and neck along with a general examination to exclude any primary cannot be more emphasized.

A case with similar circumstances as in our present one was described previously by Agarwal et al.[6] A submandibular lump initially suspected to be a submandibular gland neoplasm was operated upon which revealed a subcutaneous metastatic melanoma deposit while the submandibular gland itself was found to be free of tumor and the primary remained unknown. A patient with melanoma within a submandibular gland pleomorphic adenoma was described by Cochrane and Kenny.[7] This patient did have multiple skin moles and a melanotic lesion on the right forearm, and hence was possibly a

metastatic lesion. However, in our patient the melanoma cells were seen within the submandibular gland while the adjacent lymph nodes were free from the tumor.

A previous study has suggested that there are no lymph nodes within the submandibular glands,[8] and hence, we had to consider the possibility of primary submandibular gland melanoma. Thus, this presents a unique and rare diagnostic dilemma of its kind. The specialist melanoma MDT considered both possibilities and eventually decided to treat it as a metastatic intraglandular lymph node melanoma deposit overrun by malignant cells. This was because, the MDT reasoned that, in a balance of probabilities, possibility of this being metastatic intraglandular lymph nodes was greater than a primary melanoma lesion which is hitherto unknown within the submandibular gland. Also, an assumption was made that the metastatic disease was limited only to the intraglandular lymph nodes, since the surrounding extraglandular lymph nodes were free from the tumor.

In conclusion, as demonstrated in our patient, melanoma can appear similar to other lesions both clinically and histologically. A fair degree of suspicion aided by immunohistochemical studies with markers such as S-100, HMB-45, MITF, SOX-10, and MART-1, which remain the most important tools for distinguishing melanomas from other tumors,[9] can often achieve a histological diagnosis despite a very uncommon presentation such as in our patient.

ACKNOWLEDGMENT

Published with written consent of the patient.

CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

SGK: collected patient data, drafted the manuscript, performed literature search, managed correspondence, and revised the manuscript. SARS: collected patient data, collected necessary images, and helped author writing the manuscript. SCJ: clinically responsible for the patient, operating surgeon, represented the team in the MDT, supervised and guided the drafting, and contributed to revision of the manuscript. RCH: supplied necessary data relating to histopathology, and provided the relevant histology images and description.

ETHICAL STANDARDS

All authors (Srinish Gopala Krishnan, Ali Sherazi, Sharan Chakkyath Jayaram, and Rachel Hall) undertake that this case report complies with accepted Ethical standards as applicable.

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REFERENCES

1. Prayson RA, Sebek BA. Parotid gland malignant melanomas. *Arch Pathol Lab Med*. 2000;124(12):1780-1784.
2. Greene GW Jr, Bernier JL. Primary malignant melanomas of the parotid gland. *Oral Surg Oral Med Oral Pathol*. 1961;14:108-116.
3. Ribero S, Pampena R, Bataille V, et al. Unknown primary melanoma: worldwide survey on clinical management. *Dermatology*. 2016;232(6):704-707.
4. Smith JL Jr, Stehlin JS Jr. Spontaneous regression of primary malignant melanomas with regional metastases. *Cancer*. 1965;18(11):1399-1415.
5. Kılıçkaya MM, Aynali G, Ceyhan AM, Çiriş M. Metastatic malignant melanoma of parotid gland with a regressed primary tumor. *Case Rep Otolaryngol*. 2016;2016:5393404.
6. Agarwal K, Jain S, Dass J, Andley M. Malignant melanoma of unknown primary site simulating a salivary gland neoplasm. *Turk Patoloji Derg*. 2011;27(1):77-79.
7. Cochrane HR, Kenny RA. Metastatic malignant melanoma in a submandibular pleomorphic salivary adenoma. *Histopathology*. 1993;23(5):483-484.
8. Fives C, Feeley L, Sadadcharam M, O'Leary G, Sheahan P. Incidence of intraglandular lymph nodes within submandibular gland, and involvement by floor of mouth cancer. *Eur Arch Otorhinolaryngol*. 2017;274(1):461-466.
9. Ohsie SJ, Sarantopoulos GP, Cochran AJ, Binder SW. Immunohistochemical characteristics of melanoma. *J Cutan Pathol*. 2008;35(5):433-444.

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