СЛУЧАЙ ИЗ ПРАКТИКИ

TRANSFORMATION OF AN OSTEOID OSTEOMA OF T2 VERTEBRA INTO AN OSTEOBLASTOMA AFTER RADIOFREQUENCY ABLATION – A RARE OCCURRENCE - A CASE REPORT AND LITERATURE REVIEW

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We present a case of a young caucasian male who had a typical presentation of an osteoid osteoma at T2 level with characteristic imaging features of a small nidus on cross-sectional imaging in the left T2 pedicle. An elective, uncomplicated CT-guided radiofrequency themoablation was performed at the presenting hospital.

Within four weeks, he represented with recurrent symptoms and repeat imaging at eight weeks demonstrated a large expansile and aggressive lesion at the previous site with aneurysmal bone cyst formation. A needle biopsy confirmed a diagnosis of osteoblastoma which was treated with en-bloc resection (hemivertebrectomy) at the regional bone tumour centre. We propose that the primary lesion was indeed an osteoid osteoma due to its typical imaging features. The subsequent imaging characteristics of the lesion were consistent with an osteoblastoma rather than a simple recurrence.

Osteoblastomic transformation of osteoid osteomas has been described previously in literature in both axial and appendicular skeleton but never within such a short period after thermocoagulation. This rare occurrence should be considered and would warrant imaging surveillance particularly with worsening clinical features by clinicians managing this condition.

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Keywords: osteoblastoma (OB), osteoid osteoma (OO), radiofrequency ablation (RFA).

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ТРАНСФОРМАЦИЯ ОСТЕОИД-ОСТЕОМЫ ВТОРОГО ГРУДНОГО ПОЗВОНКА В ОСТЕОБЛАСТОМУ ПОСЛЕ РАДИОЧАСТОТНОЙ АБЛЯЦИИ – РЕДКО ВСТРЕЧАЕМАЯ ПАТОЛОГИЯ. СЛУЧАЙ ИЗ ПРАКТИКИ И ОБЗОР ЛИТЕРАТУРЫ

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В статье представлен случай типичной клинической картины остеоид-остеомы на уровне Th2 позвонка с характерными признаками при лучевом исследовании в виде маленького очага в области левой дужки позвонка Th2 у молодого мужчины кавказкой национальности. В нашем госпитале ему была проведена радиочастотная термоабляция очага под КТ-навигацией в плановом порядке.

Через четыре недели пациент обратился в госпиталь с жалобами на повторяющиеся симптомы, через 8 недель пациенту было проведено повторное обследование и в той же области были выявлены выраженные повреждения костной ткани с формированием кистозной трансформации. Биопсия подтвердила диагноз остеобластомы, после чего пациенту была проведена гемиэктомия позвонка в региональном онкологическом

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Основываясь на характерных признаках лучевого обследования, мы предполагаем, что первичным поражением позвонка являлась остеоид-остеома. Рентгенологические признаки при последующем исследовании указывали на развитие остеобластомы, а не на рецидив заболевания.

Ранее в литературе описывались случаи остеобластической трансформации остеоид-остеомы осевого и аппендикулярного скелета, но никогда - за такой короткий период времени после термокоагуляции. Редкая частота возникновения остеобластической трансформации всегда должна приниматься во внимание, обеспечивая наилучший результат лучевого обследования.

Ключевые слова: остеобластома, остеоид-остеома, радиочастотная абляция.

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Osteoid osteoma is a benign reactive bone lesion most frequently seen in adolescents with a slight male preponderance [2]. It was first reported in 1935 by Jaffe1. Approximately 10-25% of osteoid osteomas occur in the spine, with more than two thirds (87%) involving the posterior elements particularly the lamina [27, 29, 30]. The clinical presentation is typically of localized pain classically at night and relieved by anti-inflammatory medications. It is a recognized cause of painful scoliosis with the lesion typically located in the concavity of the curve [28].

Osteoid osteoma and osteoblastoma are considered to be closely related processes which are histologically similar 2. Osteoblastomas are larger in size (more than 1.5-2cm) and exhibit an aggressive growth pattern with localized expansion and destruction which is also often associated with intra-lesional haemorrhage and aneurysmal bone cyst formation [16, 26, 27]. Previously these have been described as 'giant osteoid osteomas' but the name 'benign osteoblastoma' was first coined separately in 1951 and 1956 by Lichtenstein and Jaffe [7, 8, 22]. More than one third of the cases of OB are seen in the spine, with the lumbar spine most commonly affected [16]. Extraosseous growth and paravertebral mass or muscular oedema have all been described with concomitant neurological symptoms.

Case Report:

A 19 year old Caucasian male was referred to a tertiary care hospital with a one year history of worsening upper thoracic backache and left arm pain. An osteoid osteoma with a small nidus was found on computed tomography, at the junction of the left pedicle and posterior vertebral body of T2 with associated reactive sclerosis (fig. 1). There were supportive MR imaging features of reactive marrow oedema extending into the vertebral body with no neurological compression. The patient was considered appropriate for a CT-guided radiofrequency ablation (RFA) of the lesion, under general anaesthesia. It was performed electively as a standard procedure, via a posterior approach (fig. 2) at the presenting hospital by an experienced radiologist (with over 15 years of consultant experience). There were no immediate complications and the patient discharged home on the same day.

One month after the procedure, the patient started developing recurrent symptoms which were concerning for recurrence. At his first clinical follow-up, repeat CT and MR imaging were performed. The CT showed a large destructive lesion with scalloping the left lateral border of the T2 vertebral body and associated sclerosis and soft tissue ossification (fig. 3). The MRI revealed presence of a large complex enhancing lesion centered on the left T2 pedicle, with some fluid-fluid levels suggestive of intra-lesional haemorrhage and consequent aneurysmal bone cyst-like (ABC) change, with encroachment into the left neural foramen (fig. 4). A differential diagnosis of post-operative infection or transformation into osteoblastoma was considered. The lesion was certainly larger to be re-treated with radiofrequency ablation and patient was referred to the regional bone tumour centre.

A CT-guided biopsy was performed that showed numerous small tumour fragments made up of anastomosing trabeculae of woven bone rimmed by plump osteoblasts set in a vascularized fibrous background. These features were consistent with the diagnosis of an osteoblatoma rather than osteosarcoma or infection. The patient thereafter underwent an enbloc excision by partial vertebrectomy, laminectomy and costotransversectomy via a posteror approach (fig. 5). The histopathological diagnosis of an osteoblastoma with aneurysmal bone formation was confirmed on the resection specimen.

The patient made a slow postoperative recovery which was complicated by a moderate sized left haemothorax. He was symptom free at three months postoperatively with the MR imaging showing a tiny residual enhancing lesion with ABC change which remained stable on subsequent imaging, at six months post-operatively.

Discussion:

Osteoid osteomas and osteoblastomas are osteoblastic lesions with similar histopathological features but differences in imaging morphology and behaviour. Osteoblastomas are typically larger and destructive in nature and in the spine are often associated with neurological symptoms [14].

The imaging features in our patient on the initial presentation were typical of an osteoid osteoma with a well defined tiny nidus at the junction of the vertebral body and the left pedicle. The classic features of reactive sclerosis and marrow oedema were also present [15, 16, 17]. Although OO-mimicking lesion are described in literature but the smaller size with robust surrounding osteosclerosis and absence of a periosteal reaction were features favouring the diagnosis of an osteoid osteoma [10, 13]. The lesion was treated with a standard CT-guided RF thermocaogulation via a posterior approach under general anaesthesia. This has been increasingly recognised as an effective and preferred modality of treatment for isolated osteoid osteomas in the axial as well as appendicular skeleton [4, 5, 12].

The repeat imaging at the second presentation at two months interval demonstrated an entirely different lesion with localized bone destruction and soft tissue ossification, which could not be accounted for by post-RFA change. The presence of a paravertebral enhancing soft tissue lesion with ABC change was again suggestive of an osteoblastoma, as previously described in literature [11, 16, 26]. Osteoblastomic transformation or progression of an osteoid osteoma is a rare albeit a well documented complication [18, 19, 20, 21, 23]. All the cases reported in literature have occurred after conservative, medical or surgical management and none of these occurred soon after radiofrequencty ablation.

Sung et al described a case of an OO (in 1979) at L3 level which was surgically treated with resection of the left inferior articular facet [23]. The patient presented 16 years later with new symptoms and a much larger 5x10 cm, aggressive paraspinal osteoblastic lesion extending from the posterior elements of L3 - L5 was found. Unfortunately, the patient could not survive the surgery but a resection biopsy did reveal a benign osteoblatoma. The authors proposed a gradation classification based on the presence of benign or aggressive features.

In 1983, Pieterse et al described a case of OO in the right knee of a 14 year old boy with a 5mm radiolucent nidus in an atypical location of the lateral tibial epiphysis [21]. This was surgically resected with histopathologocal confirmation of an osteoid osteoma. The patient represented with night pain at 2 months and the radiograph showed a larger lytic / sclerotic lesion which was again resected showing histopathological features of an osteoblastoma. Six weeks later, there was a larger sclerotic lesion consistent with an osteoblastoma. Another resection was performed which showed aggressive histopathological features regarded as an aggressive or low grade malignant osteoblastoma with no frank sarcomatous change. Four months after the third operation, there was re-appearance of the a pedunculated sclerotic lesion with a separate lesion in the distal lateral femoral epiphysis. The patient underwent an above knee amputation which again demonstrated features of aggressive multifocal osteoblastoma. This case is very atypical with an epiphyseal location with lack of cross-sectional imaging and an above-knee-amputation as the final management, however the rapid aggressive transformation is worth noting.

Morton et al reported a case of a recurrent sclerotic lesion in the right second metacarpal in a 30 year old man [24]. He underwent enbloc resection for a presumed diagnosis of OO with iliac grafting. This developed subsequent aggressive features with involvement of adjacent third and fourth metacarpal bases and the capitate. The patient underwent a total of 11 operations with histological features more typical of an aggressive osteoblastoma rather than an osteoid osteoma or a slow growing osteosarcoma.

Bettelli et al, in 1991, reported two cases of recurring osteoblastomas in the appendicular skeleton, which were initially treated as typical osteoid osteomas [20]. The first case was in a 24 year old lady with a typical OO in the medial cortex of the right femur which was treated with excision and drilling. The histology confirmed osteoid osteoma, but the patient represented eight months later with a dense lesion measuring about 10 cm of the mid-third femur with few lytic areas.

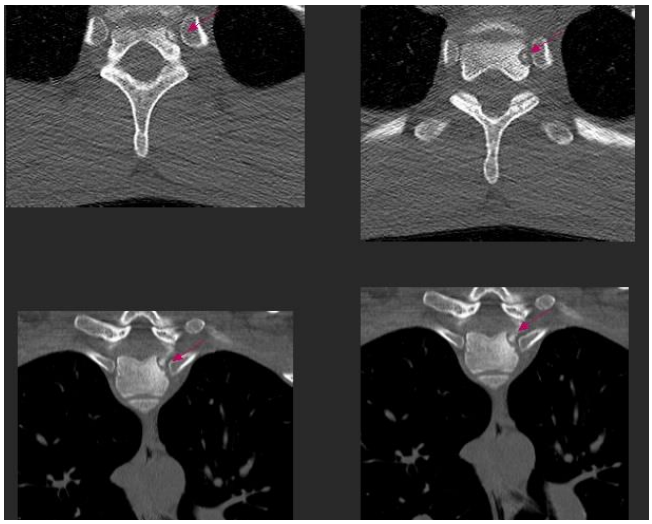

Fig. 1, a.

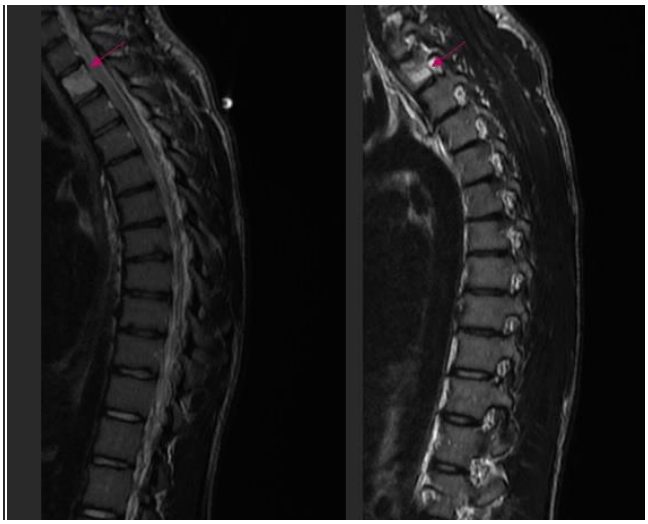

Fig. 1, b.

Fig. 1. CT, axial and coronal images. MR, Sagittal T2W images.

On presentation demonstrating the nidus of the osteoid osteoma (red arrows) in the posterior T2 vertebral body which shows some reactive sclerosis and marrow oedema.

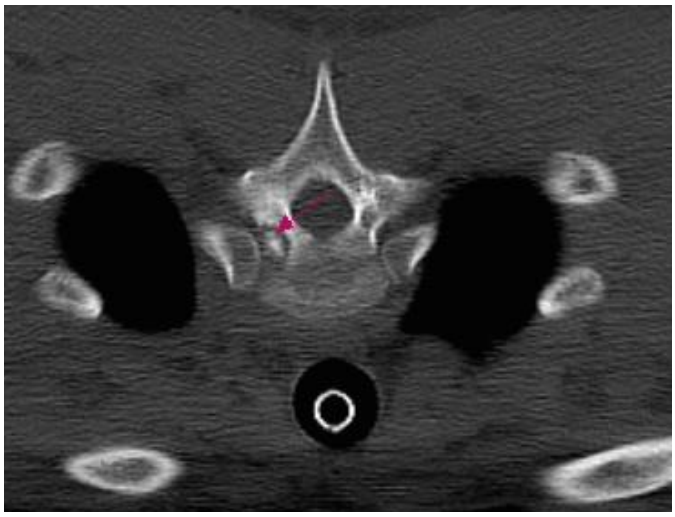

Fig. 2, a.

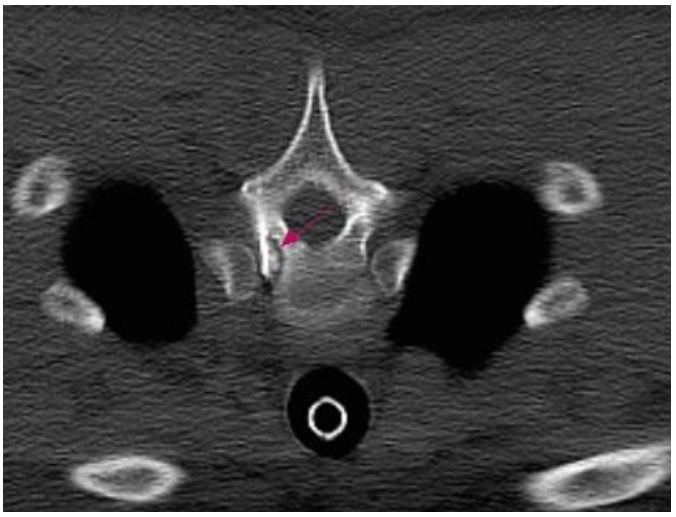

Fig. 2, b.

Fig. 2. CT, axial images.

Showing the nidus and the RFA probe sited within the nidus. The procedure was done under general anaesthesia.

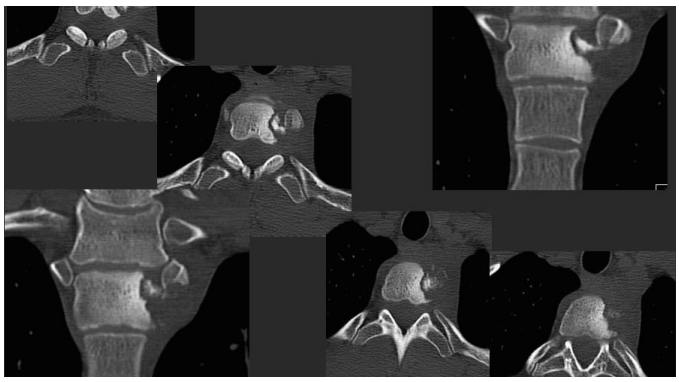

Fig. 3.

Fig. 3. CT images.

At 2 months post-RFA showing a locally destructive and larger lesion (osteoblastoma).

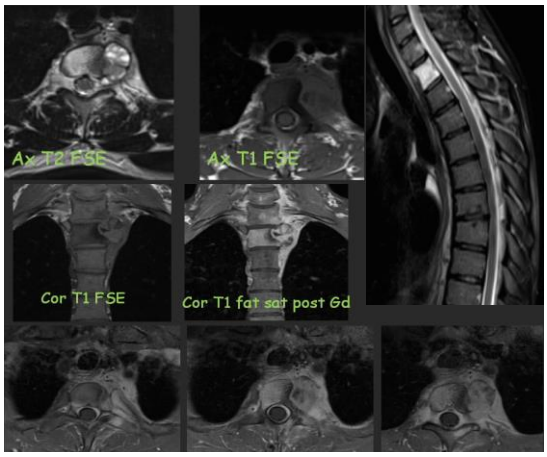

Fig. 4.

Fig. 4. MR images.

At 2 months post-RFA showing a large lesion with ABC like features (osteoblastoma). The bottom row images are Postcontrast Ax T1 fat saturated, demonstrating the enhancing left paraspinal lesion with encroachment into the left neural foramen.

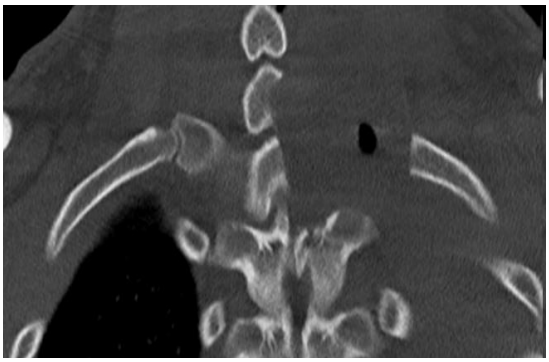

Fig. 5, a.

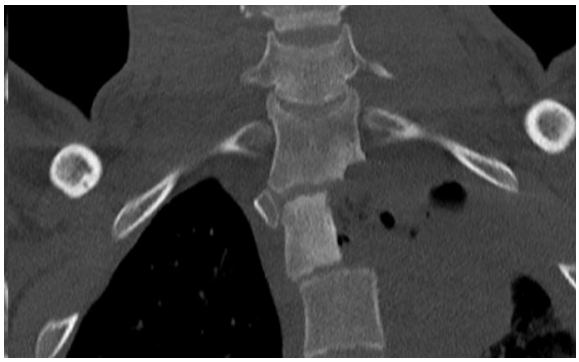

Fig. 5, b.

Fig. 5. CT images.

Immediate postsurgical coronal images showing the enbloc excision with a T2 partial vertebrectomy; at 4 months after the first radiofrequency ablation. There is an associated moderate sized left haemothorax, which resolved after few weeks.

The MR imaging features were suggestive of an aggressive lesion and a diaphyseal resection and autograft reconstruction was performed, with a histopathological confirmation of an osteoblastoma. The second case was of an 18 year old man, with a 10mm lytic lesion in the medial cortex of the left femoral neck. The lesion was excised as an osteoid osteoma, with the patient representing fourteen month later with a visible groin mass. A large lytic lesion was seen on cross-sectional imaging and resection specimen confirmed a histopathological diagnosis of osteoblastoma. These two cases of appendicular OO resemble our case in that both were typical of OO on their initial presentation but evolved into a recurrence which showed markedly aggressive features suggesting of osteoblastomic transformation.

Bruneau et al in 2005, described a case of an osteoid osteoma at the left posterolateral mass of C1 in a 25 year old man [18]. At initial presentation, there was an 8mm radiolucent nidus with surrounding sclerotic bone on CT, in proximity to the vertebral artery. The patient was managed medically with anti-inflammatory medications for his occipitocervical pain. Seven years later, there were progressive symptoms with reduced neck mobility. Repeat CT imaging showed doubling in size of the lesion, measuring 16mm, with features suggestive of an osteoblastoma. The patient underwent an open resection via an anterolateral approach and transposition of the left vertebral artery with opening of the C1 transverse process. The lesion was drilled and osteoblastoma was confirmed on histopathological examination. This case again demonstrates the need for a low threshold of re-imaging patients with spinal OO particularly with progressive symptoms. A multimodality approach is essential in managing these cases with technically challenging locations. Occasionally pre-operative embolization can be employed as a useful adjunct prior to surgical resection 12.

The most recent case of a cervical osteoid osteoma progressing to an osteoblastoma has been described by Cappuccio et al [19]. They reported a case of an OO of the right pedicle of C6 in a 16 year old boy with typical CT and MR imaging features. The patient declined an operation initially but represented with worsening symptoms one year later. Repeat CT imaging showed expansion of the lesion, with local destruction of the articular process, and involvement of the C6 nerve root and the vertebral artery. An intralesional excision with posterior C4-7 fusion was performed. The histolo-

gy was consistent with osteoblastoma ; with no recurrence seen at five years follow-up. This case shows spontaneous transformation of an OO into an osteoblastoma. Our case was treated with RF ablation with a favorable immediate result. The rate of recurrence in osteoid osteomas after radiofrequency ablation is quoted to be approximately 7 per cent, with higher rates in younger patients [25, 27]. The different imaging features at 8 weeks post-ablation were suggestive of a transformation into an osteoblastoma rather than a recurrence.

In conclusion, our case is peculiar in the list of previous reports of osteoblastoma arising in an osteoid osteoma as none occurred after an RF themo-ablation. The interventional or musculoskeletal radiologists performing RFA for osteoid osteomas should be aware of this complication in addition to recurrence. A low threshold to perform repeat imaging is advised, particularly if the patient develops recurrent or new symptoms. The presence of aggressive feature and ABC-like change should warrant a close liaison with the spinal surgical team, for further management of this particular subset of patients.

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