

# EPIGASTRIC DISCOMFORT, ABDOMINAL DISTENTION AND NAUSEA HIDING SPINE OSTEIOD OSTEOMA: A CASE REPORT OF GASTROINTESTINAL SYMPTOMS BRINGING TO AN ELUSIVE DIAGNOSIS

A. Bruschi<sup>1</sup>, L. Boriani<sup>1</sup>, P. Spinnato<sup>2</sup>, G. Facchini<sup>2</sup>, A. Gasbarrini<sup>1</sup>

<sup>1</sup>Department of Spine Surgery, IRCCS Istituto Ortopedico Rizzoli, Bologna, Italy

<sup>2</sup>Department of Radiology, IRCCS Istituto Ortopedico Rizzoli, Bologna, Italy

Corresponding Author: Alessandro Bruschi, MD; e-mail: [alessandro.bruschi@ior.it](mailto:alessandro.bruschi@ior.it)

**Abstract** – Spine osteoid osteoma can be a challenging diagnosis. Radiating pain to thorax, abdomen or thigh due to radiculopathy is known to mislead diagnostic path to non-orthopedic conditions. This is the cause of delayed diagnoses and treatments. Therefore, osteoid osteoma should be considered when a nocturnal recurrent pain finds no diagnostic resolution. Gastrointestinal symptoms have never been described as cause of misdiagnosis in spine osteoid osteoma.

This is a case of a 17-year-old girl with recurrent nocturnal back pain with epigastric discomfort, abdominal distension and nausea due to a D12 spine osteoid osteoma. The patient underwent a 12-month diagnostic path for gastroenterological, nephrological and gynecological causes of abdominal pain before reaching the correct diagnosis. Bone scintigraphy and thin slices (0.2 mm) CT scan revealed the spine osteoid osteoma in the left articular process of D12 confirmed by histology. CT guided radiofrequency of the lesion led to a resolution of symptoms. Further investigations revealed *Helicobacter pylori* infection and small intestinal bacterial overgrowth proving that gastrointestinal symptoms were present only when osteoid osteoma was active despite the persistence of bacterial infection. Therefore, hypothesis of spine conditions revealing gastrointestinal diseases due to alteration of microbiota physiology is investigated.

Nocturnal recurrent pain and painful scoliosis should rise the suspicion of spine osteoid osteoma. As shown in this case, even in the absence of radiculopathy, gastrointestinal symptoms can be present and can mislead diagnosis. Moreover, when back symptoms are associated with epigastric discomfort, abdominal distension and nausea, breath tests should be performed in order to avoid missing diagnosis of *Helicobacter pylori* infection and SIBO.

**Keywords:** Osteoid osteoma, Abdominal pain, *Helicobacter pylori*, SIBO, Microbiota, Radiofrequency.

## INTRODUCTION

Osteoid osteoma can be sometimes a challenging diagnosis. Although it is usually located in the appendicular skeleton, spine is involved in 10% of osteoid osteoma<sup>1,2</sup>. In these cases, back pain has differential diagnosis with different abdominal condition and for this it can be some-

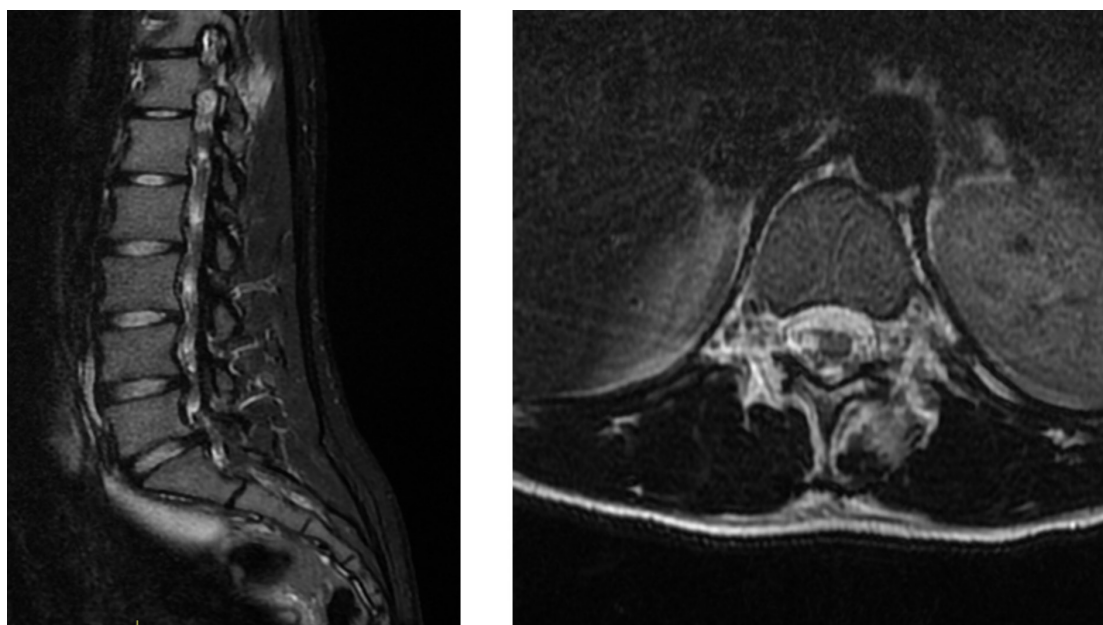


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times misdiagnosed. Other case reports in the literature confirm the diagnostic challenge that spine osteoid osteoma can provide<sup>3-5</sup>. We present a case of a 17-year-old girl with recurrent nocturnal back pain with epigastric discomfort, abdominal distention and nausea who underwent a 12-month diagnostic path for internal and gynecological conditions before reaching the diagnosis of spine osteoid osteoma. Moreover, after the diagnosis, further investigations with urea breath test and lactulose breath test were positive for *Helicobacter pylori* infection and small intestinal bacterial overgrowth (SIBO). Therefore, the hypothesis of gastrointestinal pathologies revealed by spine conditions due to a common innervation is investigated.

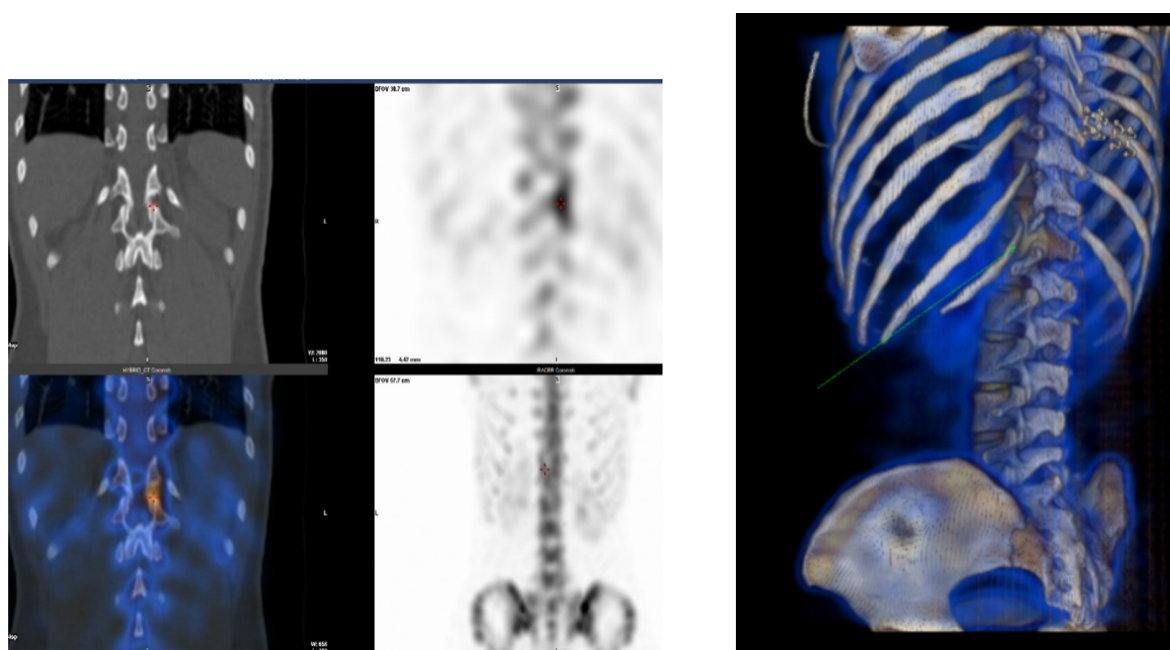
## Case Presentation

A 17-year-old girl presented to orthopedic office visit for recurrent nocturnal back pain that begun twelve months before after a fall riding a horse. In the fall, the patient had a direct contusion of the left thigh and back pain presented soon after. The physical condition of the patient did not require emergency care, but she was immobilized and brought to emergency department for evaluation. The patient had a negative medical history and referred no alcohol or drug consumption. Physical parameters and blood count were normal. Physical examination was positive for diffuse pain at the left dorso-lumbar tract during palpation, with no vascular or nervous abnormalities. Abdominal examination was negative. Spine X-rays were negative for fracture. The pain was relieved with NSAIDs and the patient was dismissed with diagnosis of no fracture, nor abdominal conditions. In the next days, the dorsal pain started to be present at night with epigastric discomfort and episodic nausea. The patient reported that nausea started four months before the first episode of back pain and increased the frequency of presentation after the onset of back symptoms. No symptoms of meteorism were reported. After ten days of these symptoms, the patient presented again to emergency department. Blood count was normal, celiac serologic testing (tissue transglutaminase, endomysial and deamidated gliadin peptide antibodies) already performed under family doctor indication was negative and pregnancy was excluded. However, due to discomfort in the left upper quadrant during palpation and left kidney percussion (Giordano's maneuver) positivity, the patient underwent abdominal ultrasonography with result of 3 mm left caliceal urinary gravel and dismissed with renal colic diagnosis treated with painkillers. In the next months, the patient kept presenting epigastric discomfort and nausea with nocturnal back pain. She underwent a new renal ultrasonography, a complete contrast enhanced thorax-abdomen CT scan and a gynecologic examination in the next three months with no diagnostic benefits. Due to persistent nausea and epigastric discomfort the patient underwent a gastrointestinal endoscopy (EGDS) which resulted normal in all districts examined except for a thinning of the gastric mucosa associated with chronic gastritis. Further, EGDS biopsy, performed in antrum, angulus and corpus using Sydney System, resulted negative for *Helicobacter pylori*. The patient therefore started pantoprazole for gastritis and metoclopramide for the nausea. At orthopedic first office presentation, 12 months after the beginning of symptoms, the patient complained about nocturnal recurrent exacerbation of the pain limiting sleeping. The pain was located in the dorso-lumbar tract and was elicited by intense physical activity during the day. The physical examination showed no scoliosis in orthostatic, right convex dorso-lumbar scoliosis in bending (14° scoliometer) and palpation pain at the left dorso-lumbar tract. X-rays were negative for bone lesions of the spine and ultrasonography of the painful region showed a contracture of the left dorso-lumbar paravertebral muscles. Considering the apparently traumatic genesis of the pain and the still present contracture of the paravertebral muscles, the patient underwent physiotherapy and chiropractic therapy, with few benefits. After a month, due to the persistent nocturnal symptoms, an MRI of the dorso-lumbar spine was done. The MRI was negative for bone lesions but showed an important oedema in the paravertebral muscles left to the D12-L1 intervertebral space with hyperintense signal going through the foramen (Figure 1 and 2). A following bone scintigraphy showed an hypercaptation between D12 and L1 in the left intervertebral space (Figure 3 and 4). A CT scan was made to complete the MRI and scintigraphy findings, but it was negative for bone lesions. Due to the positivity of the bone scintigraphy and the negativity of the latest CT, another CT scan was performed, this time with thin slices (0.2 mm). This CT scan finally showed the cause of

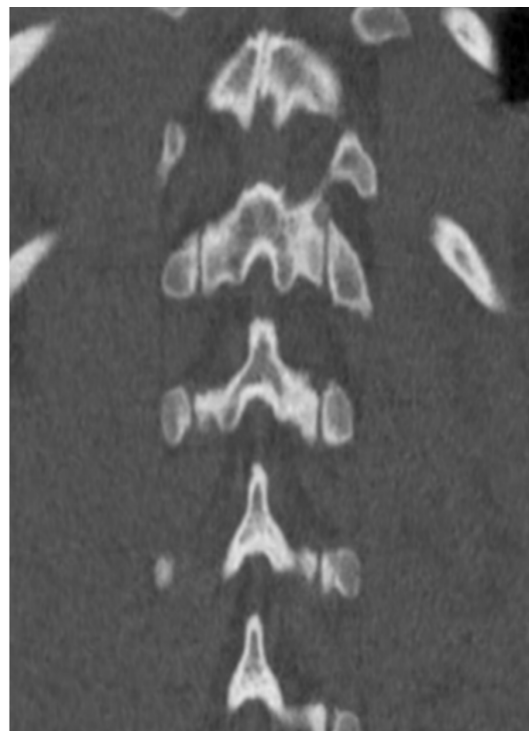


Figures 1 and 2. MRI with oedema in the left dorso-lumbar paravertebral muscles.

the pain: an osteoid osteoma of the left articular process of D12 (Figure 5 and 6). After the diagnosis, the patient underwent CT-guided radiofrequency with intraoperative histological finding confirming osteoid osteoma (Figure 7). The patients reported resolution of the pain immediately after the procedure and no episodes of nausea or abdominal discomfort presented again in the following 18 months follow up (Figure 8). Despite the diagnosis and resolution of clinical condition, gastrointestinal symptoms remained still unexplained. Therefore, patient underwent further investigations with urea breath test and lactulose breath test. Both tests were positive with diagnosis of *Helicobacter pylori* infection and small intestine bacterial overgrowth (SIBO) (Figure 9). The patient has provided informed consent for this case report and her anonymity has been preserved.



Figures 3 and 4. Scintigraphic hypercaptation in the left D12 pedicle.



**Figures 5 and 6.** CT scan: diagnosis of osteoid osteoma of the left articular process of D12.

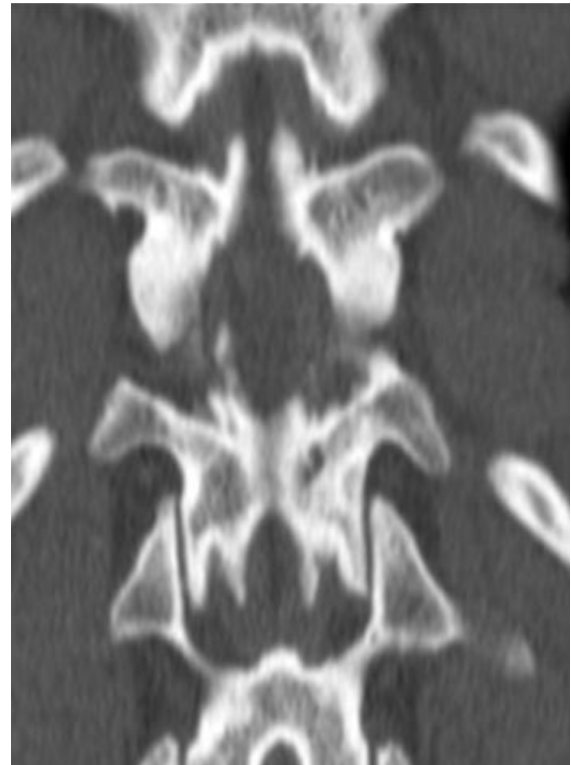
## DISCUSSION

After the fall during a horse riding, the patient's back pain was thought to be linked to the trauma. But in the next days, as the nocturnal pain did not relieve and epigastric discomfort with nausea presented, the patient underwent a diagnostic path in order to find abdominal causes of pain. The positivity of the palpation in the left upper abdominal quadrant and to left kidney percussion (Giordano's maneuver) could have been the consequence of compression of the dorsal region irritated by the osteoid osteoma. Anyway, these findings have been interpreted as an abdominal cause of pain with indication to an abdominal ultrasonography. The finding of the 3 mm renal gravel in the ultrasonography, pivoted the attention to urological problems with a renal colic. It is unlikely that nausea was linked to NSAIDs-related

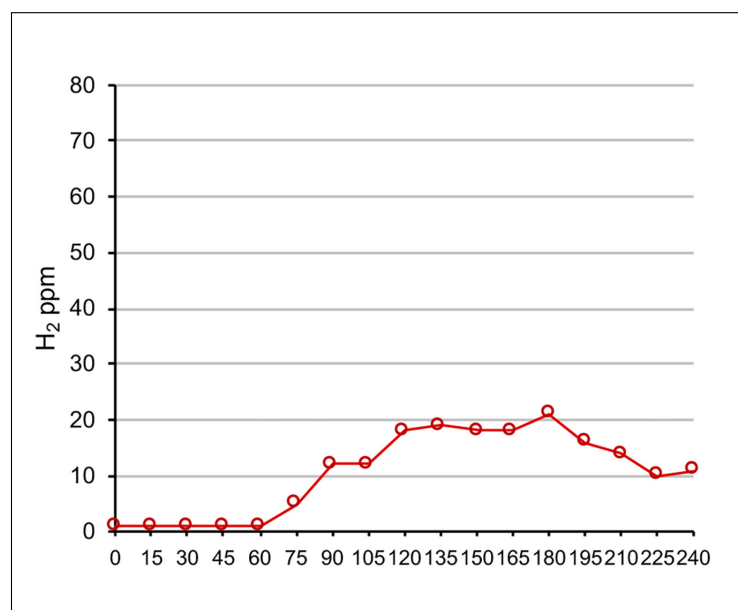


**Figure 7.** Treatment with radiofrequency on osteoid osteoma of D12 left pedicle.

**Figure 8.** Post treatment CT scan.



gastritis as nausea was already present four months before the assumption of painkillers for back pain. The persistence of this symptom shifted the attention to gastroenterological problems and gastroscopy with biopsy was performed with suspicion of gastritis, resulting negative. A complete thorax-abdomen CT scan discarded urological or gastroenterological organic conditions. Gynecological context was investigated with no results as well. This created a lengthening of the diagnostic resolution as a bone genesis of the pain was discarded. In fact, as the last CT scan showed, the cause of the pain was an osteoid osteoma of the left articular process of D12. The diagnostic CT scan was independently seen by three radiologists. Just one of the three succeeded in finding the nidus of the osteoma osteoid in a sagittal



**Figure 9.** Lactulose breath test diagnostic for SIBO.



plan view, due to a localization difficult to assess. It is well documented that the symptoms of spine osteoid osteoma are back pain with tendency to exacerbate during night and to be relieved by NSAIDs<sup>6</sup>, painful scoliosis in more than half of the patients<sup>7,8</sup> and radiculopathy<sup>4,9</sup> in up to 50% of cases<sup>10</sup>. In our case, we think that the epigastric discomfort with nausea mislead the diagnostic path. There are no reported cases of spine osteoid osteoma presenting with these symptoms. There are few case reports about differential non-orthopedic diagnosis with osteoma osteoid. Dhaliwal et al<sup>3</sup> published a case of a 33-year-old man with abdominal pain caused by a spine osteoid osteoma with left D11 radiculopathy radiating to the left abdominal wall. Zenmyo et al<sup>4</sup> reported to cases of prostaglandin mediated radiculopathy resulting in sciatica and thoracic pain caused by spine osteoid osteoma. Another case of initially undefined thoracic pain was linked to a left D11 pedicle osteoid osteoma with radiating pain<sup>11</sup>. A case of cervical osteoid osteoma has been reported for having been diagnosed and treated as tonsillitis before reaching the correct diagnosis<sup>5</sup>. Due to the localization between D12 and L1, the epigastric discomfort, abdominal distension with nausea in our case needs could be considered related to *Helicobacter pylori* and SIBO diagnosis. Our opinion is that the gastrointestinal symptoms have been revealed by the inflammatory response linked to osteoid osteoma. Actually, it is realistic to think that *Helicobacter pylori* infection and SIBO were already present before the onset of symptoms despite being asymptomatic<sup>12,13</sup>; moreover, once treated osteoid osteoma, gastrointestinal symptoms disappeared. Nevertheless, breath tests were still positive 18 months after radiofrequency. This proves that gastrointestinal symptoms were present only when spine osteoid osteoma was active, despite bacterial contamination being still present after its treatment. We think that this could be explained by a common innervation involving spine and abdominal organs interfering with microbiota physiology. It is very uncommon that the epigastric discomfort of our case was linked to a radiculopathy, as the root interested should have been the D12 root; in fact, the pubic region receives D12 root dermatomeric innervation and the patient never complained about pain in that area. So, in our case the epigastric discomfort and nausea mislead the diagnostic path; on the other side, the recurrent nocturnal pain, the typical symptom of osteoid osteoma, was finally helpful in having the correct suspicion to reach the final diagnosis during the orthopedic evaluation. Painful scoliosis was present in this patient and, as primary idiopathic scoliosis is rarely symptomatic<sup>14</sup>, we found this presentation a favorable element to our suspicion of spinal osteoid osteoma.

Osteoid osteoma is a benign bone lesion. It accounts for the 3% of all bone tumors and for the 11% of the benign bone tumors and it shows a male predilection, in the second decade of life<sup>6,15-17</sup>; it is mainly located in the appendicular bone or in the posterior arch of the vertebrae<sup>6,15</sup>; the characteristic symptom is pain with nocturnal exacerbation and tendency to be relieved by NSAIDs<sup>6</sup>. When it is close to a joint, it can cause a swelling of the joint and motion difficulties<sup>18</sup>; when it is in the spine, it can cause stiffness of the paravertebral muscles with secondary scoliosis<sup>7,8,18</sup>, as in this patient; in spine osteoid osteoma radiculopathy is common<sup>4,9</sup> and it is present in up to 50% of cases in this paper<sup>10</sup>. The pathognomonic radiological finding is a "nidus", a small (less than 2 cm) rounded osteolytic area surrounded by a halo of sclerotic bone<sup>15</sup>. Intramedullary osteoid osteoma or osteoid osteoma of spine, pelvis and small bones, cannot easily be assessed with plan radiography, therefore, the CT scan is considered the method of choice<sup>6</sup>. MRI is very useful for showing the important oedema caused by this lesion, but sometimes it can be misleading, as the oedema hinders the characterization of the bone lesion<sup>6</sup>. Scintigraphy is a very sensitive technique, helpful for confirming the suspicion<sup>19</sup>. Nowadays, the gold-standard treatment is CT-guided percutaneous radiofrequency, with 90% of success rate due to pain resolution; intralesional curettage remains an option in cases refractory to radiofrequency<sup>18</sup>.

## CONCLUSIONS

Spine osteoid osteoma can be a challenging diagnosis. Nocturnal recurrent pain and painful scoliosis should rise the suspicion of spine osteoid osteoma. Spine osteoid osteoma can be present with radiating pain in another region, such as abdomen, thorax or thigh due to radiculopathy. As shown in this case, even in the absence of radiculopathy, gastrointestinal

symptoms can be present and can mislead diagnostic path. Moreover, when back symptoms are associated with epigastric discomfort, abdominal distension and nausea, breath tests should be performed in order to avoid missing diagnosis of *Helicobacter pylori* infection and SIBO.

### Conflict of Interest

The authors declare no conflict of interest.

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