PULMONARY PUZZLES

Forty-two-year-old woman with incidental bilateral nodular opacities on chest X-ray

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A 42-year-old Filipino woman was referred to a respiratory clinic after incidental bilateral nodular opacities were discovered on a routine immigration chest X-ray (figure 1). She reported a 5-month history of intermittent dry cough, but no haemoptysis, chest pain, breathlessness or weight loss. She was systemically well. She has a medical history significant for ameloblastoma, a benign tumour of the jaw, which was excised in 2004, and previous ovarian chocolate cyst in 2002 that was complicated by the need for ureteric stenting. She is a lifelong non-smoker. She immigrated to New Zealand on a work visa in 2016, and works full time in a supermarket.

Clinical examination revealed a body mass index of 28. There was no evidence of jaundice, anaemia, cyanosis or clubbing and she had no palpable lymphadenopathy. Her chest was resonant to percussion and clear on auscultation. She had a large scar on the left side of her abdomen, which was otherwise soft with no organomegaly. She had no peripheral oedema.

She had a normal full blood count, coagulation screen, calcium level and alpha-fetoprotein. She had mild renal impairment with a creatinine of 126 µmol/L (estimated glomerular filtration rate 45 mL/min/1.73 m²).

Spirometry was within normal limits showing an FEV₁ of 2.02 L, 90% predicted.

A CT scan showed numerous bilateral cannonball lung lesions but no mediastinal lymphadenopathy (figure 2). Bronchoscopy was suspicious for extrinsic compression at the right upper lobe. Bronchial washings were obtained. There were no organisms on culture and microscopy, and no malignant cells on cytology.

QUESTION
What is the diagnosis and what is the management of this condition?

ANSWER
The CT shows multiple lesions suspicious for metastatic malignancy. Core biopsy revealed metastatic ameloblastoma.

Ameloblastomas are rare, benign odontogenic tumours that classically occur in the mandible. They are locally aggressive with a propensity to recur despite resection. Only rarely, in approximately 2% of cases, do they metastasise. Metastatic ameloblastomas have a benign histological appearance no different from those that have not metastasised in contrast to ameloblastic carcinomas characterised by cytological atypia.

Though not well understood, factors associated with the likelihood of developing metastatic disease include extent and duration of the primary tumour, local recurrence and multiple surgeries. The lung is involved in more than 80% of cases. Other sites of metastasis include lymph nodes, pleura, vertebra, skull, diaphragm, liver and the parotid gland.

The disease-free interval between initial diagnosis and appearance of metastases averages 10–12
years. The median survival time with metastatic disease is 3 months to 5 years while the longest reported survival is 37 years. It is generally agreed that prevention with early and complete resection of the primary tumour is optimal. There is less consensus regarding treatment of metastatic lesions. Where amenable, surgical resection of metastatic lesions may be effective. Chemotherapy and radiotherapy may also play a role, particularly in palliation of symptoms.

Our patient had local resection of her ameloblastoma in 2004 with no adjuvant therapy at the time. She is currently asymptomatic from her metastases and will be managed conservatively with regular surveillance.

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