

## Short communication

# Ectopic parathyroid adenoma in the submandibular region: a case report

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## Abstract

Ectopic parathyroid adenomas that affect the submandibular region have not been widely reported. We describe a 34-year-old man who presented with a painless swelling of the submandibular region. The identification of hypercalcaemia encouraged us to engage a multidisciplinary team to evaluate further serum changes. Parathyroid hormone analysis, <sup>99m</sup>Tc-methoxy-isobutyl-isonitrile (<sup>99m</sup>Tc-MIBI) scintigraphy, and single-photon emission computed tomography (SPECT-CT) were done to rule out hyperparathyroidism. Raised parathyroid hormone together with <sup>99m</sup>Tc-MIBI and SPECT-CT examination were consistent with a tumour caused by the hyperparathyroidism. Removal of the lesion resulted in rapid improvement in serum calcium and parathyroid hormone, and the normalisation of the serum creatinine, concentrations. Histopathological analysis confirmed a parathyroid adenoma. We conclude that ectopic parathyroid adenomas should be considered as part of a differential diagnosis for tumours of the submandibular region.

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**Keywords:** ectopic parathyroid adenoma; mandibular region; hypercalcaemia.

## Introduction

Ectopic parathyroid adenomas occur in anomalous anatomical locations such as the mediastinum and retropharyngeal region, but cases that affect the submandibular region have not been well described.<sup>1,2</sup> Ectopic parathyroid adenomas are commonly diagnosed using parathyroid scintigraphy with <sup>99m</sup>Tc-methoxy-isobutyliso-nitrile (<sup>99m</sup>Tc-MIBI) in com-

ination with ultrasonography or single-photon emission computed tomography (SPECT-CT) of the neck.<sup>3</sup> We present the case of a 34-year-old man who was diagnosed with an ectopic parathyroid adenoma in the submandibular region based on histopathological results.

## Case report

A 34-year-old Chinese man presented with a painless swelling in the submandibular region that he had had for two months. Preoperative laboratory tests showed a raised serum concentration of calcium at 4.96 mmol/L (normal range 2.11–2.52 mmol/L). The serum creatinine concentration (298 mmol/L) was also raised (normal range 31–133 mmol/L). According to the suggestions of the multidisciplinary team, we tested for hyperparathyroidism. Parathyroid hormone was also high (1073 pg/ml (normal

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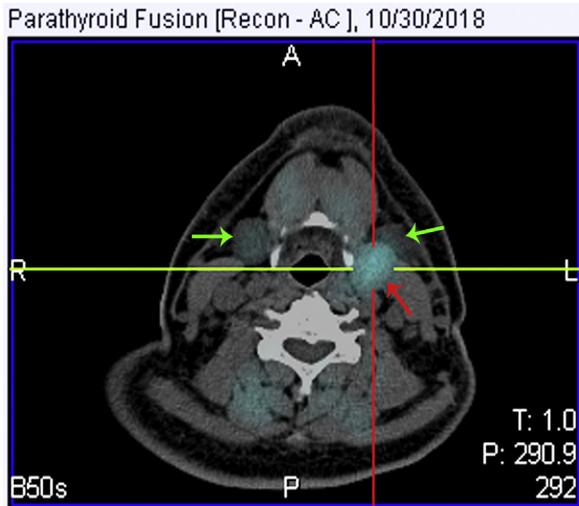


Fig. 1. Preoperative radiographic assessment of ectopic parathyroid adenoma. The fused  $^{99m}\text{Tc}$ -MIBI and SPECT-CT images show an accurate localisation of the mass on the medial-posterior aspect of the left submandibular gland. (the adenoma is shown by the red arrow and the submandibular gland is indicated by the green arrow).

range 15–65 pg/ml). CT of the neck showed a well-defined, heterogeneous mass in the left submandibular region that measured  $36 \times 21 \times 26$  mm.  $^{99m}\text{Tc}$ -MIBI scintigraphy showed focal activity in the left submandibular region at 15 minutes, and rapid  $^{99m}\text{Tc}$ -MIBI washout after three hours. The fused  $^{99m}\text{Tc}$ -MIBI and SPECT-CT images helped to locate the mass accurately on the medioposterior aspect of the left submandibular gland, and we excised it (Fig. 1). Histopathological examination of the specimen provided a final diagnosis of ectopic parathyroid adenoma. Three months after discharge, the patients' serum calcium and parathyroid hormone concentrations were within normal limits once more (Fig. 2).

## Discussion

Cases of primary hyperparathyroidism secondary to ectopic parathyroid adenoma have been reported,<sup>4</sup> along with some in unusual locations such as the pyriform sinus and soft palate.<sup>5,6</sup>

Patients with submandibular tumours often present initially to a doctor associated with oral and maxillofacial surgery, who could fail to consider ectopic parathyroid adenoma as part of the differential diagnosis for a submandibular tumour.

A combination of imaging methods should be used for detection of these lesions. As shown in this case, the fusion of SPECT-CT and  $^{99m}\text{Tc}$ -MIBI images provided accurate and direct localisation for the excision. Complete ablation is the gold standard for the management of such hyperfunctioning adenomas. This should be done regardless of symptoms because patients who have meaningful changes in markers related to hyperparathyroidism may be more prone to pro-

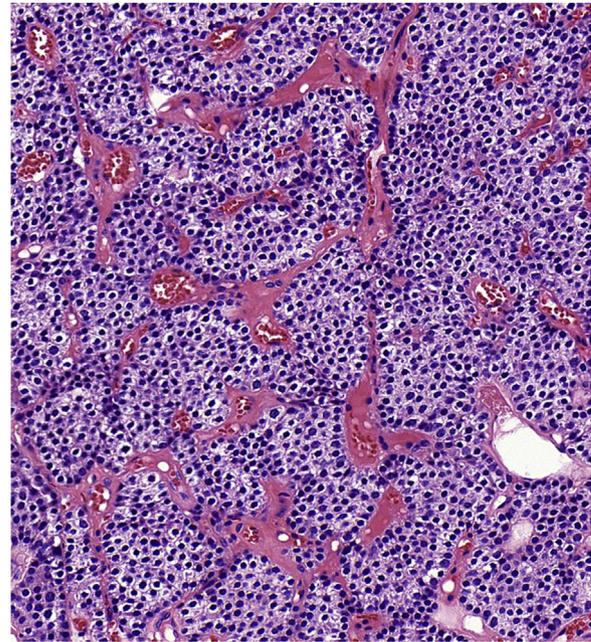


Fig. 2. Histopathological examination confirmed the final diagnosis of parathyroid adenoma (haematoxylin and eosin stain, original magnification  $\times 200$ ).

gressive disease, even with no symptoms, as was seen in this case.<sup>7</sup> Postoperatively, hypocalcaemia should also be treated regardless of the patient's symptoms because uncontrolled hypocalcaemia can lead to life-threatening cardiac arrhythmias postoperatively.<sup>8</sup>

## Conflict of interest

We have no conflicts of interest.

## Ethics approval and patient's permission

Ethics approval for this case was not required according to the guidelines stated by the Research Ethics Board of Qingdao University. The patient's consent was obtained.

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## Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:<https://doi.org/10.1016/j.bjoms.2019.10.296>.

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