

# Sesamoid ossicles within the nuchal ligament: a report of two cases and review of the literature

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Radiology Case. 2011 Aug; 5(8):22-29 :: DOI: 10.3941/jrcr.v5i8.708

## ABSTRACT

Sesamoid ossicles are ovoid or round nodular small bones found usually within a joint or tendon of the upper and lower limb and rarely at other regions. They are usually asymptomatic, resulting in an increased propensity to injuries. We report on two cases where sesamoid ossicles were detected within the nuchal ligament. The first one was a 55-years-old Caucasian male office employee, complaining of chronic pain in the cervical spine region accompanied by numbness of the left upper limb. During the routine lateral X-ray imaging two sesamoid ossicles were found within the nuchal ligament at the C4 and C5 spinous processes level. The second case was a 64-years-old Caucasian female store employee, complaining of chronic cervical spine pain. The routine lateral X-ray exhibited two sesamoid ossicles within the nuchal ligament at the C5 and C6 spinous processes level. Sesamoid ossicles found in the nuchal ligament in rare cases may become symptomatic and the pathogenesis mechanism should be differentially diagnosed by the physician. We discuss the precise morphology of the trait, the possible etiological mechanisms for their appearance, the histological features, as well as differential diagnosis.

## CASE REPORT

### CASE REPORT

#### CASE 1

A 55-years-old Caucasian male visited a private orthopaedic centre complaining of chronic pain in cervical spine region accompanied by mild non-localized numbness of the left upper limb. No trauma or other pathological condition has been reported in his medical history. The patient was occupied as an office employee. During physical examination we found a decrease in range of movement of the cervical spine accompanied by local tenderness in the middle and lower third of the cervical spine with normal reflexes and muscle strength, and numbness of the left upper limb. Lateral view X-ray imaging of the cervical spine was performed and the existence of two supernumerary ossicles within the nuchal ligament was noticed at the level of C4 and C5 vertebral spinous processes, respectively (Figure 1a). Both ossicles had smooth borders, whereas the distal one had nearly ovoid and

the proximal nearly round shape (Figure 1b). Avulsion fractures of the spinal processes were excluded from differential diagnosis due to absence of tenderness at the C4 and C5 cervical spine processes and lack of irregular circumference of the lesions, while the tips of the corresponding spinous processes show no defects. The chronic neck pain, as well as, the numbness of the left upper limb, was attributed to the co-existence of cervical spondylosis at the C4 and C5 level.

#### CASE 2

A 64-years-old Caucasian female, occupied as a store employee, visited her physician complaining of chronic cervical spine pain over the last 14 months. No traumatic injury or other pathological condition has been recorded in her medical history. The physical examination revealed a decrease in range of movement of the cervical spine accompanied by local tenderness in the middle and lower third of the cervical

spine with normal reflexes and muscle strength. The routine lateral X-ray exhibited two sesamoid ossicles within the nuchal ligament at the C5 and C6 spinous processes level, respectively (Figure 2a). Both ossicles were found in close relation to the corresponding cervical spinous processes. The distal sesamoid ossicle was round in shape with regular borders, while the proximal one was larger and ovoid shaped (Figure 2b). The possibility of avulsion fractures of the spinal processes were excluded due to absence of any tenderness at the C5 and C6 cervical spine processes and lack of irregular circumference of the lesions, while the tips of the corresponding spinous processes had no defects. Due to the nature and location of the diffuse pain, the symptoms of chronic neck pain were attributed to cervical spondylosis symptoms.

## DISCUSSION

Sesamoid ossicles within the nuchal ligament can be an incidental finding during lateral radiographs or other imaging of the neck. The exact pathogenesis of such ossicles is unknown but is believed that they are classic sesamoid ossicles formed within ligamentous or tendinous structures. The trait may be misdiagnosed as myositis ossificans circumscripta, nuchal fibrocartilaginous pseudotumor, calcinosis circumscripta, Clay-shoveler's fracture, neoplasms, and diseases with concomitant calcification deposition [1-5].

The terminology of sesamoid ossicles (*ossa sesamoidea*) derives from the seeds of *sesamum indicum* plant. Their composition can be fibrous, cartilaginous, bony or mixture of the previous, whereas their broad functional commission is in: alteration of axonal pull of tendons, friction reduction, underlying cartilage protection, friction reduction due to slide between cartilages, scattering of induced force in an articulation, and contribution in aesthetics of the joints. Sesamoid ossicles are always found within a tendon unlike accessory ossicles, and they are prone to injuries [6].

The nuchal ligament portrays as supportive structure for the head weight, muscle attachment location, neck midline orientation landmark, restrains cervical spine flexion [7], maintains the lordotic alignment, stabilizes the head during rotation of the cervical spine [8], and forms a septum between the muscles of the posterior aspect of the neck. It is triangular in cross section and divides in funicular (dorsal) and lamellar (ventral) segments [9]. The superior border attaches to the median nuchal line of the skull and to the external occipital protuberance, the posterior border extends to C7 vertebral spine and attaches to the trapezoid muscle, while the anterior border attaches to the cervical spines and the intervening ligaments. Below the C7 vertebra it unifies with the supraspinatus ligament. The upper trapezoid, rhomboid minor, splenius capitis and serratus posterior superior muscles are attached to the ligament [10]. In humans, nuchal ligament is composed mostly of collagen and less by elastic fibers [9]. Mercer and Bogduk, in a cadaveric study reported that the ligament does not exhibit any notable anatomical variations among the studied material [10]. In animals, the nuchal

ligament serve to sustain the weight of the head due to tetrapod walking and contribute in proper head balance, while the erect position of humans led to great decrease in its size [9].

The true sesamoid ossicles within the nuchal ligament are usually painless, single, and the majority of them ovoid or round. The reported incidence by Scapinelli is 11.3% in males, and 3.5% in females. Usually, these ossicles are fully developed after the third decade of life, and in 80% of cases are found at the C5/C6 or C6/C7 level. Histologically, Scapinelli found that these ossicles are produced after gradual osseous metaplasia of calcified fibrocartilaginous nodules. Mechanical factors are responsible for the ossicles' pathogenesis. Nuchal ligament is subjected to pressure against the apex of the spinous processes during forward flexion of the neck leading ultimately to ossicles formation at the higher mobility region thus C5-C6 level (Table 2) [11].

Acute, repeated injury or irritation over long periods can induce metaplasia of connective and muscular tissue into bone, a condition widely known as myositis ossificans [12]. Noble, in 1924 classified myositis ossificans into three types, the progressive, the circumscripta and the localized [13]. Myositis ossificans circumscripta is induced by repeated injury and irritation over a long period of time onto a limited area, and can be found within nuchal ligament as painful osseous nodules hardly differentiated by true sesamoid ossicles [14]. The very rare case of heterotopic appearance of a secondary nucleus of the C6 spinous process could lead to misdiagnosis of a sesamoid of the nuchal ligament [15]. Calcinosis circumscripta of the nuchal ligament could confuse the radiologist and the clinician, leading to establishment of sesamoid ossicle diagnosis. Nevertheless, in cases of calcinosis circumscripta there are multiple focal calcific nodules with presumably a history of acute injury and neck pain [16]. Other pathological conditions associated with calcification should be differentiated such as calcium hydroxyapatite deposition disease [4] and calcareus bursitis [15]. Nuchal fibrocartilaginous pseudotumors could be misdiagnosed as true sesamoid ossicles since they are developed in the nuchal ligament as hard or soft nodular masses accompanied by stiffness and vague neck pain [17]. These recently reported in the literature lesions are non-neoplastic masses developed mostly as a reaction to soft-tissue injury and they are generated after fibrocartilaginous metaplasia of the inferior most portion of the nuchal ligament [3,18]. Moreover, nevoid basal cell carcinoma syndrome, an autosomal-dominant disorder, is characterized by multiple basal cell carcinomas, jaw cysts, spine and rib anomalies, palmar or plantar pits, and in 18% calcification of the nuchal ligament [19].

Calcifications may appear in various conditions such as burns, fluoride poisoning, surgical trauma, sarcoidosis, amyloid deposition, systemic lupus erythematosus or hyperparathyroidism should also be taken into account during differential diagnosis of the nuchal ligament sesamoid ossicles [5,20]. Other conditions in which "suspicious lesions" mimicking sesamoid ossicles are ossification of ligamentum flavum and diffuse idiopathic skeletal hyperostosis in which the presented masses are located at the ligamentum flavum and retrospinal muscles, respectively [4]. Benign tumors such as

osteoma and neurofibroma, and malignant neoplasms such as synovial sarcoma could be misdiagnosed as sesamoid ossicles. These lesions are occasionally calcified and situated usually adjacent to the nuchal ligament [4,21]. Clay-shoveler fracture is the fracture of C7 spinous process, which resembles to true sesamoid ossicle within the nuchal ligament, whilst laminar fracture of the lower cervical vertebrae could, under circumstances, induce a diagnostic dilemma. At last, expansion of the septic facet joint arthritis to the posterior paraspinal muscles and abscess developing within these muscles could confuse the physician and the radiologist in case of calcareous deposition [4] (Table 1).

#### TEACHING POINT

The existence of sesamoid ossicles within the nuchal ligament can be an incidental radiological finding in adults with chronic neck pain or injury of the cervical spine. Usually, they do not cause any symptoms and they are painless. They are presumably formed due to repeated injury and metaplastic localized modification over a long period of time mostly at the C4/C5, C5/C6, and C6/C7 level.

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FIGURES



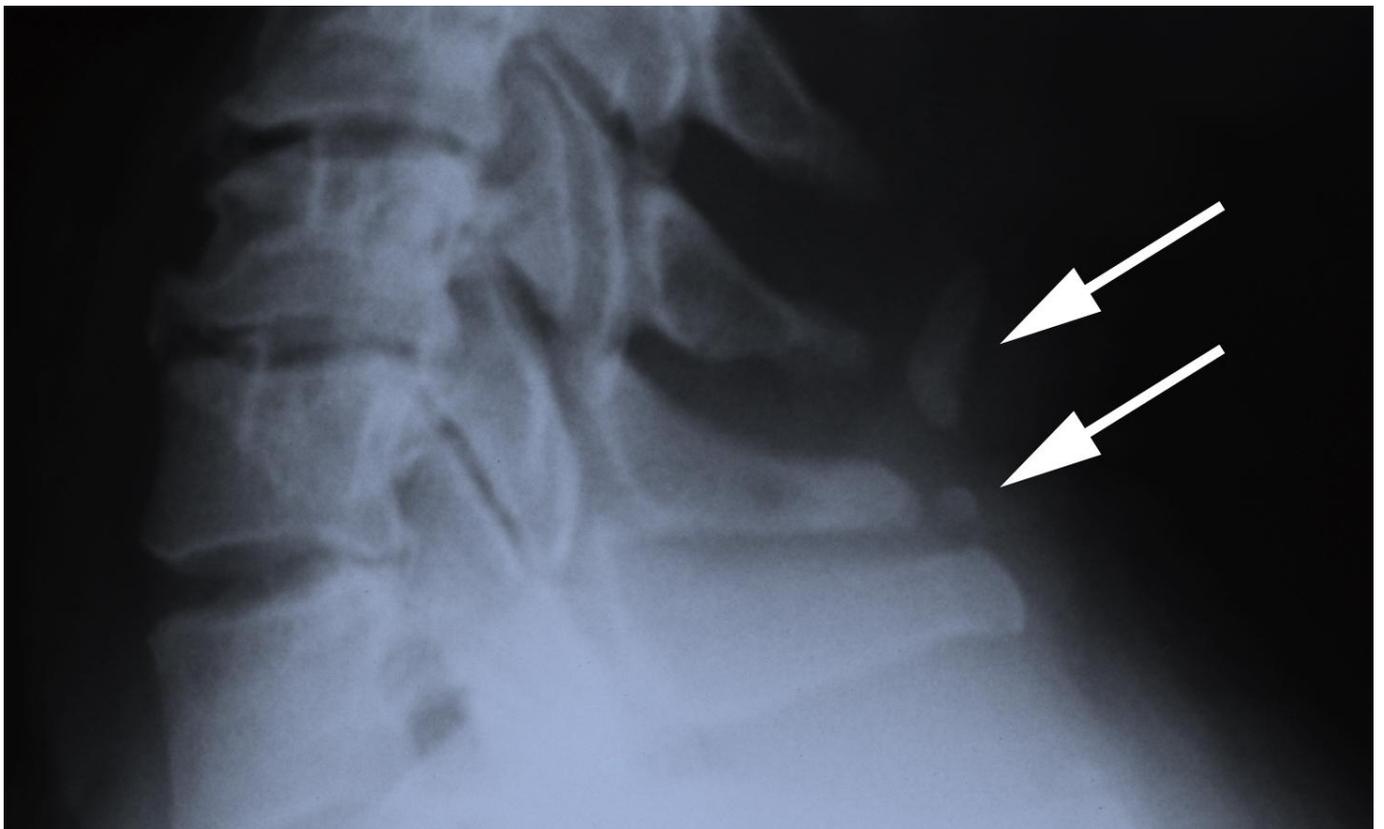
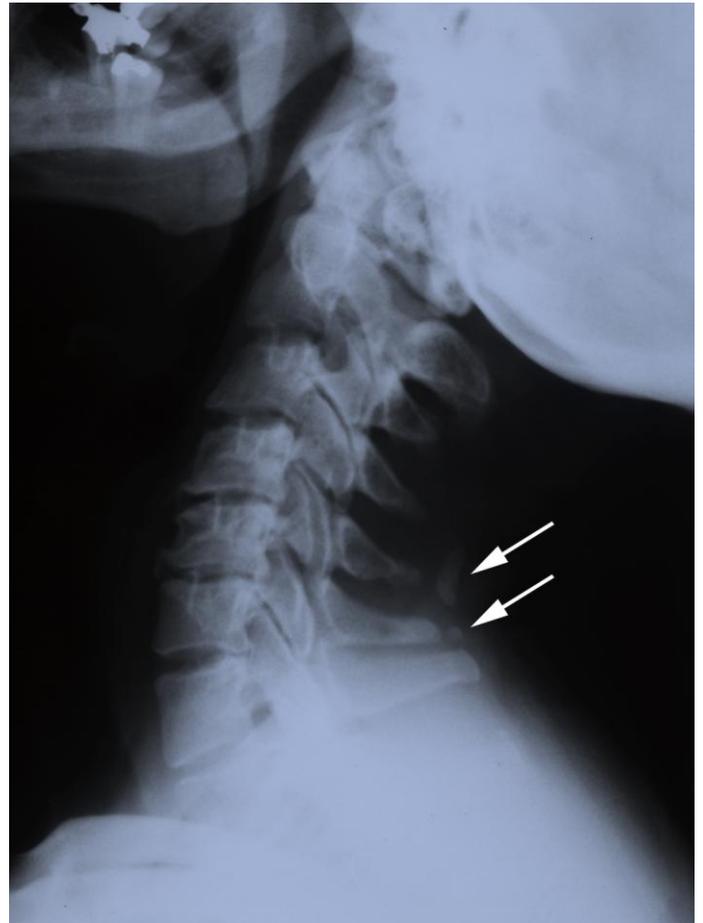
**Figure 1a (left):** Lateral plain radiograph of a 55-years-old male patient with sesamoid ossicles within the nuchal ligament. Two sesamoid ossicles (arrows) within the dorsal (funicular) portion of the nuchal ligament at the C4 and C5 spinous process level are noticed. The distal one is nearly ovoid whereas the proximal is nearly round in shape. Both ossicles have regular borders.

**Figure 1b (bottom):** Magnified view of Figure 1a.



**Figure 2a (right):** Lateral plain radiograph of a 64-years-old female patient with sesamoid ossicles within the nuchal ligament. Two sesamoid ossicles (arrows) are seen within the dorsal (funicular) portion of the nuchal ligament at the C5 and C6 spinous process level. The distal sesamoid ossicles is round in shape with regular borders, while the proximal one is larger and ovoid shaped.

**Figure 2b (bottom):** Magnified view of Figure 2a.



Pathology/Traumatology	Plain X-ray	CT	MRI
Sesamoid ossicles	Focal well-defined osseous masses, well-circumscribed with regular shape	Cortical bone surrounding central marrow space	T1W1: Usually hypointensity. Occasionally, high signal intensity in cases of large amount of marrow fat. T2W1: Similar findings to T1W1
Myositis ossificans circumscripta	Solitary or multiple irregular or linear shaped dense and sharply defined osseous formations	Well-circumscribed dense bony formations	T1W1: Low signal intensity. T2W1: Low signal intensity
Heterotopic development of the secondary nucleus of the spinous process of C6 vertebra	Solitary dense regular bony formation	Solitary well-defined bony mass	T1W1: Low signal intensity, occasionally high signal in instances with considerable marrow fat. T2W1: Similar findings to T1W1
Calcinosis circumscripta	Multifocal well-circumscribed calcified masses	Multiple irregular calcifications	The calcifications are usually devoid of signal in MRI
Calcific tendinitis or calcium hydroxyapatite disease of posterior paraspinal muscles (very rare; usually located to the retropharyngeal space)	Retrospinal amorphous focal calcifications with soft tissue swelling	Amorphous focal calcifications within the nuchal muscles, with or without fluid collection along fascial planes	T1W1: Calcific deposits of low signal intensity with soft tissue swelling. T2W1: Calcific deposits of low signal intensity with occasional fluid collection (high signal)
Calcareous bursitis	Calcified nodule	Usually single calcified nodule	T1W1: Low signal intensity. T2W1: High signal intensity
Nuchal fibrocartilaginous pseudotumor	Mass of low intensity	Nodular thickening of the ligament without calcifications	Focal thickening of the nuchal ligament
Nevoid basal cell carcinoma syndrome	Multifocal calcification nodules	Multiple irregular calcified masses	Calcifications are devoid of signal on MRI
Calcifications presented in various conditions (burns, surgical trauma, fluoride poisoning, neoplasms, sarcoidosis, systemic lupus erythymatosus, hyperparathyroidism, amyloid deposition, etc.)	Diffuse irregular nodular calcifications	Diffuse calcifications	Calcifications are devoid of signal on MRI
Ossification of ligamentum flavum (selective location in that ligament and not in nuchal ligament)	Curvilinear calcification ventral and not dorsal to lamina	Hyperdense linear or nodular ossification ventral to lamina	T1W1: Hypo- to hyperintense masses ventral to lamina. T2W1: Hypointense masses ventral to lamina
Diffuse idiopathic skeletal hyperostosis (occasionally paraspinal ossification)	Multiple well-defined ossifications	Thick ossified masses	T1W1: Hypointense if calcified and iso- to hyperintense if marrow fat exists. T2W1: Similar findings to T1W1

**Table 1:** Differential diagnosis of masses potentially developed within or adjacent to the nuchal ligament. (continued on next page)

Pathology/Traumatology	Plain X-ray	CT	MRI
Osteoma	Extremely dense well-circumscribed round lesions	Well-defined bony lesions	T1W1: Low signal intensity. T2W1: Low signal intensity
Synovial sarcoma (occasionally calcified)	Amorphous diffuse calcifications	Multifocal irregular calcifications	T1W1: Low signal intensity. T2W1: High signal intensity
Neurofibroma (occasionally retrospinal with rare calcifications)	Diffuse or localized calcifications	Calcifications	T1W1: Low signal intensity. T2W1: Similar findings to T1W1
Clay-shoveler fracture (fracture of C7 spinous process)	Solitary (usually) triangular or quadrangular dense bony fragment with bony defect of the adjacent spinous process	Solitary bony fragment	T1W1: Low signal intensity. Higher intensity is found when considerable marrow fat exists. T2W1: Similar findings to T1W1
Laminar fracture of lower cervical vertebrae	Solitary (usually) irregular bony fragment with concomitant disturbance of the adjacent laminar morphology	Solitary bony fragment	T1W1: Low signal intensity. T2W1: Low signal intensity
Abscess of posterior paraspinal muscles	Density of posterior paraspinal muscles	Amorphous soft tissue density, occasionally calcified	T1W1: Iso- to hypointense. T2W1 with fat suppression detects early retrospinal inflammation with or without diffuse calcifications
Expansion of septic facet joint arthritis to the posterior paraspinal muscles	Posterior paraspinal soft tissue density with rare diffuse multifocal calcifications. Concomitant osteolytic or sclerotic facet joint	Amorphous soft tissue density with rare calcifications. Mixed osteolytic and sclerotic facet changes	T1W1: Hypointensity. T2W1: Hyperintensity with retrospinal soft tissue edema with rare calcifications

**Table 1:** Differential diagnosis of masses potentially developed within or adjacent to the nuchal ligament. (continued)

<b>Etiology*</b>	Mechanical pressure of the nuchal ligament against the apex of the spinous processes during forward flexion of the neck
<b>Incidence*</b>	11.3% in males, 3.5% in females
<b>Gender ratio*</b>	Males appear to be more commonly affected than males. Ratio: 3.2:1
<b>Age prediction</b>	Unknown
<b>Risk factors</b>	Repeated local injury over long periods
<b>Clinical features</b>	Asymptomatic
<b>Treatment</b>	No treatment is needed
<b>Prognosis</b>	Excellent. It is a benign disorder
<b>Finding on imaging</b>	One or more ossicles with regular ovoid or circular shape and smooth surface within the nuchal ligament at the level of the lower spinous processes of the cervical vertebrae

**Table 2:** Summary table of sesamoid ossicles within the nuchal ligament.

\*Data derived from Scapinelli's (1963) work.

**ABBREVIATIONS**

C4 = 4<sup>th</sup> cervical level

**KEYWORDS**

Accessory ossicles; Nuchal ligament

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