

Submission of abstract for Scientific Program of 2015 Meeting of the International Society of Limb Salvage (ISOLS)

Title: Adamantinoma of the Long Bones\_ a Retrospective Study of 18 Cases

Author: Deng, Zhiping 1 MD dengzhiping2000@163.com

Coauthors: Liao, Feng 1 MD liaofeng@bjmu.edu.cn

Chan, Chung Ming 2 MD chancm@ortho.ufl.edu

Zhang, Qing 1 MD zhangqing6863@vip.sohu.com

Hao Lin 1 MD haolin851@163.com

Scarborough, Mark 2 MD scarbmt@ortho.ufl.edu

Niu, Xiaohui 1 MD niuxiaohui@263.net

Institution: 1 Department of Orthopaedic Oncology Surgery, Beijing Ji Shui Tan Hospital,  
Peking University, Beijing, People's Republic of China.

2: Division of Orthopaedic Oncology, Department of Orthopaedic Surgery and  
Rehabilitation, University of Florida, Gainesville, Florida USA

Level of Evidence: IV

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Abstract:

Background:

Adamantinoma of long bones is an extremely rare tumor, which is described by this name because of its close morphological resemblance to adamantinoma of the Jaws. The oncological aggressiveness of this tumor, including the local recurrence and distal metastasis, is not well reported. Limb salvage is currently the treatment of choice for most adamantinomas.

Questions/Purposes:

The purposes of this study were: (1) to evaluate the characteristics of adamantinoma of long bones; (2) to evaluate treatment and the oncological result of this tumor.

Patients and Methods:

A retrospective study was obtained to evaluate the clinical outcomes of the treatment of adamantinoma at long bones. Data was collected from the JST orthopaedic oncology database. Eighteen cases were enrolled from 1999 Dec to 2014 Mar. The median age was 23 years old (range 15-46). There was 9 male and 9 female. The tumor located at tibia in 14 cases, at both ipsilateral tibia and fibula in 3 cases and at fibula in 1 case. Eleven cases were original and seven cases were referred to JST hospital after the curettage in the outside hospital.

Results:

Eleven of fourteen tibia cases were segmented resected and reconstructed with allograft or recycle bone. Three of fourteen tibia cases were curetted, enhanced with cancellous allograft in two and cemented in one. In three multicentric cases, two were resected in both tibial and fibular lesions, one was curetted in tibia and resected in fibula. The fibular case was resected without reconstruction.

A wide surgical margin was achieved in 12 cases, a marginal margin in 2 cases and an intralesional margin in 4 cases. The median duration of follow-up was 40 months (range 12-128 months). There were 4 local recurrences. Three patients that had a local recurrence underwent amputation and one patient underwent intralesional curettage. The local recurrence rate was 22% (4/18), 3 in the inadequate group and 1 in the adequate group. The local recurrence rate was 8.3% (1/12) and 50% (3/6), adequate and inadequate group respectively,  $P=0.045$ . The four local recurrent cases had been curetted in outside hospital before referring to JST hospital. The local recurrence rate was 0% (0/11) and 57.1% (4/7), in original cases and previous surgical involved cases respectively,  $P=0.01$ . Three cases developed the lung metastasis. The metastasis rate was 16.6% (3/18). At the end of follow-up, 15 patients were alive without disease. 3 patients were alive with disease. Limb salvage was attempted in all patients. Three cases were amputated because of the local recurrence. The final rate of limb preservation was 83.3% (15/18).

#### Conclusions:

This series is a large single institution series of this rare tumor. Wide surgical margin is essential to adamantinoma of long bones. A wide margin was adequate and the marginal or intralesional margin was inadequate. This tumor should be treated at the orthopaedic oncology referring center rather than general orthopaedic service. As a low aggressive malignant tumor, the local recurrence and lung metastasis can occur. Long term follow-up is necessary to these patients.