

CASE REPORT

직장암 수술 후 하부 장간막동맥 절단부의 외상신경종: 증례 보고 및 문헌 고찰

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Traumatic Neuroma at the Inferior Mesenteric Artery Stump after Rectal Cancer Surgery: A Case Report and Literature Review

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Traumatic neuroma results from regeneration attempts of the proximal end of an injured or severed nerve, resulting in a non-neoplastic nodular lesion. The lower extremity after amputation is the most common site, followed by the head and neck. Traumatic neuromas occurring in the abdomen, however, are rare. In the abdominal region, traumatic neuromas occur in the cystic duct stump and the common bile ducts as well as around the celiac trunk. This study reports a case of a 59-year-old man who presented with a traumatic neuroma arising at the stump of the inferior mesenteric artery after rectal cancer surgery. Traumatic neuromas at the stump of the inferior mesenteric artery have not been previously reported. The lesion exhibited atypical imaging features, including a well-enhanced nodule, a significant interval growth in size and a mild increase in 18F-fluorodeoxyglucose uptake, resembling lymph node metastasis. This case report will help physicians understand the sites of occurrence and imaging features of traumatic neuromas in the abdomen. (*Korean J Gastroenterol* 2016;68:279-283)

Key Words: Neuroma; Trauma; Injuries; Rectal neoplasms; Inferior mesenteric artery

INTRODUCTION

Traumatic neuroma occurs by regeneration attempts of the proximal end of an injured or severed nerve, resulting in a non-neoplastic nodular lesion.^{1,2} The lesion typically arises one to 12 months after transection and exhibits slow or no interval size changes over months to years.³ This condition was first described by Odier in 1811 in war-wounded veterans with amputated limbs presenting with disabling pain.^{4,5} Traumatic neuromas can occur in any part of the body. The

lower extremity after amputation is the most common site, followed by the head and neck (frequently after tooth extraction or radical dissection).⁶ Traumatic neuromas occurring in the abdomen, however, are rare.

In the abdominal region, traumatic neuromas occur in the cystic duct stump and less commonly in the common bile ducts (CBD), or a choledochointestinal anastomotic site after cholecystectomy, bile duct surgery or blunt abdominal trauma.⁷⁻¹² Other sites reported are around the celiac trunk after gastrectomy or in the rectal wall after polypectomy.^{13,14}

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We present a case of traumatic neuroma at the stump of the inferior mesenteric artery (IMA) that has rapid interval growth in size and hypermetabolic activity in a patient with prior rectal cancer surgery. To the best of our knowledge, this is the first report of traumatic neuroma occurring after rectal cancer surgery.

CASE REPORT

A 59-year-old Korean male underwent an ultra-low anterior resection (U-LAR) with colo-anal anastomosis for rectal cancer (an annular constricting mass located 5 cm above the anal verge, 4 cm segment involvement of the mid-rectum). He did not have any underlying disease or significant medical history at the time of diagnosis of rectal cancer. According to the TNM staging system of the American Joint Committee on Cancer and the International Union Against Cancer, the diagnosed rectal cancer was clinically determined as stage 2 (cT3N+M0). After neoadjuvant chemoradiotherapy, the lesion was downgraded to stage 1 (ypT2N0M0), which was surgically confirmed. The patient received adjuvant chemotherapy after U-LAR.

The patient exhibited no apparent symptoms, and physical examinations remained normal during later visits. Laborato-

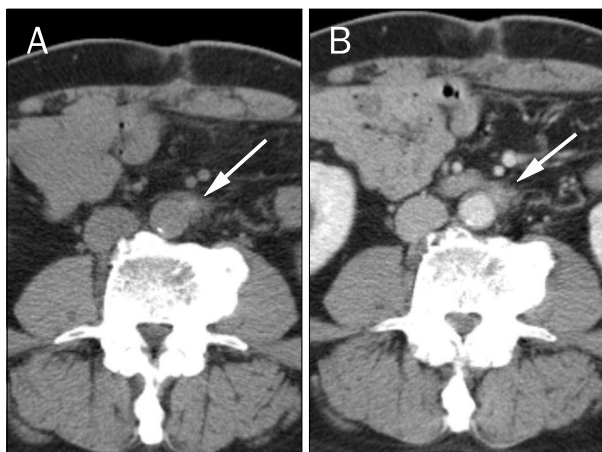


Fig. 1. An ill-defined left para-aortic lesion on CT images obtained two weeks after surgery. (A) Non-contrast CT reveals an ill-defined infiltrative para-aortic soft tissue attenuation lesion (arrow, 30.53 ± 27.25 HU) at the inferior mesenteric artery stump area. (B) Portal-venous phase CT indicates that the lesion (arrow) is persistently enhancing (53.64 ± 28.65 HU). This lesion was interpreted as a postoperative granulation tissue because the occurrence of a lesion at a stump is considered a typical finding.

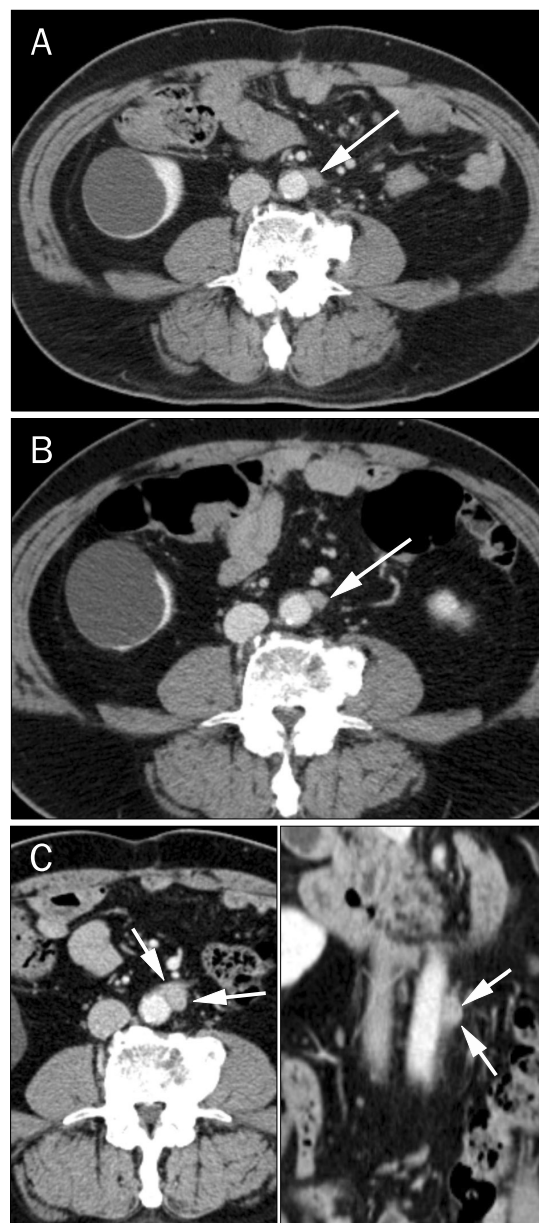


Fig. 2. A left para-aortic lesion changing into a discrete nodule on follow-up CT scans. (A) Portal-venous phase CT obtained seven months after operation depicts a 1.2 cm discrete enhancing nodule (arrow) at the inferior mesenteric artery stump area (73.17 ± 10.37 HU). (B) Portal-venous phase of a contrast-enhanced CT obtained 20 months after operation depicts a 1.2 cm discrete enhancing mass (arrow) at the inferior mesenteric artery stump area (96.16 ± 14.04 HU) at the portal-venous phase. No significant interval change in size is noted compared with the previous CT (Fig. 2A). (C) Portal-venous phase of a contrast-enhanced CT obtained 32 months after operation reveals a 1.8 cm discrete enhancing mass (arrows) at the inferior mesenteric artery stump area (110.00 ± 14.30 HU) at the portal-venous phase. An interval increase in size is noted compared with the previous CT (1.2 cm to 1.8 cm). Coronal portal-venous phase image also reveals a well-demarcated slightly elongated mass (arrows) at the left para-aortic area.

ry tests revealed no abnormal findings, and carcinoembryonic antigen level ranged from 0.5 ng/mL to 2.3 ng/mL (normal range, < 5.0 ng/mL).

In the first postoperative CT obtained two weeks after U-LAR, a soft tissue lesion at the stump of IMA was detected (Fig. 1). The lesion had an ill-defined margin and was slightly contrast-enhanced. The lesion, which was irregular in margin and slightly contrast-enhanced, was diagnosed as a post-operative granulation tissue. When viewed at seven and 20 months after U-LAR, the lesion changed into a nodular, well-circumscribed, homogeneously enhancing lesion measuring 1.2 cm in diameter (Fig. 2A, B). By 32 months, the lesion size increased from 1.2 cm to 1.8 cm in diameter (Fig. 2C). The interval change in size raised a strong suspicion of a metastatic lymph node.

A PET-CT scan revealed that the lesion was hypermetabolic (standardized uptake value [SUV], 2.87) (Fig. 3). This finding strengthens our suspicion of a metastatic lymph node. To confirm whether the lesion was a nodal metastasis, a surgical biopsy was performed. Histological examination of the surgical specimen revealed haphazardly arranged proliferation of variably sized nerve fascicles, including axons, Schwann cells, and fibroblasts, but no evidence of malignant cells (Fig. 4). The lesion was pathologically confirmed as traumatic neuroma.

DISCUSSION

Most intra-abdominal traumatic neuromas are asymptomatic.^{4,13,14} Some traumatic neuromas cause symptoms, such as epigastric pain, vomiting, or obstructive jaundice, with varying degrees and time of onset.⁸ Although asymptomatic traumatic neuromas do not require treatment, as their natural history is benign, accurate differentiation of traumatic neuromas from recurring tumors or metastatic lymph nodes in the clinical context of prior malignancy is crucial.^{1,2,4,8,13}

Imaging is one of the most valuable examination methods, paired with pathologic examination, to confirm the diagnosis of traumatic neuromas.^{1,2,4,15} Radiologic descriptions of traumatic neuromas are primarily based on cases involving amputated extremities or the head and neck.^{1,3,6,15} Characteristics of traumatic neuromas on ultrasonography include a small short/long diameter ratio, a small short-axis diameter, and the presence of a central hyperechoic area.² On CT imaging, traumatic neuromas are characteristically stable nodules with a low attenuation.^{1,2} MRI features of traumatic neuromas include a signal intensity that is similar to that of muscle on T1-weighted images, contrast enhancement, and slightly heterogeneous, intermediate to high signal intensity frequently combined with a hypointense rim on T2-weighted images.^{2,3,15} On metabolic imaging using 18F-fluorodeoxyglucose (FDG) PET, traumatic neuromas exhibit no increased uptake of FDG.^{13,16}

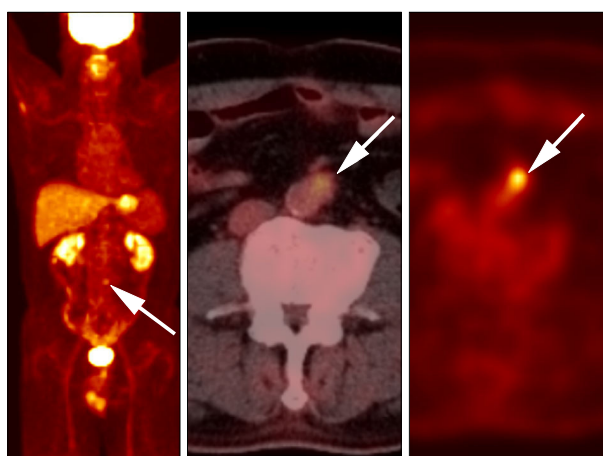


Fig. 3. A hypermetabolic lesion at the left para-aortic area. A 18F-fluorodeoxyglucose PET-CT scan depicts a hypermetabolic lesion (arrows) at the left para-aortic inferior mesenteric artery stump area (standardized uptake value, 2.87).

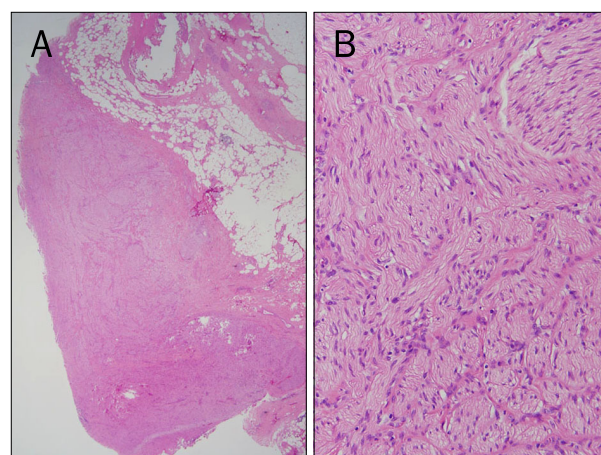


Fig. 4. Microscopic findings (H&E). (A) The lesion is composed of variably sized proliferating nerve fascicles in a background of collagen and adipose tissue ($\times 12$). (B) The image reveals haphazard proliferation of nerve fascicles including axons, Schwann cells, and fibroblasts ($\times 200$).

Although understanding the imaging characteristics of traumatic neuroma is important to make a correct diagnosis and differentials, the imaging features need further description in the literature. In a report of traumatic neuromas occurring in four patients after neck dissections, the characteristic CT features were described as a nodule with a central hypoattenuation, a hyperattenuating rim, and intact overlying fat.¹ However, another author asserted that a hyperattenuating rim on CT cannot be used as a differentiating radiologic feature because this feature is observed in both traumatic neuroma and recurrent lymphadenopathy.² Traumatic neuroma is currently described as a stable nodule with low attenuation on a contrast-enhanced CT scan that occurs one to 12 months after the clinical setting related to its development, trauma or surgery.^{2,13}

The MRI features of traumatic neuromas are described in several studies.^{2,3,15} Although no significant difference in the signal intensity on T1- and T2-weighted images is apparent between traumatic neuroma and recurrent lymphadenopathy, a hypointense rim on T2-weighted MRI is seen only in traumatic neuromas.^{2,3} A recent review of 13 subjects with 20 neuromas concluded that traumatic neuromas exhibit enhancement and lack a T2 hyperintense rim surrounding a central area of low signal, called a target sign.¹⁵

Altered glucose metabolism is characteristic of numerous malignancies, and FDG-PET is increasingly used for staging, restaging, detecting recurrence or metastasis, or differentiating between benign and malignant lesions. However, because FDG uptake is not tumor-specific, false positive results are possible in many non-neoplastic and benign diseases, such as inflammation or posttraumatic repair.¹⁶ Compared with other conventional modalities, data on FDG-PET features of traumatic neuromas have been extremely limited. The report that traumatic neuromas exhibited no increased uptake on FDG-PET scans is consistent with the natural history of traumatic neuroma as a benign entity.¹³

We present a case of a traumatic neuroma at the stump of the IMA that presented radiologic findings inconsistent with prior reports, making it difficult to differentiate this lesion from a malignancy pre-operatively. First, a contrast-enhanced CT scan exhibited a well-enhanced nodule (73 HU to 110 HU at portal-venous phase CT). This finding is not typical for a traumatic neuroma, usually described as hypoattenuating in the arterial, portal, and equilibrium phases of an enhanced

CT scan.¹³ Second, a FDG-PET scan revealed a hypermetabolic nodule in the left para-aortic IMA stump area (SUV 2.87), later found to be a false positive after surgical excision. Third, the lesion increased from 1.2 cm to 1.8 cm in diameter over one year. The growth rate during the year prior to neuroma resection (0.6 cm/year) raised strong suspicion of malignancy.

Only one case of a traumatic neuroma exhibiting imaging findings consistent with our case has been reported. A 70-year-old man presented with late-onset jaundice as a result of a polypoid mass in CBD.⁸ The patient underwent a cholecystectomy 25 years prior to presentation and a choledochojunostomy 12 years prior. A contrast-enhanced abdominal CT scan exhibited a polypoid mass in mid-CBD with contrast enhancement. Magnetic resonance cholangiography exhibited a polypoid filling defect in the mid-CBD, and the PET-CT scan showed a mild hypermetabolic lesion in the CBD (SUV 1.9). An extensive dissection called pylorus-preserving pancreaticoduodenectomy was performed under the impression of CBD cancer, but the lesion was finally confirmed as a CBD neuroma. Therefore, traumatic neuroma can show contrast enhancement on CT and mild hypermetabolism on PET-CT, even though it is not typical for traumatic neuroma.

With regard to its anatomic location, a traumatic neuroma at the IMA stump has not been reported in the literature. The lesion may involve the inferior mesenteric plexus (interconnected with the celiac plexus) surrounding the root of IMA that is resected during U-LAR performed in the rectal cancer patient of this case. The risk of biliary traumatic neuromas can be increased due to any injury to the surrounding nerves (mainly derived from the celiac plexus) or arteries or the bile ducts themselves during surgical manipulations, such as vascular ligation, thermocoagulation, and excessive exploration.⁸ Thus, the traumatic neuroma of our case likely resulted from injury to the inferior mesenteric plexus during the IMA resection or lymph node dissection for rectal cancer surgery.

In this patient, an infiltrative soft tissue lesion was seen at the IMA stump area two weeks after rectal cancer surgery (Fig. 1). This lesion showed typical imaging findings of post-operative granulation tissue that is commonly seen after lymph node dissection. Therefore, the lesion is not thought to be the initial finding of traumatic neuroma. A well-defined nodular mass developed seven months after surgery, possi-

bly the beginning of traumatic neuroma.

In conclusion, a traumatic neuroma occurred at the stump of the IMA. Traumatic neuromas are rarely observed at this site. The lesion exhibited atypical imaging features, such as a well-enhanced nodule, rapid interval growth in size and a mild increase of FDG uptake. Understanding this case will help enhance our knowledge of imaging features of traumatic neuromas.

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