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Background

Sarcoidosis is a chronic granulomatous disease of unknown origin with clinical manifestations(表現) being highly variable between affected patients. An exaggerated(誇張) immune response without distinct stimulus is characteristic for sarcoidosis. So far, no specific environment trigger could be identified which leads to this obscure(模糊) disease. The lungs are most commonly affected with more than 90% of the patients having pulmonary involvement. Bony lesions usually occur bilateral and are characterized by an osteolytic zone, while the cortical part remains intact.

The clinical appearance is highly variable, involving sustained cough, erythema nodosum(結節性紅斑), periarticular inflammation, fever and occurring fatigue(疲勞). The diagnosis is based on three pillars: (1) clinical assessment, (2) radiographic signs and (3) histological evidence of non-caseating granulomas. Therapy consists of immunosuppressive treatment with oral glucocorticoids being the standard first-line treatment.

Case presentation

A 74 year old European female patient approached her dentist due to pain in the right lower jaw. A panoramic X-ray was performed, presenting an osteolysis of unknown origin in the mandible on both sides, right more than left.



The patient was then referred to the clinic of cranio-maxillo-facial surgery for further evaluation. She reported pain in the area of the right angle of the mandible for several months; additionally, pain in the right calcaneus and in both forearms has been noted. She denied fever and weight loss, but reported cough and dyspnea(呼吸困難) on exertion(用力). Furthermore, a sicca(結膜乾燥症)-symptomatic in both eyes and the oral cavity was present. The patient was diagnosed with hypertensive cardiac disease, rhytmogene cardiopathy(心臟病), hyperlipidaemia, cutaneous psoriasis(牛皮 mathered mathematical mathe spironolactone, and simvastatine.

At the day of first presentation there were no signs for locale infection on the oral and extraoral examination, and no palpable lymph nodes in the cervical region. Only the tooth 47 showed percussion sensitivity and was not sensitive to cold. There were no abnormal laboratory findings, specifically normal C-reactive protein, serum calcium and alkaline(鹼性) phosphatase.

A computed tomography with 3D reconstruction of the osteolysis was performed. The radiography showed loosening of cancellous(網肤骨) bone with extension of the mandibular canal and partially interrupted cortical bone on the right side. The findings were suspicious for osteomyelitis or a malignant lesion. Additional Single Photon Emission(排放) Computed Tomography (SPECT-CT) examination was completed presenting increased bone metabolism in the area of the vertex and the jaw angles of both sides, right more than left (Fig. 2). Further increased metabolism was detected in both forearms and in the right calcaneus.



A biopsy of the alveolar process of the lower jaw was carried out for further investigation of the unknown osteolysis together with the removal of the second lower molar (Fig. 3). Histology revealed chronic

granulomatous inflammation with non-caseating epitheloid granuloma, consistent with sarcoidosis. Ziehl-Neelsen's staining was negative and mycobacterial(分枝桿菌) culture showed no growth. Fungal and other bacterial infections were excluded. To confirm the suspicion of sarcoidosis further examinations were initiated: The CT-Scan of the lung showed enlarged intrathoracic lymph nodes, and pulmonary function testing revealed a restriction(限制) of diffusion capacity of carbon monoxide. Based on a recently developed vertigo(眩暈) a MRI scan of the brain and the spinal cord was performed, showing a suspicious lesion in the area of the vertex compatible(兼容) with neuro-sarcoidosis. Levels of interleukin-2-receptor and angiotensin(血管緊張素)-converting enzyme were in the range of normal. Finally, biopsy of an additional skin lesion in the area of the medial canthus(眼角) of the left eye histologically confirmed the diagnosis of sarcoidosis.

Due to relevant multi-organ disease with symptomatic bone involvement and strong suspicion of neurosarcoidosis, immunosuppressive therapy with azathioprine was installed. A follow-up SPECT-CT scan after six months of treatment revealed regressive(何退) bone metabolism of all previously described lesions, and no new

lesions were detected. The patient was free of pain and the immunosuppressive treatment was well tolerated.

A couple of differential diagnosis should be considered when evaluating intrabony lesions of the jaws, in particular of the lower jaw: osteomyelitis, metastatic tumors, multiple myeloma(多發性骨髓瘤), Langerhans histiocytosis(細胞增生). All these findings may present a similar radiographic image and could be confounded(困惑) with a sarcoidotic lesion and had therefore to be considered in our patient. Conclusions

The first symptoms of multi-organ sarcoidosis can be variable and symptomatic bone involvement is a rare presentation of this disease. Histologic confirmation is mandatory, followed by further diagnostic assessments with regard to other organ involvement. This case is of utmost clinical importance as it demonstrates that an accurate anamnesis(病 \mathbb{E}) and clinical and radiological examination are required for an early diagnosis of this rare disease.

題號	題目	
1	Sarcoidosis is not	
	(A) Well-known cause	
	(B) Formation of noncaseating granulomatous inflammation	
	(C) most commonly appears acutely over a period of days to weeks	
	(D) Common clinical symptoms include dyspnea, dry cough, chest pain,	
	fever, malaise, fatigue, arthralgia, and weight loss	
答案	出處: oral and maxillofacial pathology, third edition	
(A)		
題號	題目	
2	Sarcoidosis's predominant site is less on	
	(A) Lungs	
	(B) Lymph nodes, salivary gland	
	(C) Skin, eyes	
	(D) Bone	
答案	出處: oral and maxillofacial pathology, third edition	
(D)		