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CASE REPORT

Multiple spontaneous tendon ruptures from enthesis failure in primary hyperparathyroidism: a case report and review of imaging findings

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Abstract
Cases of spontaneous tendon ruptures have been previously reported in the literature. Although both renal failure and hyperparathyroidism have been implicated as separate aetiologies, their frequent co-existence has confounded analysis. We report a case of a young man with primary hyperparathyroidism presenting with multiple acute spontaneous tendon ruptures. This case affords an opportunity to evaluate the imaging features of acute spontaneous tendon ruptures in the context of primary hyperparathyroidism, and in conjunction with a review of previous literature reports, an imaging-based hypothesis on the pathophysiology and aetiology of spontaneous tendon ruptures is proposed.

Keywords Primary hyperparathyroidism · Spontaneous tendon rupture · Enthesis failure

Introduction

Spontaneous tendon ruptures, although not a common occurrence, have been previously reported in the literature [1–6]. A range of aetiological factors have been implicated [7–11]. Two of these proposed aetiologies (hyperparathyroidism and chronic renal failure) frequently co-exist in subjects. Recent reports have emphasised hyperparathyroidism as being the main pathological mechanism leading to tendon rupture in chronic renal failure, as will be discussed.

Review of the existing literature reveals no primarily imaging-focused treatment of this topic. This current case of primary hyperparathyroidism presenting with multiple tendon ruptures affords an opportunity to analyse the pattern of tendon failure in primary hyperparathyroidism, compared with that in chronic renal failure. Based on this, an imaging-based hypothesis of the pathophysiology and aetiology of spontaneous tendon ruptures in the context of hyperparathyroidism and chronic renal failure is proposed.

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Case report

A 27-year-old man with no significant past medical history presented to the emergency department with bilateral knee and left elbow pain following a fall 2 days earlier. This occurred while he was alighting from a bus and felt both knees spontaneously "give way", landing on his back and left elbow. He felt a popping sensation in both knees and left elbow. There was no significant occupational history, recent injury or strenuous activity.

On clinical examination, the patient was unable to flex or extend either knee because of pain, but could move his ankles and toes freely. He was unable to weight-bear because of pain. Neurovascular status of both lower limbs was otherwise intact.

Radiographs of both knees and left elbow were performed. Initial assessment showed mild abnormal anterior angulation of the left patella, and superior subluxation of the right patella with prominent soft-tissue swelling along the anterior aspect of the right knee. Flaky amorphous radio-opaque densities were seen in the soft tissues superior to the left patella, inferior to the right patella and posterior to the left elbow joint (Figs. 1, 2, and 3).

Initial blood investigations revealed normal renal and liver function tests. Subsequent blood investigations revealed a markedly elevated serum calcium level of 3.87 mmol/L (normal range: 2.10–2.60 mmol/L) and serum parathyroid hormone level of 141.9 pmol/L (normal range: 1.30–

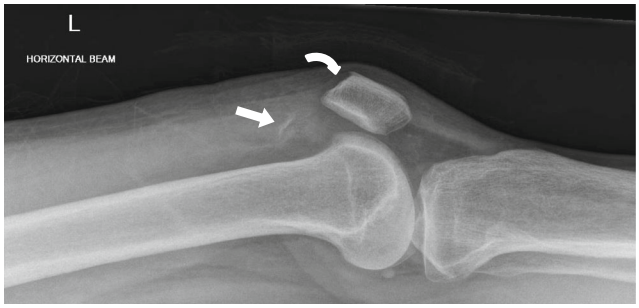

Fig. 1 Plain radiograph of the left knee in the emergency department, showing mild abnormal anterior angulation of the left patella. On retrospective review of this radiograph in conjunction with the subsequently performed left knee MRI, it was shown that the suprapatellar amorphous radio-opaque densities (*straight arrow*) seen here represent the avulsed enthesis of the quadriceps tendon, with a corresponding defect at the donor site in the patellar superior pole (*curved arrow*)

7.60 pmol/L). Fluid aspiration of the right knee yielded no abnormal microorganism growth.

Magnetic resonance imaging scans of both knees showed bilateral acute extensor mechanism disruptions. In the right knee, there was a complete acute tear at the patellar attachment of the proximal infrapatellar tendon and a partial width tear involving the medial portion of the distal infrapatellar tendon tibial attachment (Fig. 4). In the left knee, there was complete acute rupture of the distal quadriceps tendon at its patellar attachment (Fig. 5). The avulsed tendons were attached to small fragments of the entheses, corresponding to the amorphous radio-opaque fragments on the radiographs. Both knees also showed areas of bony resorption at the entheseal attachments of the avulsed tendons.

The diagnosis of acute spontaneous tendon ruptures secondary to hyperparathyroidism was made based on the biochemical and imaging results. Bilateral elbow MRI scans and bilateral ankle radiographs were subsequently obtained to evaluate for other at-risk tendons. A subacute near complete

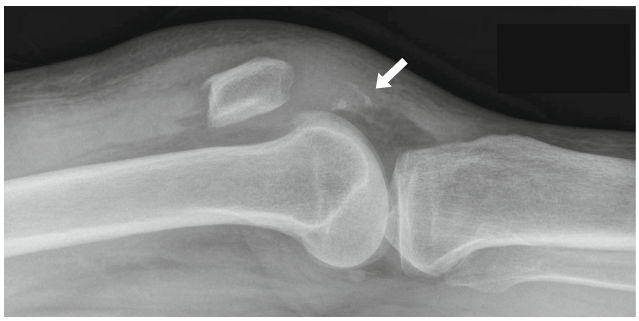

Fig. 2 An accompanying plain radiograph of the contralateral right knee demonstrating superior subluxation of the right patella, with prominent soft-tissue swelling along the anterior aspect of the right knee. Amorphous radio-opaque densities (*arrow*) projected anterior to the femoral condyles were shown on subsequent MRI to represent avulsion of the proximal infrapatellar tendon enthesis from the patellar lower pole. The donor site is not readily apparent in this projection

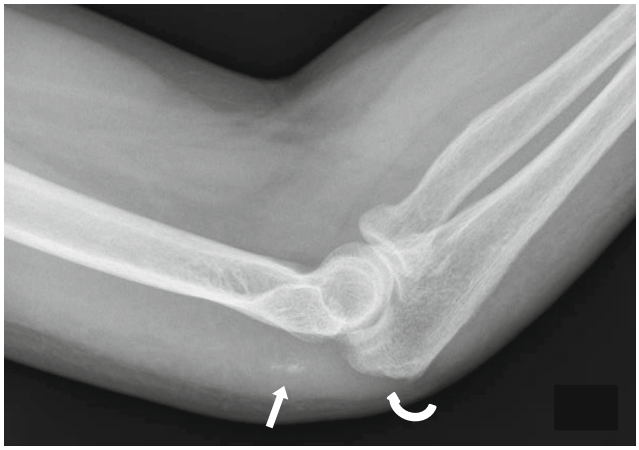

Fig. 3 Plain radiograph of the left elbow showing soft-tissue swelling at its posterior aspect. Amorphous radio-opaque densities (*straight arrow*) posterior to the distal humerus were shown on subsequent MRI to represent the avulsed enthesis of the triceps brachii tendon, with the donor site appreciated of the olecranon (*curved arrow*)

avulsion of the left triceps brachii tendon from the olecranon process attached to small fragments of its enthesis was demonstrated (Fig. 6), corresponding to the amorphous radio-opaque fragments on the radiographs. The right elbow MRI showed hyperintense signal, suspected bony resorption at the olecranon enthesis attachment of the distal triceps brachii tendon, likely secondary to hyperparathyroidism and deemed to be at risk of avulsion (Fig. 7). The bilateral ankle radiographs (not shown) revealed no acute disruption of the Achilles tendons.

A CT scan of the thorax, abdomen and pelvis was performed. This revealed a well-defined, solid, enhancing soft-tissue nodule abutting the posterior aspect of the right thyroid gland, a suspected parathyroid adenoma. Other features of hyperparathyroidism, including medullary nephrocalcinosis, urinary tract calculi and bony resorption at the acromioclavicular joints and fingers, were also evident (Fig. 8).

A neck ultrasound was performed to further assess the nodule adjacent to the right thyroid lobe. This showed a well-defined solid nodule adjacent to the posterior margin of the lower pole of the right thyroid gland, homogeneously hypoechoic relative to thyroid parenchyma. A characteristic feeding vessel was also demonstrated on Doppler interrogation (Fig. 9). A sestamibi nuclear medicine study confirmed the presence of a hyperfunctioning right inferior parathyroid nodule, a suspected adenoma, resulting in primary hyperparathyroidism (Fig. 10).

Following pre-surgical optimisation including normalisation of serum calcium with subcutaneous calcitonin and intravenous pamidronate, the patient underwent surgical repair of the right patellar, left quadriceps and left triceps brachii tendons. On the third post-operative day, the patient underwent excision of the right inferior parathyroid adenoma. Intra-

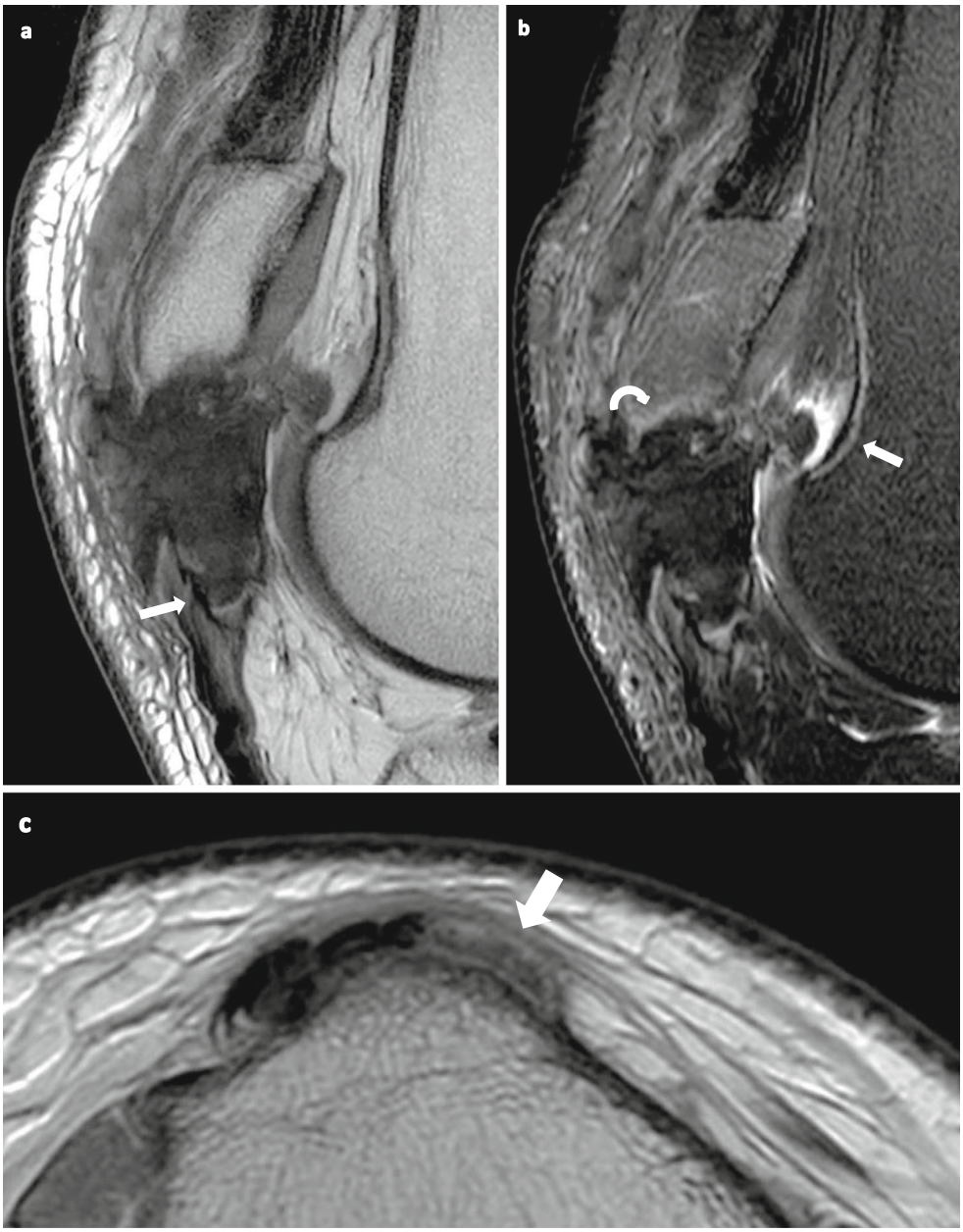

Fig. 4 **a** Right knee MRI: sagittal proton density (PD)-weighted image demonstrating a complete tear of the proximal infrapatellar tendon, being avulsed from the patellar lower pole at its enthesis, associated with an intervening hematoma. The avulsed enthesis, seen on a previous plain radiograph (Fig. 2), is demonstrated as a curvilinear structure of low signal intensity at the margin of the torn tendon (*arrow*). **b** Right knee MRI: sagittal inversion recovery (IR) image best demonstrates the subperiosteal (*straight arrow*) and subtendinous (*curved arrow*) bony resorption secondary to hyperparathyroidism. **c** Right knee MRI: axial PD image shows a partial width tear of the medial portion of the distal infrapatellar tendon at its tibial enthesis attachment (*arrow*)

operative serum parathyroid hormone dropped from 180.9 pmol/L to 11.26 pmol/L, and frozen section confirmed the presence of parathyroid tissue. The parathyroid nodule on gross examination measured 2.0 × 1.2 × 1.0 cm and weighed 4.4 g. Post-operatively, hungry bone syndrome was anticipated and managed with appropriate replacement and regular monitoring by the endocrinology team. The final histological diagnosis was of a cellular parathyroid lesion consistent with a parathyroid adenoma without evidence of malignancy (Fig. 11).

The patient's post-operative recovery was uneventful and he was discharged to a community hospital for further rehabilitative management and therapy.

Discussion

Spontaneous tendon rupture is a rare presentation of primary hyperparathyroidism [1]. The predominant histopathological mechanism of tendon failure in hyperparathyroidism has been proposed to be chronic osteitis fibrosa leading to repeated micro-avulsion fractures of the bone at the tendon insertion site and eventually to total enthesis avulsion [2].

Our case of three separate major tendons in the same patient simultaneously failing in the same manner (enthesis avulsion) reflects the systemic sequelae of hyperparathyroidism as detailed above and presents an opportunity to describe the

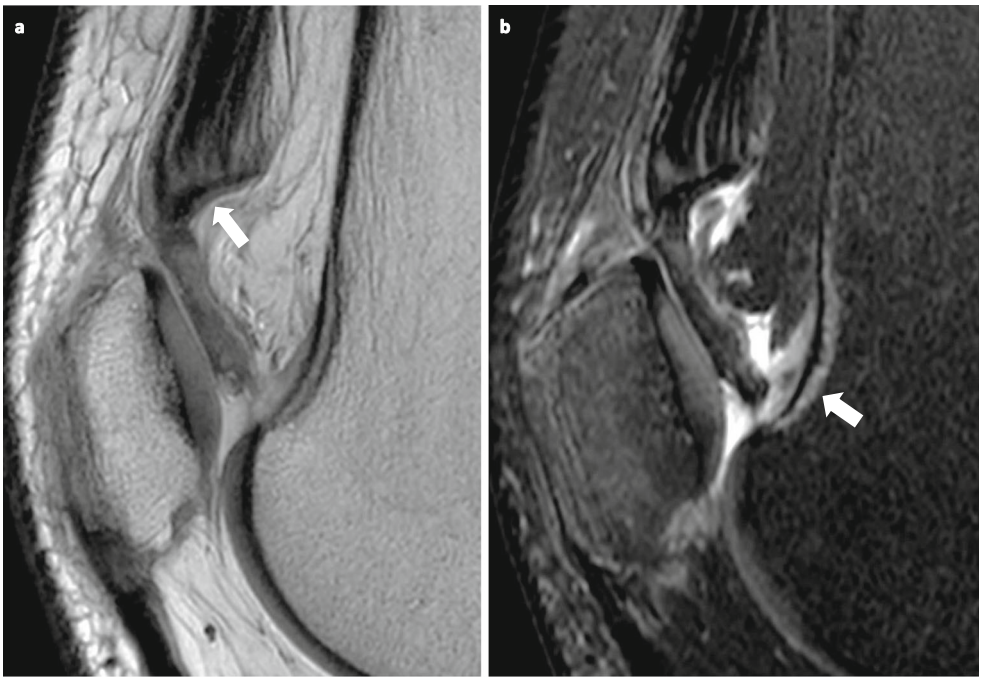

Fig. 5 **a** Left knee MRI: sagittal PD image demonstrating a complete tear of the distal quadriceps tendon at its patellar attachment with an avulsed curvilinear enthesis fragment (*arrow*), corresponding to the findings on plain radiograph (Fig. 1). **b** Left knee MRI: sagittal IR image shows incidental areas of subperiosteal bony resorption in the distal femur (*arrow*)

imaging correlations of this condition. The flaky curvilinear high radiodensity fragments seen on plain radiographs at all three sites of tendon failure were shown on MRI to represent the avulsed entheses, rather than dystrophic calcifications, as previously alluded to in earlier reports [3, 4]. Irregular linear or serpiginous areas of subperiosteal/subentheseal bone

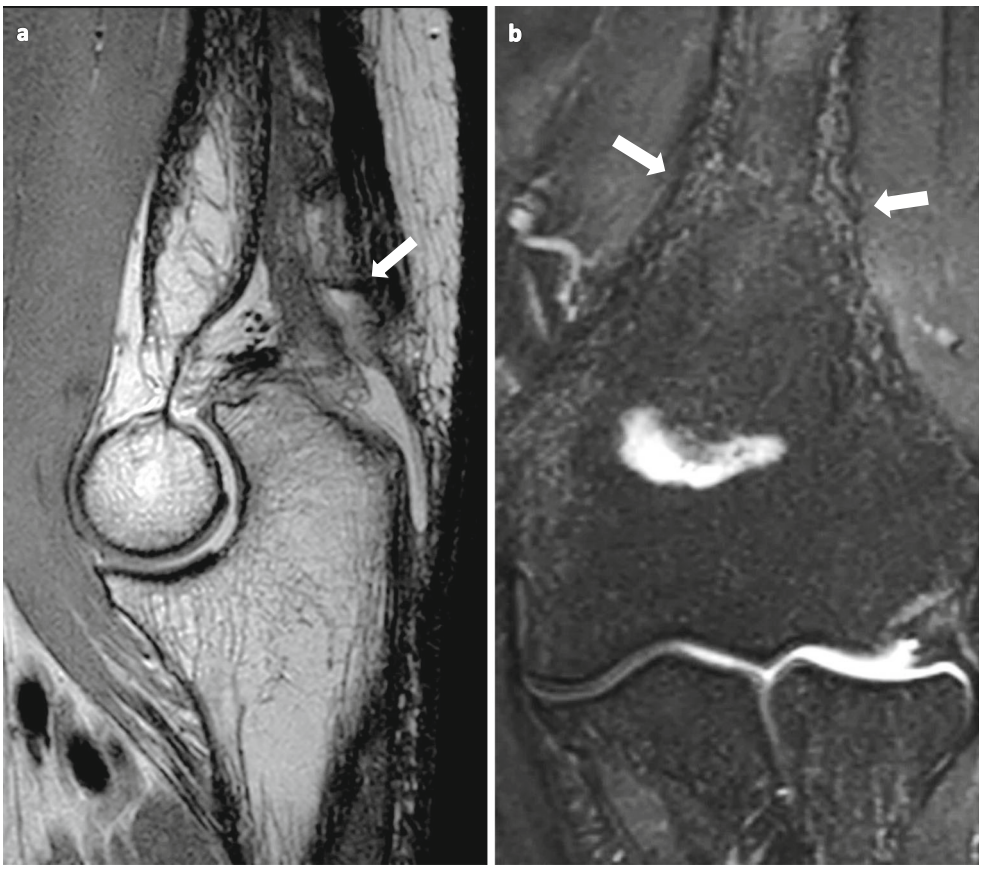

Fig. 6 **a** Left elbow MRI: sagittal PD image demonstrating near complete avulsion of the left triceps brachii tendon and enthesis (*arrow*) from the left olecranon process, as seen on a plain radiograph (Fig. 3). **b** Left elbow MRI: coronal IR image shows areas of subperiosteal bone resorption in the distal humerus (*arrows*)

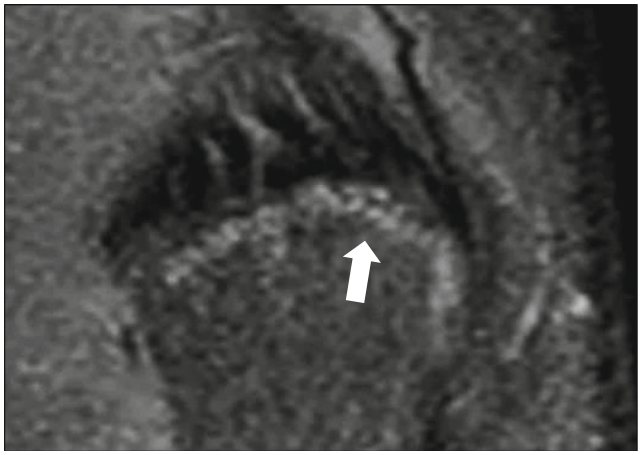

Fig. 7 Right elbow MRI: coronal IR image shows subtendinous bone resorption in the olecranon (*arrow*), indicating an at-risk right triceps enthesis

marrow oedema on the MRI studies of all three ruptured tendons likely represent areas of bone resorption in the context of generalised osteitis fibrosa and hyperparathyroidism.

Of note in our case is that aside from the sites of avulsion, the substance of the rest of the torn tendons otherwise displays relatively preserved signal intensity and morphology on MRI, suggesting the absence of radiologically appreciable interstitial structural abnormality within the tendon substance on conventional imaging, thus supporting the notion that the pathological mechanism in hyperparathyroidism might primarily be that of resorption and failure at the enthesis rather than failure due to internal derangement of the tendon substance per se.

A previously reported case of major tendon rupture in primary hyperparathyroidism unconfounded by other chronic co-morbidities displays similar imaging features to our case. Review of the images in the case reported by Gao et al. [4] showed enthesis avulsion (rather than tendon calcification as mentioned in the report) and distal femur subperiosteal bone resorption, with preserved tendon substance signal. However, the report did not appreciate the significance of the enthesis avulsion and relative preservation of the quadriceps tendon. Another report by Uzer et al. [5] of hyperparathyroidism secondary to vitamin D deficiency presenting as spontaneous

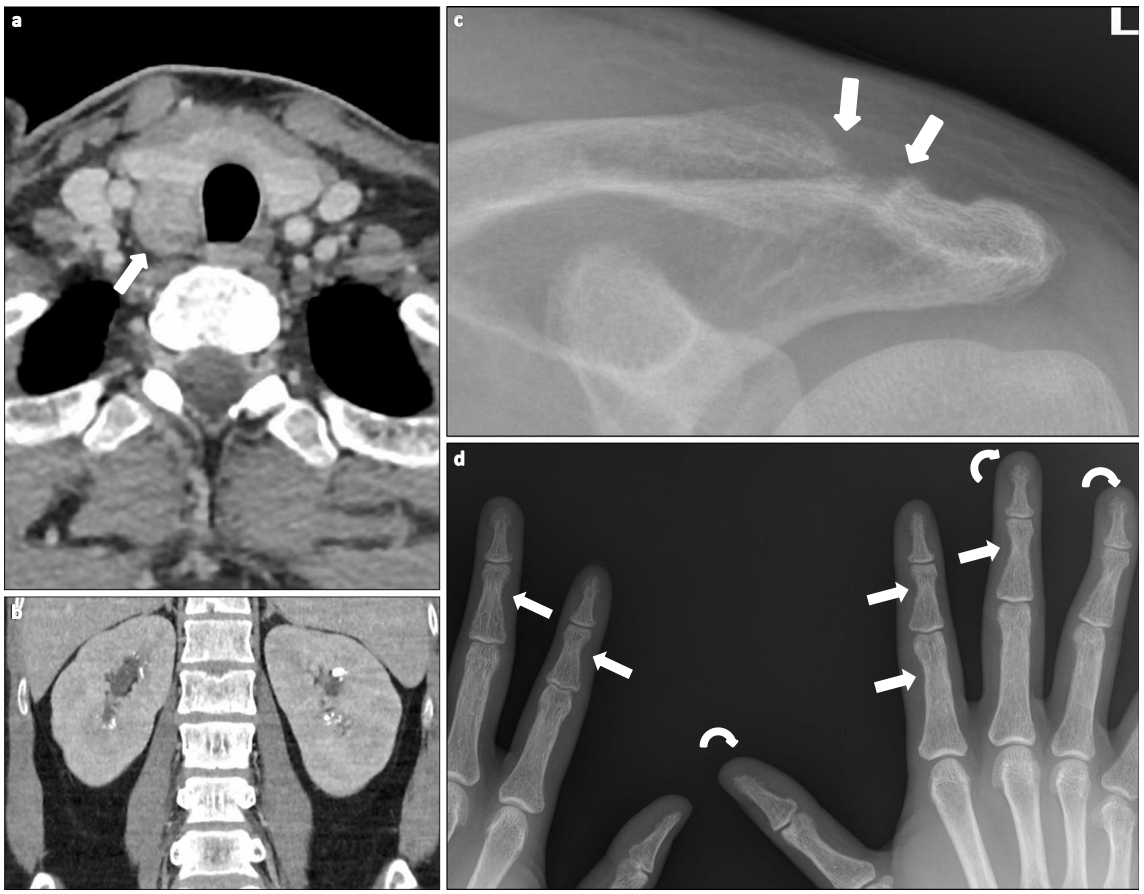

Fig. 8 **a** Axial CT image showing a well-defined solid nodule abutting the posterior aspect of the right thyroid mid-lower pole (*arrow*), a suspected parathyroid adenoma. **b** Coronal CT image of renal medullary nephrocalcinosis and caliceal calculi. **c** Plain radiograph showing subchondral bone resorption at the left acromioclavicular joint, involving both the clavicle and the acromion (*arrows*). **d** Plain radiograph of both hands showing subperiosteal bone resorption affecting the radial aspects of the proximal and middle phalanges (*straight arrows*) and acro-osteolysis (*curved arrows*)

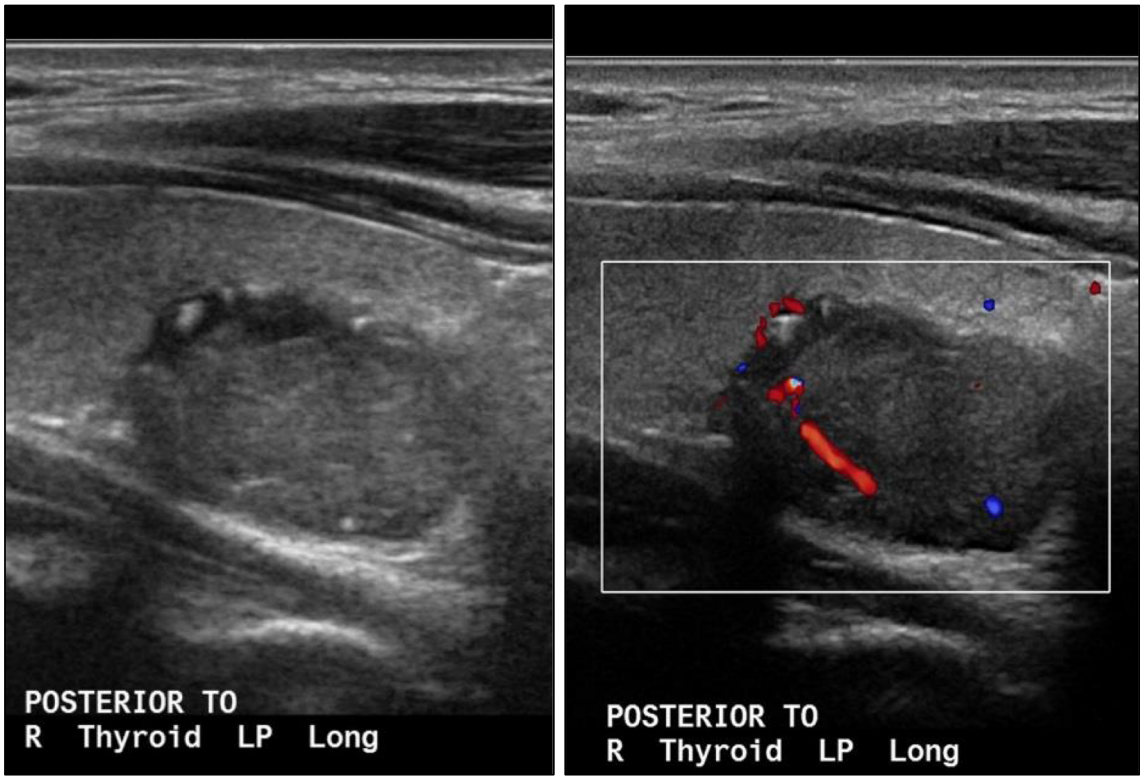

Fig. 9 Ultrasound of the neck demonstrating a well-defined solid nodule abutting the posterior margin of the lower pole of the right thyroid gland, homogeneously hypoechoic relative to thyroid parenchyma. A characteristic feeding vessel is seen on Doppler interrogation

bilateral quadriceps tendon ruptures in a young patient also showed enthesis avulsions.

Another commonly reported cause of spontaneous tendon rupture is chronic renal failure. Bilateral knee extensor mechanism ruptures generally reflect underlying systemic disease, of which chronic renal failure has been the most frequently described and commonly reported associated condition [6]. In chronic renal failure, multiple risk factors for tendon rupture have been proposed, including duration of haemodialysis [7], β_2-amyloidosis [8], vitamin D deficiency [9] and uremic toxins [10]. Recent literature, however, has emphasised secondary hyperparathyroidism as the principal pathophysiological factor for tendon ruptures in chronic renal failure [3, 11–13].

A past companion case from our hospital archives is also hereby presented of a 26-year-old man with end-stage renal disease on long-term haemodialysis presenting with spontaneous bilateral knee extensor mechanism failures. The plain radiograph and MRI studies (Figs. 12 and 13) reveal striking similarities to those of our primary patient, bearing the hallmarks of hyperparathyroidism-induced tendon ruptures at the entheses. This companion case of tendon rupture in the setting of chronic renal failure demonstrates two separate major tendons failing in exactly the same pattern and location as the currently presented case of primary hyperparathyroidism. This supports the hypothesis that secondary hyperparathyroidism, rather than the other factors mentioned above, underpins the mechanism of spontaneous tendon failure in chronic renal failure.

Review of past reports also yielded concordance with our premise. For example, the reports by Wani et al. [14], Chen et al. [15] and Gao et al. [16] all contain images demonstrating tendon avulsions at the entheses, rather than tendon substance ruptures, of bilateral knee extensor mechanisms in patients with chronic renal failure.

In contrast, spontaneous tendon failure from other aetiologies in adults, such as those reported in the context of connective tissue disease [17], fluoroquinolone use [18], steroid therapy [19] and obesity [20], display disruptions of the tendon substance with macroscopically intact entheses on imaging, thus alluding to structural weakening and failure within the tendon as opposed to the osteo-tendinous junction (enthesis).

The imaging findings in this case and review of available literature support the hypothesis that spontaneous tendon ruptures in primary hyperparathyroidism result primarily from bony resorption of the enthesis, leading to failure and avulsion of an otherwise macroscopically intact tendon. Likewise, we also hypothesise that entheseal resorption and its failure due to secondary hyperparathyroidism is the main pathological mechanism leading to spontaneous tendon ruptures in patients with chronic renal failure.

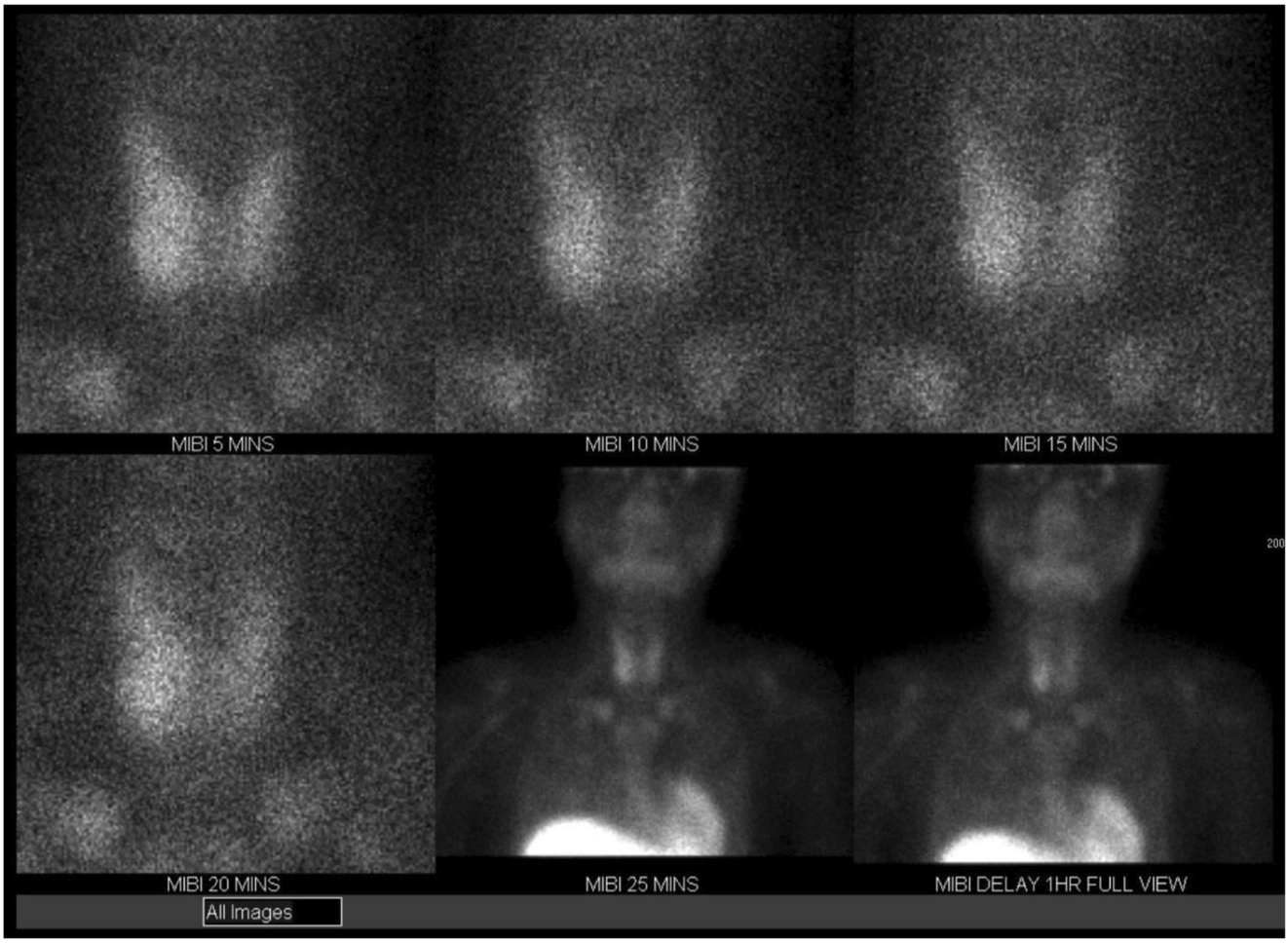

Fig. 10 A sestamibi nuclear medicine study confirmed the presence of a hyperfunctioning right inferior parathyroid nodule, and the absence of hyperfunctioning parathyroid tissue at the expected locations of the other parathyroid glands. Findings are indicative of an adenoma, consistent with the clinical diagnosis of primary hyperparathyroidism

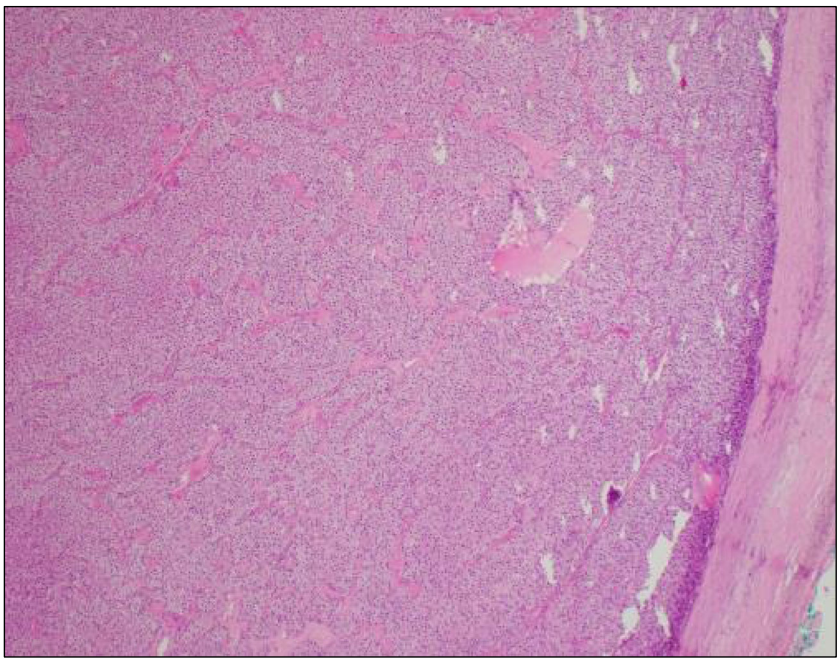

Fig. 11 Parathyroid adenoma (H&E). Histologically, the lesion was encapsulated and composed exclusively of chief cells set within a fine vascular background devoid of stromal fat. The distinction between parathyroid gland adenoma and hyperplasia is based on the well-defined encapsulation and the lack of stromal fat in an adenoma. This was substantiated by the presence of three normal remaining parathyroid glands on sestamibi nuclear medicine study; however, the best corroborative evidence was the immediate marked drop in parathyroid hormone levels upon surgical removal of the nodule from 180.9 pmol/L to 11.26 pmol/L.

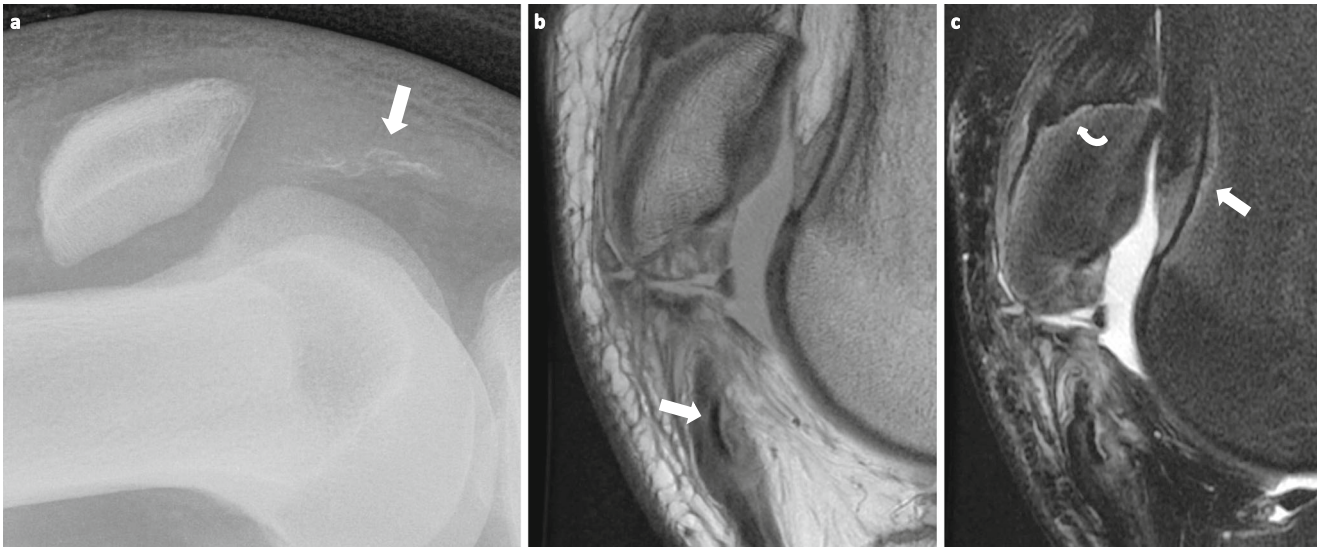

Fig. 12 **a** Companion case: right knee plain radiograph from this companion case demonstrating flaky amorphous density (*arrow*) projected anterior to the femoral condyles, representing an avulsed infrapatellar tendon enthesis. **b** Companion case: right knee sagittal PD MRI again demonstrating the avulsed enthesis (*arrow*). **c** Companion case: right knee sagittal IR MRI reveals subperiosteal (*straight arrow*) and subtendinous (*curved arrow*) bone resorption, rendering the right quadriceps tendon at risk of enthesis avulsion

We thus posit that various medical conditions and diseases result in hyperparathyroidism as the common pathophysiological pathway underpinning this distinctive pattern of tendon failure at its enthesis, whether from primary, secondary or tertiary causes. Although chronic renal failure is by far the most common cause of hyperparathyroidism, other less common and rare causes, such as parathyroid hyperplasia, parathyroid malignancy, multiple endocrine neoplasia syndrome, vitamin D deficiency, severe calcium deficiency, previous neck irradiation [21] and chronic lithium therapy [22] are on the list of differential diagnoses.

We also note that MRI was able to demonstrate ongoing subtendinous bony resorption in otherwise clinically intact tendons, i.e. identifying tendons at risk of future failure. These insights may potentially augment the clinical management of chronic renal failure with respect to focusing therapy on the correction of hyperparathyroidism, in addition to performing targeted MRI screening of major tendons to identify those at risk and to prevent their potential failure. Furthermore, recognising on plain radiographs the flaky radio-opaque densities (representing the avulsed entheses) adjacent to the bony attachments of spontaneously ruptured

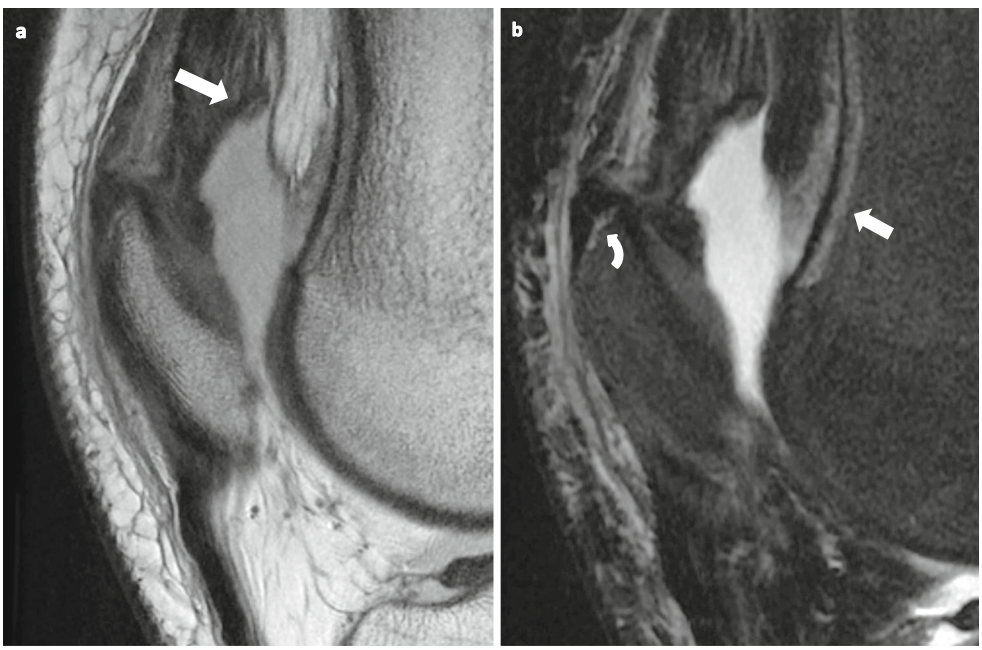

Fig. 13 **a** Companion case: left knee sagittal PD MRI demonstrating the avulsed enthesis, represented by curvilinear hypointensity at the free margin of the quadriceps tendon (*arrow*). **b** Companion case: left knee sagittal IR MRI reveals the subperiosteal (*straight arrow*) and subtendinous (*curved arrow*) bone oedema representing bony resorption due to secondary hyperparathyroidism

tendons may aid in making a specific diagnosis of hyperparathyroidism at the initial clinical presentation.

In conclusion, our reported case and review of previous literature provide an imaging basis for supporting the hypotheses that spontaneous tendon failure in the clinical setting of primary hyperparathyroidism might be due to tendon avulsion secondary to bony resorption of the enthesis rather than interstitial failure of the tendon substance per se, and that spontaneous tendon failure in the setting of chronic renal failure occurs through similar pathophysiological mechanisms due to secondary hyperparathyroidism. Less common and rare causes of hyperparathyroidism associated with spontaneous tendon ruptures might likewise be postulated to result from enthesis failure with similar distinctive imaging features.

Compliance with ethical standards

Conflicts of interest The authors declare that they have no conflicts of interest.

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