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Case report

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Case report: An inflammatory pseudotumor of the caudate lobe of the liver caused by a foreign body



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ABSTRACT

This case report explores a rare foreign body-induced inflammatory pseudotumor in the caudate lobe of a 47-year-old male. The patient was admitted to the hospital due to epigastric pain and fever. Radiological examinations led to the diagnosis of a malignant tumor, and a resection of the caudate lobe lesion was performed. The excised tumor specimen revealed a fishbone-like foreign body. Immunohistochemistry suggested that this was an inflammatory pseudotumor rather than a malignant tumor. This prompted us to contemplate the origin of the foreign body and the mechanisms by which it led to the formation of the inflammatory pseudotumor.

1. Introduction

The caudate lobe of the liver, due to its small size, although the likelihood of tumors is low, benign or malignant tumors can still occur [1,2]. Among these, cavernous hemangioma stands out as the most common benign tumor [3], while hepatocellular carcinoma is a prevalent malignancy [4]. Inflammatory pseudotumors, characterized by well-defined tumor-like masses resulting from inflammatory tissue proliferation, are exceptionally rare in the caudate lobe [5]. Inflammatory pseudotumor is mainly classified into the fibrohistiocytic type, xanthogranulomatous type, spindle cell type, and plasma cell granuloma [6]. There is no gender difference in the occurrence of inflammatory pseudotumor [7], and its incidence rate is generally low [8]. Currently, it is generally diagnosed by observing symptoms and laboratory data indicating active inflammation, lacking specific imaging diagnostic methods [9–11]. This case report focuses on an intriguing instance of a foreign body-induced inflammatory pseudotumor in the caudate lobe of the liver, shedding light on its clinical presentation, diagnosis, and management.

2. Case presentation

A 47-year-old male patient was admitted to the hospital on February 3, 2015, due to epigastric pain with a fever for more than 10 days. On January 18, 2015, the patient developed persistent pain in the upper abdomen with no apparent cause, accompanied by chills and fever, with a maximum temperature of 38.9 °C. In the previous 10 days, the patient had been mentally healthy and slept well, with loss of appetite, dark-colored urine, reduced stool frequency, and weight loss of 5 kg. He had no history of hepatitis, and other systemic reviews did not reveal significant abnormalities. The patient then underwent a comprehensive examination, which revealed clear

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consciousness, positive percussion pain in the liver area and negative shifting dullness without pale jaundice in the sclera. The abdomen was flat and soft, and there was no tenderness, rebound tenderness, or palpable mass. No palpable liver or spleen under the ribs was found through physical examination. Routine blood tests were abnormal, with white blood cell counts of 12.82×10^9 /L (normal: $4-10 \times 10^{9}$ /L), neutrophil counts of 10.13×10^{9} /L (normal: $2-7 \times 10^{9}$ /L), and higher high-sensitivity C-reactive protein levels (117.29 mg/L, normal: 0.8~8mg/L). No significant abnormalities in liver or kidney function were found in any of the biochemical tests. The hepatitis B serologic test results included the following: antibodies against the hepatitis B core antigen, 0.004 COI (normal: 0~0.9COI); antibodies against the hepatitis B e-antigen, 0.072 COI (normal: 0~1COI); and antibodies against the hepatitis B surface antigen, 521.10 IU/L (normal: > 10IU/L). The alpha-fetoprotein (AFP), carbohydrate Antigen 199 (CA199), and carcinoembryonic antigen (CEA) levels were normal. An abdominal Computed Tomography (CT) scan without contrast revealed a vertical row of mixed-density soft tissue masses at the second and third hepatic portal measuring approximately $9.2 \times 6.4 \times 5.9$ cm, with CT attenuation values ranging from 17 to 45 HU (Fig. 1A). In contrast-enhanced CT, during the arterial phase, the central region showed significant enhancement, with CT attenuation values ranging from 78 to 98 HU. Patchy liquefactive necrosis and punctate calcifications (Fig. 1B, star) were observed inside, but the boundary was still clear. The arterial phase of the contrast-enhanced CT showed obvious abnormal blood distribution in the liver, with a circular, slightly hypodense lesion (Fig. 1B, arrow) observed in the left liver and the lower pole of the right liver, measuring approximately 15 mm, with CT attenuation values of 75 HU and clear borders. No significant abnormalities were observed in the remaining liver, biliary tract, pancreas, spleen, kidneys, or adrenal glands on CT. Multiple lymph nodes were observed in the retroperitoneum. CT scan results indicated a high possibility of a malignant tumor, small nodules in the liver with minimal enhancement, abnormal blood perfusion in the liver, and multiple small lymph nodes located behind the peritoneum. The patient presented with upper abdominal discomfort on two occasions, July 4, 2014, and January 7, 2015, respectively, with no other specific symptoms noted. Both times, the patient underwent gastroscopy. The first gastroscopy showed an esophageal ulcer (Fig. 2A), and the second showed inflammatory hyperplasia in the lower esophagus (Fig. 2B). At that time, the patient was treated with standard therapies such as acid suppression, and their symptoms improved before discharge. This admission, the preliminary diagnosis reveals hepatocellular carcinoma in the caudate lobe of the liver and an esophageal ulcer. Subsequently, the patient underwent further preoperative preparation and then exploratory laparotomy and resection of the caudate lobe lesion on February 7, 2015. After performing surgery to remove the caudate lobe of the liver in the patient, the symptoms of abdominal pain and fever disappeared. Postoperatively, the patient received routine anti-inflammatory and fluid replacement therapy, with successful wound healing and no observed complications or potential adverse events. The excised tumor specimen was opened, revealing the presence of a fish bone-like foreign body (Fig. 3A). The pathology shows that it is mainly composed of significant proliferation of histiocytes, foam cells, and lymphoplasmacytic cells. Small abscesses can be seen in the center, with extensive fibrosis and residual or proliferative bile ducts and liver tissue around the lesion (Fig. 3B). The initial suspicion was of a malignant tumor, pending further confirmation through immunohistochemistry. Subsequent immunohistochemistry showed positive results for CD68, CD163, Lymphocyte Common Antigen (LCA), Vimentin, Periodic Acid-Schiff (PAS), Vascular Endothelial Growth Factor (VEGF), 5-Fluorouracil (5-FU), Topoisomerase II (TOPO II), and Ki-67 (approximately 20 % positive). This confirms that the consistent proliferative cells seen in pathology are histiocytes, accompanied by the formation of small abscesses, consistent with inflammatory pseudotumor changes. Following a consultation between our hospital and Nanjing General Hospital of Nanjing Military Command, both institutions concluded that the observed changes were indicative of an inflammatory pseudotumor.



Fig. 1. Abdominal CT scan. A) Soft tissue masses without contrast at the hepatic portal, measuring $9.2 \times 6.4 \times 5.9$ cm, with CT values ranging from 17 to 45 HU. B) The contrast-enhanced scan shows enhanced masses (78–98 HU), with liquefactive necrosis, calcifications (star), and a 15 mm hypodense lesion (arrow, 75 HU).



Fig. 2. Gastroscopic examination. A) Revealed the presence of an esophageal ulcer. B) Demonstrated inflammatory hyperplasia in the lower esophagus.



Fig. 3. Excised tumor specimen and pathology. A) A fish bone-like foreign body. B) Proliferative lesions with consistent cellular morphology.

3. Discussion

Hepatic inflammatory pseudotumors are common sites for both primary and secondary liver cancer, but inflammatory pseudotumors rarely occur. An inflammatory pseudotumor is a well-defined tumor-like mass characterized by proliferative inflammation. It is an extremely rare benign lesion with an unknown etiology. It has been reported in various organs, with the most common site being the lung [12], but it can also occur in the liver, bladder, and other locations [13,14]. Therefore, hepatic inflammatory pseudotumors are extremely rare, and their etiology and pathogenesis have not been determined. The most common symptoms of hepatic inflammatory pseudotumors include abdominal pain, fever, and abdominal distension [15]. These tumors can regress spontaneously, and the prognosis is generally favorable [16]. However, due to the similarity of clinical presentation and radiological features to malignant tumors [17], it can be challenging to differentiate inflammatory pseudotumors from malignancies [5]. As a result, it can be challenging to differentiate between inflammatory pseudotumors and malignant tumors using imaging (CT and MRI). Therefore, histopathological examination is necessary for the accurate diagnosis of inflammatory pseudotumors [11,18].

In this particular patient, the inflammatory pseudotumor was caused by inflammation triggered by a fish bone (foreign body) penetrating the liver caudate lobe through the esophagus.

The preoperative CT findings initially suggested a malignant liver tumor with patchy liquefactive necrosis and punctate calcifications, and further evaluation through coronal reconstruction of the abdominal CT clearly revealed that the punctate calcification was actually identified as an intraoperatively discovered bone foreign body. This discrepancy in the preoperative CT interpretation can be attributed to the rarity of this type of case. The final diagnosis was confirmed to be inflammatory pseudotumor through pathological and immunohistochemical results.

This patient prompted us to contemplate the origin of the bone foreign body, specifically whether it was endogenous or exogenous. Endogenous factors that may cause inflammatory pseudotumors include: abnormal immune system, long-term chronic inflammation, autoimmune diseases [19–24]; exogenous factors may include: viral infections, liver trauma or surgery-induced inflammatory

reactions [25]. If the tumor was endogenous, it could be attributed to a teratoma; however, the patient's preoperative AFP and CA199 levels were normal, and the postoperative pathological diagnosis was an inflammatory pseudotumor, ruling out the possibility of an endogenous cause. If this was exogenous, the question arises as to how it occurred in the caudate lobe of the liver. Upon reviewing the patient's medical history, it was noted that the patient resided in a coastal area and had a long-term history of fish consumption. Furthermore, the patient underwent gastroscopy examinations in July 2014 and January 2015, which indicated gastric and esophageal ulcers. It is possible that the foreign body originated from the inadvertent ingestion of a fish bone, which penetrated the gastric wall and reached the caudate lobe of the liver. Subsequently, the caudate lobe may have formed an inflammatory encapsulation, giving rise to an inflammatory pseudotumor and causing the patient's upper abdominal pain and fever. This is a remarkable process. We hope that by reporting this case study, doctors encountering cases where there is suspicion of a tumor but normal AFP levels, making it difficult to diagnose, will consider the possibility of an inflammatory pseudotumor. They can then inquire about the patient's medical history to explore the presence of foreign bodies, and if necessary, conduct a histopathological examination to differentiate inflammatory pseudotumors.

4. Conclusion

This case illuminates the diagnostic intricacies surrounding hepatic inflammatory pseudotumors in the caudate lobe, emphasizing the need for nuanced evaluation. The unexpected discovery of a foreign body prompts reflection on rare etiologies, underscoring the challenges in distinguishing benign from malignant lesions. The patient's coastal residence and fish consumption history hint at a unique trigger for inflammatory pseudotumor formation. Clinically, this case underscores the importance of considering diverse factors in diagnosis. Its broader implications contribute to the evolving understanding of hepatic inflammatory pseudotumors, guiding clinicians to approach similar cases with comprehensive scrutiny and highlighting the significance of individualized diagnostic strategies in the realm of rare hepatic lesions.

Ethics statement

The written informed consent was obtained from the patient for the publication of this case report, any accompanying data and images.

Patient identity information

Patient-identifiable information has been entirely removed.

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Additional information

No additional information is available for this paper.

CRediT authorship contribution statement

Jianfa Zhong: Writing – review & editing, Writing – original draft, Investigation, Formal analysis, Data curation. **Zhaojie Su:** Writing – review & editing, Validation, Investigation, Data curation. **Hao Li:** Writing – review & editing, Writing – original draft, Validation, Supervision, Investigation, Formal analysis, Data curation. **Jingmiao Ma:** Writing – review & editing. **Dongbin Li:** Project administration, Investigation, Data curation.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

References

M. Shimada, T. Matsumata, T. Maeda, K. Yanaga, A. Taketomi, K. Sugimachi, Characteristics of hepatocellular carcinoma originating in the caudate lobe, Hepatology 19 (1994) 911–915.

- [2] J.O. Colonna, A. Shaked, H.A. Gelabert, R.W. Busuttil, Resection of the caudate lobe through "bloody gultch,", Surg. Gynecol. Obstet. 176 (1993) 401-402.
- [3] L. Jing, H. Liang, L. Caifeng, Y. Jianjun, X. Feng, W. Mengchao, Y. Yiqun, New recognition of the natural history and growth pattern of hepatic hemangioma in adults, Hepatol. Res. 46 (2016) 727–733, https://doi.org/10.1111/hepr.12610.
- [4] J.M. Llovet, R.K. Kelley, A. Villanueva, A.G. Singal, E. Pikarsky, S. Roayaie, R. Lencioni, K. Koike, J. Zucman-Rossi, R.S. Finn, Hepatocellular carcinoma, Nat. Rev. Dis. Prim. 7 (2021) 6, https://doi.org/10.1038/s41572-020-00240-3.
- [5] J.B. Koea, G.W. Broadhurst, M.S. Rodgers, J.L. McCall, Inflammatory pseudotumor of the liver: demographics, diagnosis, and the case for nonoperative management, J. Am. Coll. Surg. 196 (2003) 226–235, https://doi.org/10.1016/S1072-7515(02)01495-3.
- [6] Y. Zen, T. Fujii, Y. Sato, S. Masuda, Y. Nakanuma, Pathological classification of hepatic inflammatory pseudotumor with respect to IgG4-related disease, Mod. Pathol. 20 (2007) 884–894, https://doi.org/10.1038/modpathol.3800836.
- [7] J.H. Kim, J.H. Cho, M.S. Park, J.H. Chung, J.G. Lee, Y.S. Kim, S.K. Kim, S.K. Kim, S.K. Kim, B.W. Choi, K.O. Choe, J. Chang, Pulmonary inflammatory pseudotumor-a report of 28 cases, Korean J Intern Med 17 (2002) 252–258, https://doi.org/10.3904/kjim.2002.17.4.252.
- [8] G.H. Li, J.Q. Li, Y.Z. Lin, Inflammatory pseudotumor of the liver, J. Surg. Oncol. 42 (1989) 244–248, https://doi.org/10.1002/jso.2930420409.
- [9] R. Horiuchi, T. Uchida, T. Kojima, T. Shikata, Inflammatory pseudotumor of the liver. Clinicopathologic study and review of the literature, Cancer 65 (1990) 1583–1590, https://doi.org/10.1002/1097-0142(19900401)65:7<1583::aid-cncr2820650722>3.0.co;2-l.
- [10] J.I. Lopez, B. Eizaguirre, M. Nevado, J.C. Ruiz-Jaureguizuria, Inflammatory pseudotumor of the liver. Report of a case and literature review, APMIS 98 (1990) 1022–1026.
- [11] T. Sakai, K. Shiraki, N. Yamamoto, T. Kawakita, S. Ohmori, I. Itoh, T. Nakano, M. Yasuda, K. Yamakado, K. Takeda, S. Yagi, K. Yamagiwa, H. Yokoi, T. Noguchi, S. Uemoto, Diagnosis of inflammatory pseudotumor of the liver, Int. J. Mol. Med. 10 (2002) 281–285.
- [12] P.J. Goldsmith, A. Loganathan, M. Jacob, N. Ahmad, G.J. Toogood, J.P.A. Lodge, K.R. Prasad, Inflammatory pseudotumours of the liver: a spectrum of presentation and management options, Eur. J. Surg. Oncol. 35 (2009) 1295–1298, https://doi.org/10.1016/j.ejso.2009.04.003.
- [13] L. Tj, B. Ew, T. Pm, M. Rf, Inflammatory pseudotumour of the liver: antecedent causes and clinical experience, J. Hepatol. 19 (1993), https://doi.org/10.1016/ s0168-8278(05)80582-3.
- [14] S.C. Hsieh, C.H. Wu, W.P. Chan, Diagnosis of inflammatory pseudotumour of the urinary bladder aided by dynamic computed tomography, Acta Clin. Belg. 65 (2010) 360, https://doi.org/10.1179/acb.2010.078.
- [15] X. Yang, J. Zhu, E. Biskup, F. Cai, A. Li, Inflammatory pseudotumors of the liver: experience of 114 cases, Tumor Biol. 36 (2015) 5143–5148, https://doi.org/ 10.1007/s13277-015-3167-v.
- [16] C. Balabaud, P. Bioulac-Sage, Z.D. Goodman, H.R. Makhlouf, Inflammatory Pseudotumor of the Liver: A Rare but Distinct Tumor-like Lesion, (n.d.).
- [17] Y. Matsuo, M. Sato, T. Shibata, M. Morimoto, K. Tsuboi, T. Shamoto, T. Hirokawa, T. Sato, H. Takahashi, H. Takeyama, Inflammatory pseudotumor of the liver diagnosed as metastatic liver tumor in a patient with a gastrointestinal stromal tumor of the rectum: report of a case, World J. Surg. Oncol. 12 (2014) 140, https://doi.org/10.1186/1477-7819-12-140.
- [18] J.Y. Park, M.S. Choi, Y.-S. Lim, J.W. Park, S.U. Kim, Y.W. Min, G.-Y. Gwak, Y.-H. Paik, J.H. Lee, K.C. Koh, S.W. Paik, B.C. Yoo, Clinical features, image findings, and prognosis of inflammatory pseudotumor of the liver: a multicenter experience of 45 cases, Gut Liver 8 (2014) 58–63, https://doi.org/10.5009/ gnl.2014.8.1.58.
- [19] D.A. Arber, O.W. Kamel, M. van de Rijn, R.E. Davis, L.J. Medeiros, E.S. Jaffe, L.M. Weiss, Frequent presence of the Epstein-Barr virus in inflammatory pseudotumor, Hum. Pathol. 26 (1995) 1093–1098, https://doi.org/10.1016/0046-8177(95)90271-6.
- [20] C.M. Coffin, P.A. Humphrey, L.P. Dehner, Extrapulmonary inflammatory myofibroblastic tumor: a clinical and pathological survey, Semin. Diagn. Pathol. 15 (1998) 85–101.
- [21] G.M. Aru, C.R. Abramowsky, R.R. Ricketts, Inflammatory pseudotumor of the spleen in a young child, Pediatr. Surg. Int. 12 (1997) 299–301, https://doi.org/ 10.1007/BF01372155.
- [22] T.S. Neuhauser, G.A. Derringer, L.D. Thompson, J.C. Fanburg-Smith, N.S. Aguilera, J. Andriko, W.S. Chu, S.L. Abbondanzo, Splenic inflammatory myofibroblastic tumor (inflammatory pseudotumor): a clinicopathologic and immunophenotypic study of 12 cases, Arch. Pathol. Lab Med. 125 (2001) 379–385, https://doi.org/10.5858/2001-125-0379-SIMTIP.
- [23] T. Kamisawa, K. Takuma, N. Egawa, K. Tsuruta, T. Sasaki, Autoimmune pancreatitis and IgG4-related sclerosing disease, Nat. Rev. Gastroenterol. Hepatol. 7 (2010) 401–409, https://doi.org/10.1038/nrgastro.2010.81.
- [24] L. Jm, V. M, R. J, S. Jh, B. U, IgG4-related diseases of the digestive tract, Nature Reviews, Gastroenterol. Hepatol. 19 (2022), https://doi.org/10.1038/s41575-021-00529-y.
- [25] W.-G. Wang, Y. Zhang, L. Wang, Y. Chen, B.-L. Tian, Chronic pancreatic inflammatory granuloma caused by foreign body presenting as a pancreatic pseudotumor: a case report and literature review, Pancreatology 15 (2015) 573–575, https://doi.org/10.1016/j.pan.2015.05.474.