Intraabdominal heterotopic ossification is a rare sequela of abdominal surgery or trauma. Only a few previous reports have been described in the literature. Here, we report the case of a 20-year-old man who underwent appendectomy 3 years ago and experienced this condition. Initially, he presented with right undescended testis with a palpable mass in the right lower abdomen. Plain radiography and computed tomographic imaging showed peripheral calcified soft tissue mass. Explorative laparotomy for excision was performed and heterotopic ossification was diagnosed by pathologic findings. The characteristic imaging features of this rare entity are discussed and the literature are reviewed.

Heterotopic ossification (HO) is the abnormal formation of true bone within extraskeletal soft tissues. Classically, many diseases sharing this common feature were lumped into the category myositis ossificans; however, the term has fallen into disfavor because primary muscle inflammation is not a necessary precursor for such ossification and the ossification does not always occur in muscle tissue. The term HO has largely replaced myositis ossificans in the literature [1].

Mature HO closely resembles normal bone both histologically and radiologically, and thus it differs from dystrophic calcification in soft tissues, in which there is no osteoblastic activity or lamellar bone. Heterotopic ossification has been reported in the context of several conditions—most notably total hip arthroplasty and neurologic injuries [2, 3]. However, intraabdominal HO is a rare sequela of abdominal surgery or trauma and of unknown etiology. Here, we present a patient who experienced intraabdominal HO after appendectomy.

CASE REPORT

A 20-year-old-young man presented with right undescended testis during a military health examination. No previous trauma was reported but he had a history of an appendectomy 3 years ago. Physical examination revealed one painless palpable mass in the right lower quadrant (RLQ) of the abdomen and nonpalpable right testis at right scrotum and inguinal area. Laboratory findings were within normal limit.

Plain radiography of the abdomen demonstrated a large rim-calcified lesion in the RLQ area (Fig. 1). Computed tomography of the abdomen identified a large peripheral mineralized soft tissue mass with central punctate calcifications (Fig. 2). On the basis of
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the clinical history and radiologic findings, the preoperative diagnosis of an undescended testicular tumor in the RLQ was suggested. An exploratory laparotomy for tumor excision was performed. Macroscopic examination of the resected specimen revealed a $7.5 \times 6 \times 4.5$ cm$^3$ whitish firm mass. Microscopically the tumor was composed of fibroadipose tissue with ossification and calcification (Fig. 3).

**DISCUSSION**

Heterotopic ossification is an uncommon condition and is characterized by new bone formation in a tissue that does not usually undergo ossification. Several mechanisms have been described, but the exact etiology is not clear. HO is usually a benign condition and the main challenge is to differentiate it from other conditions. When HO occurs intraabdominally, however, this condition may have life-threatening complications. Multiple cases of HO after abdominal surgery or trauma are reported. It is confined to the operative incision, adjacent muscle, and connective tissue.

It is quite unusual to see this process in the mesentery and fewer than 20 cases of heterotopic mesenteric ossification (HMO) are described in the English literature [1, 4]. HMO was first reported in a 55-year-old man by Hansen et al in 1983 [5]. The patient exhibited symptoms of small bowel obstruction 2 weeks after he had a coloproctectomy for ulcerative colitis. He was found to have widespread intraabdominal fibrous adhesions and nodules of heterotopic bone in the mesentery. His course was complicated by abscess formation, fistula formation,
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and protracted gastrointestinal dysfunction.

In reviewing the literature, the vast majority of cases involved adult men whose most common presentation was bowel obstruction [5-7]. Both trauma and abdominal surgery have been consistently identified as antecedents to HMO [4]. Fortunately, our patient didn’t experience significant symptoms of bowel obstruction.

The mechanism for intra-abdominal HO is unproven, but its morphologic appearance is quite similar to myositis ossificans [8], and it is likely to occur by a similar mechanism. According to Kaplan et al. [9], four key events are necessary for the development of HO in the soft tissues. The first is an inciting stimulus such as surgical or mechanical trauma; however, ischemia, venous stasis, edema and inflammation have been implicated as well. Next, inductive signaling pathways must be formed at the site of injury. During this process, there must be a population of undifferentiated mesenchymal cells, such as inducible osteoprogenitor cells, that can respond to various signals. Finally, the environment must be suitable for osteogenesis. Only a very small percentage of patients who have surgical or mechanical trauma undergo HO [10].

The differential diagnosis for intra-abdominal mass with calcification after trauma or surgical exploration should include: HO, dystrophic calcification, osseous neoplasia (teratoma, extraskeletal osteosarcoma, chondrosarcoma, etc) and foreign material [1, 11, 12]. Distinguishing among them radiologically may require a high degree of clinical suspicion. If little or no history is available, the diagnosis can be particularly problematic because of its rarity.

Densities with a well-defined cortex and internal trabecular pattern on imaging suggest mature bone rather than dystrophic calcification. Dystrophic calcification typically causes faint radiodense areas that are punctate and irregular [11]. Additionally, mineralized neoplasm such extraskeletal osteosarcoma or extraskeletal chondrosarcoma generally show irregular, immature growth patterns rather than the mature trabecular bone seen in HO [13].

The recognition of HO is important because of its propensity to recur and cause bowel obstruction [6]. Treatments that have been suggested to decrease regrowth include anti-inflammatory medications, disphosphonates, and radiotherapy [7, 12] We were unable to find any cases documented in the literature in which intraabdominal heterotopic ossification has undergone malignant degeneration.

In conclusion, intraabdominal HO is a very rare entity. It can easily be misdiagnosed and may lead to serious complications. Nevertheless, in the differential diagnosis of intraabdominal densities after abdominal surgery or trauma, HO should be considered. It should be distinguished from dystrophic calcification and ossifying neoplasm.

REFERENCE

腹部內異位性骨化十分罕見，我們報告一位 20 歲男性在三年前做過闌尾切除，因為單側陰囊症併有右下腹腫塊而被轉介。X 光素片及電腦斷層顯示環狀鈣化或骨化之腫瘤於右下腹。術前診斷為腹內睾丸腫瘤，腹腔鏡探測腫瘤切除後的病理診斷為異位性骨化。我們在文中討論這種罕見疾病的影像特徵並回顧文獻報告。