

Clinicopathological characteristics and prognostic factors for the recurrence of abdominal desmoid tumors: a retrospective study of 113 patients from two Chinese hospitals

Si-Cheng Zhou¹, Wei Pei¹, Zhen Sun², Zhi-Xiang Zhou¹, Xi-Shan Wang¹, Jian-Wei Liang¹, Qiang Feng¹

¹Department of Colorectal Surgery, National Cancer Center/National Clinical Research Center for Cancer/Cancer Hospital, Chinese Academy of Medical Sciences and Peking Union Medical College, Beijing 100021, China;

²Department of General Surgery, Beijing Hospital, Beijing 100730, China.

To the Editor: Desmoid tumors (DTs) show local aggressiveness to surrounding structures and exhibit a propensity to recur, leading to a high local recurrence rate and significant functional impairments and morbidity.^[1] It has been shown that DTs located in the abdominal wall and abdominal cavity are relatively minimally invasive and have a relatively low recurrence rate,^[2] which indicated abdominal desmoid tumors (ADTs) may have different biological make-ups and genes. This study was aimed to delineate the clinicopathological features and determine the prognostic factors for recurrence-free survival (RFS) in ADTs after macroscopic complete surgical resection.

From January 2000 to January 2019, a total of 343 patients who underwent surgical resection and were pathologically diagnosed with DTs from two different Chinese institutions were retrospectively reviewed. The inclusion criteria were as follows: (1) Patients with DTs of the abdominal wall or abdominal cavity; (2) Patients undergoing macroscopically complete surgical resection (R0 or R1); and (3) Patients who received surgical resection as the initial treatment. According to the above criteria, the remaining 113 patients were analyzed and formed the basis of the present study. The study protocol was approved by the Ethics Committee of the National Cancer Center (No. NCC 2017-YZ-026, October 17, 2017), and all patients signed an informed consent form before the study.

Clinical data and surgical outcomes were collected based on electronic records. The tumor site was categorized as intra-abdominal or abdominal wall. Surgical margin and tumor size were examined by two pathologists specialized in gastroenteric tumors, and the microscopic margin status was considered positive if the tumor was identified in the pathological specimen to be <0.5 cm from the edge of the

inkblot. According to the patient's general condition and the radiologist's experience, adjuvant radiation with a median overall dose of 55 Gy was recommended. The patients received a follow-up survey every 1 to 2 years via outpatient visits or telephone until recurrence, death, or December 31, 2019. Local recurrence was the main endpoint and was diagnosed by physical computed tomography scans or magnetic resonance imaging.

All data were analyzed using the Statistical Package for the Social Sciences (SPSS version 24.0, IBM Corp., Armonk, NY, USA). Quantitative data were expressed as the mean \pm standard deviation, and the two groups were compared with paired Student's *t* tests and Mann-Whitney *U* tests for independent values for normally and non-normally distributed values, respectively. Qualitative data and ordinal data are presented as the number of cases and percentages, and the groups were compared using χ^2 tests or Mann-Whitney *U* tests, as appropriate. The local RFS time was defined as the time interval between the date of pathological diagnosis and recurrence. RFS rates were analyzed by the Kaplan-Meier method and were compared between the subgroups with the log-rank test. In addition, the Cox proportional-hazards regression model was used to perform multivariate analysis to identify the independent prognostic factors. A *P* value of <0.05 was considered statistically significant.

All patients successfully underwent macroscopically complete surgical resection [Figs. 1A and 1B], and there were no deaths during the perioperative period. A total of 113 patients were included in this study. The median age at first diagnosis was 36 years (range 15–71 years), and the cohort was predominantly female (73.5%, *n* = 83).

Access this article online

Quick Response Code:



Website:
www.cmj.org

DOI:
10.1097/CM9.0000000000001333

Correspondence to: Jian-Wei Liang, Department of Colorectal Surgery, National Cancer Center/National Clinical Research Center for Cancer/Cancer Hospital, Chinese Academy of Medical Sciences and Peking Union Medical College, Beijing 100021, China
E-Mail: Liangjw1976@163.com

Copyright © 2020 The Chinese Medical Association, produced by Wolters Kluwer, Inc. under the CC-BY-NC-ND license. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal.

Chinese Medical Journal 2021;134(12)

Received: 07-09-2020 Edited by: Jing Ni

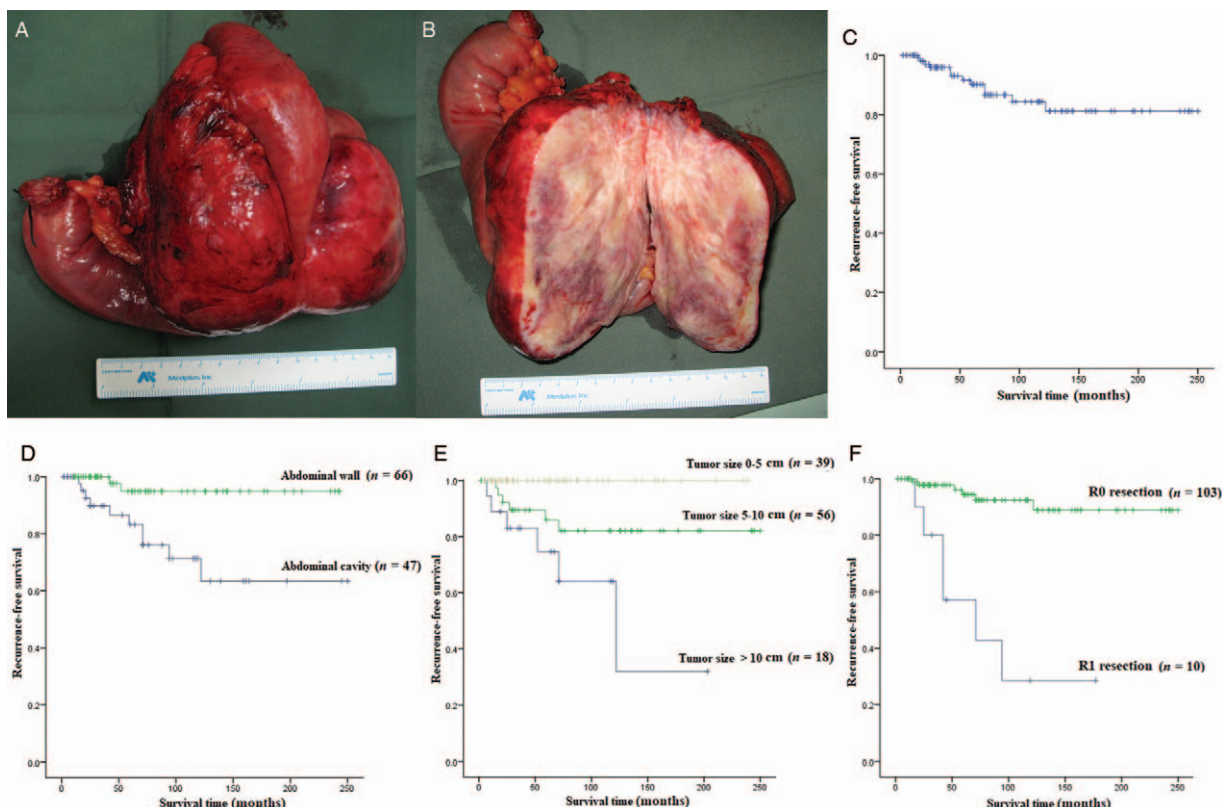


Figure 1: Desmoid tumors and RFS curves. (A) The tumor underwent macroscopic complete resection. (B) The tumor has a hard texture, white cut surface, and no necrosis. (C) The RFS rate of the entire cohort. (D) RFS according to the tumor site. (E) RFS according to the tumor size. (F) RFS according to the margin status. RFS: Recurrence-free survival.

According to the tumor sites, 113 patients were assigned to the abdominal wall group ($n = 66$) or the intra-abdominal cavity group ($n = 47$). The proportion of female patients in the abdominal wall group was significantly higher than that in the intra-abdominal group (93.9 vs. 44.7%, $P < 0.001$). In contrast, the proportion of patients aged 35 years or older was significantly higher in the intra-abdominal group than that in the abdominal wall group (91.5 vs. 63.6%, $P = 0.001$). There were more patients with a previous history of cesarean section in the abdominal wall group than those in the intra-abdominal group (34.8 vs. 6.4%, $P < 0.001$). In addition, patients with lesions in the abdominal wall were less likely to present with clinical symptoms than patients with intra-abdominal lesions (13.6 vs. 40.4%, $P = 0.001$). The number of patients in the intra-abdominal group with lesions >10 cm was significantly higher than that in the abdominal wall group (27.7 vs. 7.6%, $P = 0.011$), and the lesions in the intra-abdominal group were mostly adhered to important nerves or vasculature (42.6 vs. 13.6%, $P = 0.001$). In terms of the surgical outcomes, the abdominal wall group had a significantly shorter operation time (98.7 ± 50.0 vs. 194.5 ± 90.3 min, $P < 0.001$) and a lower amount of intraoperative blood loss than the intra-abdominal group (24.1 ± 41.1 ml vs. 136.0 ± 149.4 ml, $P < 0.001$) [Supplementary Tables 1, and 2, <http://links.lww.com/CM9/A443>].

The median follow-up interval was 61 months (range: 9–250 months). During the whole follow-up period, a total of 12 patients developed recurrence after surgery with or

without adjuvant radiotherapy: 2 cases of recurrence were found in patients presenting with intra-abdominal DTs, and 10 cases of recurrence were found in patients presenting with abdominal wall DTs. The estimated 5- and 10-year RFS rates for the entire cohort were 92.8% and 89.8%, respectively [Figure 1C]. It is worth noting that recurrence was still observed in one patient beyond 10 years. In addition, none of the 12 patients who were admitted with recurrent disease experienced recurrence during follow-up after surgical resection combined with or without radiotherapy.

According to the univariate analysis, tumor location, tumor size, and margin status significantly affected RFS ($P < 0.05$). A significantly higher rate of local recurrence was associated with abdominal wall DTs, a tumor size >10 cm, and an R1 margin status [Figs. 1D–1F]. According to the multivariate analysis, RFS was significantly affected by tumor location (Hazard ratio [HR]: 8.407; 95% confidence interval [CI]: 1.649–42.865; $P = 0.010$), tumor size (HR: 17.437; 95% CI: 3.648–83.346; $P < 0.001$), and margin status (HR: 8.045; 95% CI: 2.388–27.099; $P = 0.001$).

In the present study, we divided ADT patients into an intra-abdominal DT group and an abdominal wall DT group to compare the clinicopathological features and prognosis factors of the two groups. Consistent with previous literature,^[3] the present study revealed that compared to patients with intra-abdominal DTs, most patients with abdominal wall DTs were young women with a history of

cesarean section. Our study also found that the tumor size of patients with intra-abdominal DTs was significantly larger than that of patients with abdominal wall DTs, and most of these patients presented with corresponding clinical symptoms upon the first diagnosis. This may be due to the insidious nature of intra-abdominal DTs and the absence of obvious special clinical symptoms at the initial stage. As the tumor gradually increases and compresses the surrounding organs, it causes abdominal pain, ileus, hydronephrosis, and other clinical symptoms.

In terms of the prognostic factors targeted ADTs, our results showed that intra-abdominal DTs, large tumor size, and R0 resection were all independent predictors for RFS in DT patients. Previous studies have demonstrated that the long-term prognosis of patients with abdominal wall DTs is significantly better than that of patients with intra-abdominal DT.^[4,5] Wilkinson *et al*^[4] reported the prognosis of 50 patients with abdominal wall DTs who underwent surgical resection. Within a median follow-up period of 5 years, the local recurrence rate was only 8% (4/50). Our study found that only 3% (2/66) of patients with abdominal wall DTs had local recurrence during follow-up. However, the local recurrence rate of patients with intra-abdominal DTs was 23.1% (10/47), and the tumor size was >5 cm in all 10 patients with recurrence, of whom 4 patients had tumors larger than 10 cm. In addition, of the 10 patients with recurrence in the intra-abdominal DT group, 5 had positive margins after surgery. We believe that unlike abdominal wall DTs located on the body surface, due to the rarity of DTs and the absence of special clinical symptoms in the initial stage, lesions in the abdominal cavity are already large at the time of diagnosis and are closely related to the surrounding important nerves or vasculature. To preserve organ function as much as possible, it is difficult to achieve radical resection, which results in a significantly higher local recurrence rate.

In conclusion, compared with intra-abdominal DTs, abdominal wall DTs demonstrate different clinicopathological features and better prognosis. Moreover, R0 resection and tumor size <5 cm suggest a better prognosis after surgical resection in patients with abdominal DTs.

Under the premise of ensuring negative margins during the first surgical procedure, patients with abdominal wall DTs can obtain satisfactory prognoses through radical resection.

Funding

This work was supported by grants from the Beijing Hope Run Special Fund of Cancer Foundation of China (No. LC2017A19), the Capital's Funds for Health Improvement and Research (No. 2016-2-4022), and the Application of Clinical Features of Capital City of Science and Technology Commission China BEIJING Special Subject (No. Z151100004015120).

Conflicts of interest

None.

References

1. Kasper B, Baumgarten C, Garcia J, Bonvalot S, Haas R, Haller F, *et al*. An update on the management of sporadic desmoid-type fibromatosis: A European Consensus Initiative between Sarcoma Patients EuroNet (SPAEN) and European Organization for Research and Treatment of Cancer (EORTC)/Soft Tissue and Bone Sarcoma Group (STBSG). *Ann Oncol* 2017;28:2399–2408. doi: 10.1093/annonc/mdx323.
2. Seinen JM, Niebling MG, Bastiaannet E, Pras B, Hoekstra HJ. Four different treatment strategies in aggressive fibromatosis: a systematic review. *Clin Transl Radiat Oncol* 2018;12:1–7. doi: 10.1016/j.ctro.2018.03.001.
3. Martínez Trufero J, Pajares Bernad I, Torres Ramón I, Hernando Cubero J, Pazo Cid R. Desmoid-type fibromatosis: who, when, and how to treat. *Curr Treat Options Oncol* 2017;18:29. doi: 10.1007/s11864-017-0474-0.
4. Wilkinson MJ, Chan KE, Hayes AJ, Strauss DC. Surgical outcomes following resection for sporadic abdominal wall fibromatosis. *Ann Surg Oncol* 2014;2:2144–2149. doi: 10.1245/s10434-014-3618-5.
5. Couto Netto SD, Teixeira F, Menegozzo CAM, Leão-Filho HM, Albertini A, Ferreira FO, *et al*. Sporadic abdominal wall desmoid type fibromatosis: Treatment paradigm after thirty two years. *BMC Surg* 2018;18:37. doi: 10.1186/s12893-018-0367.

How to cite this article: Zhou SC, Pei W, Sun Z, Zhou ZX, Wang XS, Liang JW, Feng Q. Clinicopathological characteristics and prognostic factors for the recurrence of abdominal desmoid tumors: a retrospective study of 113 patients from two Chinese hospitals. *Chin Med J* 2021;134:1505–1507. doi: 10.1097/CM9.0000000000001333