CASE REPORT

Calcification of the external carotid arteries and their branches

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This patient had longstanding hypercalcaemia and hyperphosphataemia owing to chronic renal disease, then finally failure, inducing tertiary hyperparathyroidism. He also had long histories of diabetes mellitus type II, hypertension and hypercholesterolaemia. He then reported a painful expansile swelling of the anterior mandible which was diagnosed as a “brown tumour”. Subsequent review of the CT data set by an oral and maxillofacial radiologist revealed two patterns of calcification of the carotid arteries. A pipestem pattern was observed bilaterally along almost the entire lengths of the external carotid artery, a muscular artery, and its branches whereas plaque-like calcification was observed in the common and internal carotid arteries (elastic arteries). The pipestem pattern, hitherto an unreported feature affecting the external carotid artery, may represent a metastatic calcified deposit owing to hypercalcaemia and hyperphosphataemia in the tunica media of muscular arteries, resulting in arteriosclerosis, which maintains a patent lumen. The plaque-like pattern is representative of lumen-occluding calcified atherosclerosis associated with the long histories of diabetes mellitus type II, hypertension and hypercholesterolaemia. As this patient did not have any symptoms and/or signs of myofacial pain, facial dysfunction or numbness, the calcification of his external carotid arteries and branches were considered as arteriosclerosis. The brown tumour responded to the parathyroidectomy and the renal transplant.

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Introduction

Calcification of arteries is common in ageing, diabetes, dyslipidaemia, genetic disease and disturbances of calcium metabolism, *e.g.* hypercalcaemia and hyperphosphataemia.¹ There are two types of calcification of the arteries depending upon whether the tunica media or tunica intima is involved. The former is termed medial calcific sclerosis or Monckeberg's arteriosclerosis. The calcification of the tunica intima, particularly of the larger elastic arteries proximal to the heart, is associated with narrowing of the lumen (stenosis) and atheroma formation. When the carotid artery is so affected, it is called calcified carotid atheroma (CCAA). CCAA particularly affects the larger elastic arteries proximal to the heart,¹ such as the common carotid. The former's synonym, Monckeberg's arteriosclerosis,

implies vessel hardening and may be radiologically apparent as a “pipestem” or “rail-track” radio-opaque pattern in the more distal and muscular arteries.¹ This pattern is due to “metastatic calcification” that occurs within otherwise normal tissue in contrast with “dystrophic calcification”, which arises within abnormal tissue such as the atheromatous plaques of CCAA. Although Monckeberg's arteriosclerosis is supposed to be benign compared with CCAA in outcome, it can be associated with important medical conditions such as osteoporosis and hyperparathyroidism. Although most commonly observed in the limbs, it has hitherto not been reported in the head and neck. This is a report of such a case.

Case report

Table 1 shows that this 64-year-old male had a long-standing situation of hypercalcaemia and hyperphos-

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phataemia owing to chronic kidney disease (CKD) before the diagnosis of tertiary hyperparathyroidism, which was treated recently by parathyroidectomy. The other medical conditions and their treatments do not contribute to the manifestations in the present case. He presented with a painful, progressive expansile lesion at the anterior edentulous mandible of 1 year's duration. His extensive and relevant medical history and treatment are set out in Table 1. There was no relevant family history that would contribute to his condition.

A well-defined radiolucency in the anterior edentulous mandible was observed on the panoramic radiograph. Subsequent nuclear medicine investigation (^{99m}Tc methylene diphosphonate) reported a high uptake at the site of the lesion. This patient was further investigated using CT. The CT bone windows revealed a well-defined, unilocular lucency in the anterior sextant of the mandible which was edentulous (Figure 1). This lesion, measuring 3.34×1.92 cm, extended from the junction between the right canine and lateral incisor to that between the left canine and first premolar. The labial (or buccal) cortex was substantially expanded and eroded. Furthermore, the cortex appeared absent in places. The lingual cortex to the left of the midline was eroded and slightly displaced lingually.

The lesion was biopsied and the histopathological examination revealed a typical central giant cell lesion, which showed numerous multinucleated giant cells in a cellular fibroblastic background with red blood cell extravasations and obvious cellular woven bone. A diagnosis of tertiary hyperparathyroidism-induced brown tumour was consistent with a longstanding history of renal failure.

The laboratory test results 6 months after the parathyroidectomy and 4 months prior to the renal transplant revealed that alkaline phosphatase was now normal (88 units l^{-1} ; normal 40–110 units l^{-1}) but that the kidney function tests were abnormal. The estimated glomerular filtration rate was consistent with a diagnosis of chronic renal failure as it was 5 ml min^{-1} (normal rate is $>60 \text{ ml min}^{-1}$). Urea and creatine were also elevated; 11 mmol l^{-1} (normal 2–9 mmol l^{-1}) and 853 $\mu\text{mol l}^{-1}$ (normal 45–110 $\mu\text{mol l}^{-1}$), respectively.

It was at this time that the CT data set was reviewed by an oral and maxillofacial radiologist, revealing that almost all major branches of bilateral external carotid arteries exhibited pipestem calcification (Figure 2). These are the external carotid itself, the suprahyoid branch of the lingual artery, the facial artery, the internal maxillary artery, the transverse facial artery and the superficial temporal artery. The pattern of calcification exhibited in the common and proximal part of the internal carotid arteries was plaque-like rather than pipestem.

Shortly after the biopsy and this observation on the CT, the patient received a renal transplant which restored him to normal renal function. 4 months after the transplant the brown tumour had sufficiently regressed so the patient could wear his lower denture again. The patient still did not display numbness of the face and head or myalgia, or dysfunction of the muscles of mastication or facial expression.

Discussion

Although brown tumours commonly affect the long bones of the limbs, ribs and pelvis, they rarely affect the jaws.² Nevertheless, a brown tumour affecting the jaws was reported in 8% of patients with CKD who were on dialysis.³ The increasing prevalence globally of chronic renal disease² is likely to be accompanied by an increase in brown tumours overall and thereby an increase in the number of them affecting the jaws. Recently, Selvi *et al*⁴ reported a case of brown tumours affecting the jaws in a patient exhibiting a similar history of CKD and its progression to renal failure and tertiary hyperparathyroidism, as in the present case. The brown tumour in the present case had regressed sufficiently for the patient to resume normal denture wear. This reinforced Selvi *et al*'s⁴ recommendation that parathyroidectomy should be first countenanced as treatment for brown tumour. Excision or recontouring surgery of the brown tumour itself should be reserved for large tumours which have produced facial asymmetry and slowly regress.⁴

Table 1 Medical history and medications collected at the time of the investigations of the brown tumour undertaken after the parathyroidectomy and before the renal transplant

<i>Disease</i>	<i>Associated with hypercalcaemia and/or hyperphosphataemia</i>	<i>Present treatment</i>	<i>Does this treat hypercalcaemia and/or hyperphosphataemia?</i>
Diabetes mellitus type II for about 20 years	No	Insulin injection	No
Hypertension for about 12 years and coronary heart disease for 3 years	No	Metoprolol, minoxidil, irbesartan	No
Hypercholesterolaemia for 10 years	No	Atorvastatin	No
Constipation for about 2 years	No	Sennoside, docusate	No
Bone pain for about 2 years	Yes	Gabapentin	No
Tertiary hyperparathyroidism diagnosed 5 months earlier	Yes	Present total parathyroidectomy/partial autotransplantation	Yes
Chronic kidney disease for 10 years and renal failure for 3 years	Yes	Haemodialysis for 2 years, Renagel (phosphate binder) for 2 years Renal transplant ^a	Yes

^aRenal transplant was performed 8 months after definitive diagnosis of the brown tumour and almost a year after the parathyroidectomy

Figure 1 A CT axial section (bone window) at the level of the second cervical vertebra. The radiolucency is well defined. The labial (buccal) expansion is substantial. The labial cortex is not only eroded but appears to be discontinuous, which is suggestive of perforation. The lingual cortex is slightly displaced and eroded but is continuous. The parts of the course of facial arteries, exhibiting a calcified “rail-track” pattern, are obvious buccal and lingual to the mandible. There is only a short arc of dense calcification in association with an internal carotid artery

Although it appears that a pipestem pattern of calcification of the branches of the external carotid have not been hitherto reported, opacification of some of these branches was observed in Selvi *et al*’s Figure 4 and Figure 5.⁴ The differences were that not only did these arteries display no lumen but so did the ophthalmic artery, a branch of the internal carotid artery. Selvi *et al*’s Figure 4 revealed that intravenous contrast had been used, thus accounting for the opacification of these arteries.

The presentation of the calcified arteries in the present report is consistent with Monckeberg’s arteriosclerosis. Nevertheless, the definitive diagnosis of whether the calcification occurs within the intima, media or both rests upon the histopathology. Although temporal artery biopsy (TAB) is the standard investigation for giant cell arteritis,⁵ there was no longer any pressing clinical indication for TAB in the present case, whose CKD had been successfully treated by renal transplant. The brown tumour had already regressed sufficiently 4 months after the renal transplant (and more than a year after the parathyroidectomy) so that he could wear his lower denture again. Furthermore, there was no altered sensation or function within the area supplied by the external carotid artery. This, in absence of a TAB, suggests the blood supply to the face had not been compromised by the calcification.

Figure 2 A CT three-dimensional reconstruction in the coronal plane displays the extent of calcification of the carotid arteries. Note that the coronal plane has been slightly rotated to display better most of the calcified arterial branches at least on one side. The lower half of the patient’s face has been displayed in semi-profile. Although the mandible, maxilla and base of the skull were excluded so as to optimally display these arteries, the hyoid bone, styloid processes, the superior horns and the superior border of the lamina of the calcified thyroid cartilage and the anterior arch of the first cervical vertebra were retained in order to facilitate orientation. The pipestem pattern of calcification of external carotid arteries and its branches and the plaque-like denser opacification of the common and internal carotid arteries are obvious. The pipestem calcification of the branches of the external carotid arteries continues into those arteries which supply the nasal cavity and are observed above the markers for internal maxillary artery (MA) and transverse facial artery (TFA). C1, anterior arch of the first cervical vertebra; CCA, common carotid artery; FA, facial artery; HB, hyoid bone; ICC, internal carotid artery; SH, suprahyoid branch of the lingual artery; SP, styloid process; STA, superficial temporal artery; TC, superior horn of the thyroid cartilage

Although this may suggest that the lumen had been kept open by the rigid tunica media, which is expected of arteriosclerosis,⁶ it may also mean that the lumen has so far not been appreciably reduced in diameter if it were also affected by atherosclerosis. Claudication of stenotic external carotid arteries caused by atherosclerosis has been recently reported.⁷ A reason for such a consideration is arteriosclerosis, as classically defined by Monckeberg as wholly confined to the tunica media, which may not be strictly so in all cases. Although Fishbein *et al*⁸ reported that 60% of publications supported the contention that Monckeberg’s arteriosclerosis is not accompanied by calcification of the internal elastic lamina (the outer layer of the tunica intima), McCullough *et al*⁹ state that Monckeberg’s arteriosclerosis is a “pathologic misconception”. They add that “the published histological and clinical data suggest that Monckeberg’s (arterio) sclerosis describes advanced calcific atherosclerosis as the common and important pathobiology that is present in patients with

CKD".⁹ Nevertheless, Drueke¹⁰ and Amann¹ urged restraint and careful consideration before overthrowing the existing paradigm that calcification of the media and the intima is different because the case for its continued retention is very persuasive.

Conclusions

- (1) This was a hitherto longstanding case with hypercalcaemia and hyperphosphataemia prior to the parathyroidectomy, after which the brown tumour was definitively histopathologically diagnosed. 8 months after this biopsy, the underlying CKD was definitively treated by renal transplant. 4 months after this successful transplant, the brown tumour had regressed sufficiently for the patient to recommence wearing his lower denture.
- (2) Although this case displays the plaque-like calcification of the common and internal carotid

arteries which is expected of atherosclerosis and the pipestem-like calcification of the external carotid artery associated with arteriosclerosis, in the absence of histopathology, no definitive comment can be made as to whether the latter process was solely at work in the external carotid artery.

- (3) Regardless of the probable histological outcome raised in the last point, with reference to whether the tunica intima of the external carotid artery is affected or not, the atheroma-indicating plaque-like calcifications in this patient's common and internal carotid arteries indicate an increased risk of a cardiovascular accident.
- (4) Although this is the first reported finding of calcification of the external carotid artery and its branches incidental to a CT investigation for a brown tumour, it may not be unique. Therefore, the entire CT data set should be routinely reviewed for hitherto undetected incidental findings.

References

1. Amann K. Media calcification and intima calcification are distinct entities in chronic kidney disease. *Clin J Am Soc Nephrol* 2008; **3**: 1599–1605.
2. Tarrass F, Benjelloun M, Bensaha T. Severe jaw enlargement associated with uremic hyperparathyroidism. *Hemodial Int* 2008; **12**: 316–318.
3. Scutellari PN, Orzincolo C, Bedani PL, Romano C. Radiographic manifestations in teeth and jaws in chronic kidney insufficiency. *Radiol Med* 1996; **92**: 415–420 (in Italian).
4. Selvi F, Cakarar S, Tanakol R, Guler SD, Keskin C. Brown tumour of the maxilla and mandible: a rare complication of tertiary hyperparathyroidism. *Dentomaxillofac Radiol* 2009; **38**: 53–58.
5. Rieck KL, Kermani TA, Thomsen KM, Harmsen WS, Karban MJ, Warrington KJ. Evaluation for clinical predictors of positive temporal artery biopsy in giant cell arteritis. *J Oral Maxillofac Surg* 2011; **69**: 36–40.
6. Couri CE, da Silva GA, Martinez JA, Pereira Fde A, de Paula FJ. Mönckeberg's sclerosis—is the artery the only target of calcification? *BMC Cardiovasc Disord* 2005; **5**: 34. Available from: <http://www.biomedcentral.com/1471-2261/5/34>
7. Schiller A, Schwarz U, Schuknecht B, Mayer D, Hess K, Baumgartner RW. Successful treatment of cold-induced neck pain and jaw claudication with revascularization of severe atherosclerotic external carotid artery stenoses. *J Endovasc Ther* 2007; **14**: 304–306.
8. Fishbein GA, Fishbein MC. Arteriosclerosis: rethinking the current classification. *Arch Pathol Lab Med* 2009; **133**: 1309–1316.
9. McCullough PA, Agrawal V, Danielewicz E, Abela GS. Accelerated atherosclerotic calcification and Monckeberg's sclerosis: a continuum of advanced vascular pathology in chronic kidney disease. *Clin J Am Soc Nephrol* 2008; **3**: 1585–1598.
10. Drüeke TB. Arterial intima and media calcification: distinct entities with different pathogenesis or all the same? *Clin J Am Soc Nephrol* 2008; **3**: 1583–1584.