

# Osteoma mucosalis, a new entity: an interesting case report of heterotopic ossification in the nail bed and review of the literature

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Heterotopic ossification is the formation of bone tissue at an abnormal site. The ossification of soft tissue outside the skeletal system can occur anywhere and can be found in mucosal tissues. This is the first case report of an osteoma mucosalis affecting the nail bed. We also reviewed the heterotopic ossification and calcification of cutaneous and mucosal sites in the PubMed, Scopus, and Google Scholar databases.

**Keywords:** cutaneous, heterotopic ossification, heterotopic calcification, nail bed, osteoma mucosalis

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## INTRODUCTION

Osteoma cutis is a rare condition characterized by focal ossification in the dermis or subcutaneous tissue <sup>1</sup>, which can be classified as primary or secondary <sup>2</sup>. The primary one is seen among patients with predisposing diseases such as Albright hereditary osteodystrophy and fibrodysplasia ossificans <sup>3</sup>. The prevalence of primary osteoma cutis is just 15 percent of all cases <sup>2</sup>. There is a wide spectrum of differential diagnoses according to the subungual lesions. As a nail biopsy is a difficult and annoying procedure, the diagnosis of nail unit tumors is always challenging <sup>4</sup>. As subungual tumors do not have specific clinical manifestations, the nail unit biopsy can help differentiate benign

from malignant conditions and it is safe and simple when the principles are followed. Heterotopic ossification is the formation of lamellar bone tissue at an abnormal site <sup>5</sup>. The term osteoma cutis describes ectopic bone formation in the dermis or subcutaneous tissue but ossification of soft tissue can occur anywhere outside the skeletal system. However, the etiology of secondary heterotopic ossification remains unclear. Some studies showed that traumatic or inflammatory processes can play an important role in the ectopic formation of bone tissue <sup>6</sup>.

Important diseases in the differential diagnosis of tender subungual lesions include neuroma and glomus tumor, although many subungual lesions like exostosis can cause pain <sup>5</sup>. We decided to

report a case of bone formation at the nail bed as a new entity called 'osteoma mucosalis'. This represents the first case of primary ossification of the nail bed as a mucosa-like tissue.

## CASE PRESENTATION

A 30-year-old female patient who presented with a tender lesion under the great toenail was referred to the dermatology clinic of a hospital in August 2015. The patient gave informed consent for participation in the study and publication of her photos.

On physical examination, the color and the texture of her nail plate were normal, whereas, a slight pain caused by trauma without radiation to the shoulder was provoked. This pain did not have any relation to the alteration of temperature.

In the subungual area, there was a fusiform-like tumor emerging in the tiptoe (Figure 1). On the X-ray of the distal great toe, there was no depression on the dorsal side of the pharyngeal bone. Prior to the surgical procedure, a complete medical history about drug usage, systemic illnesses, and peripheral vascular diseases was taken. Moreover, routine laboratory tests were requested and consent was obtained. Prior to local anesthesia and surgery, the field was prepared and draped. A proximal digital nerve block was done by introducing a

combination of lidocaine and bupivacaine below the dermis. The nerve block was begun by injecting 1 ml within the midway between each side dorsally and ventrally.

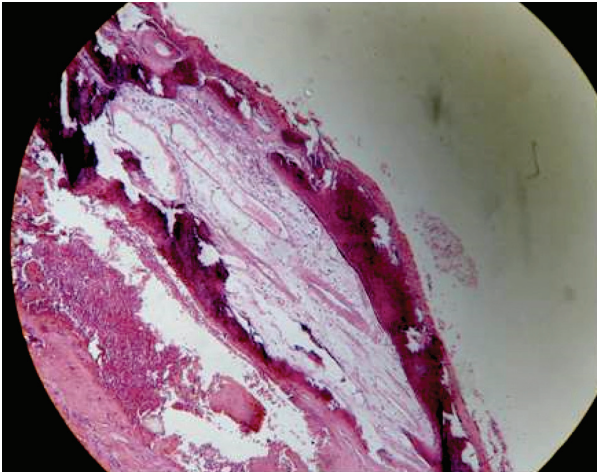
After about 10 minutes of introducing the anesthetic drug, we started to avulse a partial nail plate covering the tumor underside. A compact and firm fusiform soft tissue tumor residing on the nail bed was resected carefully down to the base and the specimen was sent for pathological evaluation. The histopathologic examination showed the nail bed including the epithelium, nail matrix, and hyponychium with a filiform lesion that seemed to be arising from the nail bed. This lesion was composed of a loose and edematous vascular core mildly infiltrated by chronic inflammatory cells, surrounded by calcified trabecular bone (Figure 2). These histopathologic features could be suggestive of a new entity termed 'osteoma mucosalis'.

## DISCUSSION

An osteoma is a benign, slow-growing bone mounted on another bone or non-bone tissue, referred to as a homoplastic or heteroplastic osteoma, respectively. Homoplastic osteomas usually occur in skull bones. Peripheral osteomas arise from the periosteum with unknown etiology.



**Figure 1.** A firm, fusiform, horn-shaped lesion in the distal one-third of the nail bed.



**Figure 2.** Fusiform lesion deposits of mature bone fragments (hematoxylin and eosin staining; 100×).

They are usually asymptomatic with no need for any surgical procedures. However, in symptomatic, growing, or cosmetically disturbing cases, surgical management should be considered. The recurrence rate is low. They usually occur in the mandible bone or nasal and paranasal sinuses (frontal, ethmoidal, maxillary, and rarely sphenoidal). Osteomas rarely occur in maxillary bones like the hard palate and zygomatic arch<sup>7-14</sup>.

Calcifications of mandibular soft tissues are common findings in imaging. Rarely, metastatic ossification and dystrophic calcification can be seen in the nasal septum or polyps<sup>15-17</sup>.

Heteroplastic osteomas can occur in non-bone tissues like skin<sup>18,19</sup> and mucosa<sup>20-47</sup>. There are many reports of heterotopic ossification and calcification in cutaneous<sup>18,19</sup>, oral<sup>20-24</sup>, uro/ano-genital, vaginal<sup>25</sup>, cervical<sup>26</sup>, salpingeal<sup>27</sup>, endometrial<sup>28-35</sup>, orbital<sup>36-39</sup>, and mucosal regions. In addition, there are some cases of gastrointestinal<sup>40-42</sup> and airway (e.g., pulmonary)<sup>43-47</sup> mucosal calcification and ossification in the literature. These entities could be managed conservatively or surgically.

Primary or secondary osteoma cutis is the bone formation in the dermis and/or subcutaneous tissues. This usually presents as multiple, small, and asymptomatic papular lesions in the face and is considered an important cosmetic problem<sup>18,19</sup>.

The nail bed is situated underneath the nail plate. The lunula and hyponychium are the proximal and distal edges of the nail bed, respectively. The epithelium of the nail bed is composed of basal and spinous layers without any granulosum or

corneum layers. There are few melanocytes in the basal layer. The dermis of the nail bed is composed of uniform collagen and elastic fibers in a longitudinal vascular medium. There are also some thermo-regulatory glomus bodies (arteriovenous anastomoses) in the dermis. Based on histopathology, the epithelial and sub-epithelial structure of the nail bed is somewhat similar to that of mucosal tissue<sup>48,49</sup>.

An osteoid osteoma is a benign tumoral lesion arising from the bone and is considered a rare disorder of the nail's bony structure<sup>50</sup>.

An osteoma cutis of the foot is a very rare and reportable entity in the nail. Although in-growing toenails are often seen, they could have some rare or never-before-seen differential diagnoses like an osteoma cutis of the nail<sup>51</sup>.

One of the most common differential diagnoses of nail-related bony structure abnormalities is exostosis. An exostosis is a benign lesion, often appearing in the toe's distal phalanges. It rarely occurs in the fingers or other bones. Squamous cell carcinoma (SCC) of the nail apparatus is the most common neoplasm of this area but represents a rare malignancy overall. It could be misdiagnosed as conditions like in-growing toenail-related granulation tissue, exostosis, wart, an infectious disorder, and pyogenic granuloma<sup>52-56</sup>.

Nails may be affected by many pure dermatologic, non-dermatologic systemic inflammatory, nutritional, or immune-related disorders, as well as benign or malignant neoplastic lesions<sup>57</sup>. Here, we report the first case of a nail bed osteoma. Based on the histopathologic similarity of the nail bed to mucosal tissues and considering the various reports of mucosal ectopic calcification-ossification, we named our observation as 'osteoma mucosalis' of the nail bed and managed it surgically.

The reported case had no underlying disease. Due to the presence of chronic inflammatory cells in the pathological specimen, trauma was assumed as the precursor event of ectopic bone formation in the nail bed. Complete local resection of the lesion was performed to treat this case of osteoma mucosalis. The patient reported no complications or recurrence during six months of follow-up.

In the case of a calcified-ossified subungual lesion, nail bed osteoma could be a rare differential diagnosis of an exostosis, osteoid osteoma, or osteoma cutis.

## CONCLUSION

In osteomas, although bone formation usually occurs in subcutaneous tissues, it can also be seen in mucosal tissues.

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