Enchondroma of the spine: a report of eleven cases

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Research

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Abstract

Purpose To explore clinical characteristics, surgical strategies, and prognosis of spinal enchondroma.

Methods We retrospectively analyzed eleven cases with primary benign spinal enchondroma who were treated in our institution between January 2008 and December 2016.

Results Of the 11 patients included in our study, the average age was 43 years(range from 5–66 years). There were 7 men and 4 women. The lesions were mainly located in the lumbar and thoracic column with 5 cases and 4 cases respectively, and following by the cervical and sacral vertebrae with both 1 cases. Total tumor resection was used in 10 cases and curettage in one case. The average follow-up was 3.3 years(range from 2 to 6 years). Most of the cases had achieved satisfied therapeutic outcome with the symptoms disappeared. However, Case 9 developed into local recurrence and a second surgery was performed. Although the lesion have not recured any more after the second operation during the follow-up, the lower extremity paralysis had no improvement. Case 2 was sent to our hospital for the recurrence of the sacral tumor which was very closed to the common iliac artery.When the second operation was conducted, the artery was ruptured and the patient died from hemorrhagic shock.

Conclusion To establish the diagnosis of spinal enchondroma is not always a simple job for the absence of specific clinical characteristics. The optimal treatment is total resection. Although spinal enchondroma is benign, a few cases can develop into local recurrence. Sacral enchondroma should be more vigilant for its fatal complications.

Introduction

Chondroma is known as a type of benign bone tumor originated from cartilage[1]. According to the growth methods, it can be further divided into two types: periosteal chondromas and enchondromas.Periosteal chondromas are characterized by growing outside the surface of the bone cortex,while enchondroma was defined as lesion arising within the medullary cavity[2]. Enchondroma, accounting for 3%~10% of all bone tumors, is defined as the most common phalangeal tumors[3]. Spinal enchondroma is extremely rare, accounting for 2% of all enchondroma[4]. The treatment of choice of the spinal enchondroma is total recection[5,6]. Although the lesion is detemined as benign tumor, malignant transformation and local recurrence can be observed in a few cases[6,7]. Cancerous and recurrent lesion will invade spinal cord, nerve root and surrounding soft tissue, and even lead to chronic pain and paraplegia.

There is little published information concerning spinal enchondroma for its rare morbidity,and the limited information mainly stem from a few case reports[4,5,8,9]. The authors of these reports presented us a lot of valuable experience in diagnosis and treatment of spinal enchondroma. However, the sample size of their researches is so small that some bad prognosis can not be revealed.

So, it is significant to analysis and sum up clinical experience of spinal enchondroma based on a larger sample. To our best knowledge, this has been the largest sample size at present.

Materials And Methods

Inclusion/Exclusion criteria

We retrospectively collected the patients'data including general medical records, imaging and pathological data from our institutional databases and all patients were pathologically diagnosed as "enchondroma". Then we defined the inclusion and exclusion criteria to selecte the cases that we want to explore. The inclusions were defined as following:(1) The lesion was verified as "enchondroma" by preoperative imaging and postoperative histopathology; (2) The lesion was located in vertebral column; (3) The minimum following-up was at least 2 years. The exclusion criteria were: (1) The lesion was diagnosed as enchondroma by histopathology but was not consistent with preoperative imaging; (2) Patient with incompleted medical record. (3) Patient who lost follow-up or died from non-research illness.

Imaging And Biopsy

X-ray was taken routinely in all patients. CT and MRI were used to further definite the nature, size,and the degree of invasion of the lesion.In the situation that malignant tumors was suspected, the biopsy should be performed.

Surgical Methods

The approched were determined by the tumor locations. Classically, the lesions located in middle and posterior columns was removed by posterior approach, while the anterior approache were required when the tumor was growth arising from anterior colum. Antero-posterior approach adopted to the lumbosacral tumors where anterior reconstruction was inadequate to provide spinal stability. After excision of the lesion, compressed endorachis and nerve roots should be released. To maintain spinal stability, instrumentation with fusion was always necessary. Spinal pedicle screw and titanium cage with autogenous bone were used to fuse the vertebral body for anterior and posterior approache, respectively.

Follow Up Measures

Patients returned to the hospital for a check every three months after surgery in the first year, every six months for second year, and then annually for life. Clinical examination including visual analog scale (VAS) and American Spinal Injury Association (ASIA) was conducted to evaluate the pain and neurological function repectively. X xay was the routine imaging examination. When the recurrence was suspected, CT or MRI was further used.

Results

Demographic and Clinical Characteristics

366 patients diagnosed pathologically with enchondroma were found from our institution database between 2005 to 2016.Among them, 18 cases involved spine. According our Inclusion and Exclusion criteria, 4 cases was excluded for inadequate follow-up, 2 cases for incompleted medical record, and 1 case for loss to follow-up.Among the other 11 cases included in our study, there were 7 men and 4 women.All patients accepted their primary surgical resection in our institution except case 2, who was sent to our hospital for local recurrence after initial surgery in other institution. The diagnostic age was 5–66 years with a mean of 43. All stages of the spine can be involved but often occur in the lumbar and thoracic, with 5 cases and 4 cases respectively. Posterior column, middle column and anterior column were all likely to be invased with 4case, 2 cases and 2cases respectively. Posterior + middle column was both involved in 2cases and middle + anterior column in 1case. The pain was the most commen symptom. These symptoms can combine with numbness of lower limbs if the nerve root canals were involved. Patients would suffer from weakness when the spinal cord was compressed.

Imaging

In X-xay, radiolucent lesion can be observed in 3 cases, ambiguous vertebral pedicle in 2case, pathological fracture of vertebra in 3 cases, and no abnormal sign in 3cases. The CT scan showed that the lesion was characterized by expansive and osteolytic destruction complicated with scattered calcification. MR images revealed well-circumscribed mass with hypointense on T1-weighted images and hyperintense on T2-weighted images. A total of 5 patients underwent preoperative biopsy, and all petients were firmly diagnosed as chondroma. To the histological features, there were no significant difference between spinal enchondroma and conventional enchondroma.

Diagnosis

According to the clinical manifestation, imaging examination and biogsy, 8 patients were diagnosed as spinal enchondroma correctly.However, two patients were misdiagnosed as spinal giant cell tumor and one as spinal tuberculosis.

Treatment Effects And Follow-up

Posterior approach was conducted in six patients to removed the lesions, anterior approache in three,and antero-posterior approach in two.To keep spinal stability,all patients had their fusion with instrumentation. The average intraoperative bleeding volume was approximately 800 ml(range 350–1500 ml),and the mean operative time was 190 min(range from 120–250 min).

The follow-up ranged from 2 to 6 years with an average of 3.3 years.Nine patients improved after surgery with their VAS pain score dropping from 4.96 ± 1.77 (range 4–8) preoperatively to 1.06 ± 1.07 (range 0–6).For the six patients who had myelopathy complaint, the ASIA grade was increased significantly. However, the other two patients developed into bad prognosis. Case 9 developed into local recurrence and a second surgery was performed. Although the lesion have not recured any more after re-operation during the follow-up, the lower extremity paralysis had no improvement. Case 2 was sent to our hospital for the recurrence of the sacral tumor which was very closed to the common iliac artery. When the second operation was conducted,the artery was ruptured and the patient died from hemorrhagic shock.

Table 1
Demographics and operative data of 11 cases

No.	Sex /age	Location	Symptom	Approach	Method	Follow-up(year)	Final follow-up
1	F/63	L4	Pain numbness	Anterior	Resection	4	No recurrence
2	F/49	S1	Pain numbness weakness	Posterior	Resection	-	Death
3	M/5	L2/L3	Pain numbness weakness	Anterior	Resection	3	No recurrence
4	M/65	T6	Pain	Posterior	Resection	2	No recurrence
5	M/52	T1	Pain numbness weakness	Anterior	Resection	2	No recurrence
6	F/6	T8	Pain	Posterior	Resection	3	No recurrence
7	M/35	L4	Pain	Anterior/ posterior	Resection	6	No recurrence
8	M/32	C6	Pain numbness	Posterior	Resection	2	No recurrence
9	M/42	T7	Pain	Posterior	Curettage	5	Recurrence
10	F/66	L3	Pain numbness	Anterior/ posterior	Resection	4	No recurrence
11	M/53	L5	Pain	Posterior	Resection	2	No recurrence

Discussion

Epidemiology

Enchondroma is a common benign bone tumor which is quite specific to phalange[10]. Spinal enchondroma is rare, accounting for approximately 2% of the all enchondroma[4]. In present study, the morbidity of spinal enchondroma is 3.05%(11/366), which is slightly higher compared with the previous study.Additionally, all age stages are susceptible to this disease with no gender dominance.

Clinical Features

Mordard M et al revealed that thoracic vertebrae are more susceptible to be invased than other spinal regions[11]. This view was confirmed by Jing Guo et al in their case report[5].However, we found that lumbar and thoracic region are both susceptible to enchondroma. The tumor can invaded any column of the vertebra,including vertebral body, pedicles and spinous process. A case report involving three cervical chondromas by Gaetani P showed posterior elements were commonly affected. In present study, there were six lesions affecting posterior elements,accounting for 54.4% of all cases. Thus, enchondroma can attack the the total spine but mainly the lumbar and thoracic vertebra, and the posterior elements of the vertebra were commonly involved.

Local pain is the chief clinical manifestation. Some authors hold that there was a well demarcation between tumor and adjacent structures. Additionally, enchondroma was characterized by slow growth. So they suggested that neurological symptoms were rarely occurred in spinal enchondroma[4,12]. However,there were 6 patients complaining for their numbness and weakness, which accounts for the majority in our cases series. The cause of this divergence should be attributed to the different location of the lesions. The tumor reported by previous study mainly involved the vertebral body and spinous process, while our 6 lesions are all involved in pedicles, foramen intervertebrale and spinal cord. In fact, whether neurological symptoms occur or not is mainly due to the involved scope of tumors, but not to their integrity and growth rate.

X-ray is of little value in diagnosis of enchondroma. However, some indications including radiolucent lesion, ambiguous vertebral pedicle and pathological fracture of vertebra suggest that a toumor lesion may exist. In CT scan, the lesion is characterized by expansive and osteolytic destruction.In a few cases, scattered calcification can be observed. MR images revealed well-circumscribed mass with hypointense on T1-weighted images and hyperintense on T2-weighted images. Classically, the cortices is always intact and the soft-tissue is rarely invased in CT scan and MRI[4]. However, the continuity of the cortex was interrupted and the soft-tissue developed in our two cases of recurrence.

Diagnosis And Differential Diagnosis

It is difficult for physician to diagnose spinal enchondroma initially for the rarity of this lesion. Additionly, there are no characteristic changes in radiology. In case reports presented by Willis BK and S.H. Apurva,

the lesion was under suspicion as aneurysmal bone cyst or a giant cell tumor[4,8].The same misdiagnosis was established in our two cases. Fortunately, this error has not result in any undesirable outcomes for the consistent treatment of those two disease. One patient was misdiagnosed as spinal tuberculosis.Although the lesion of this patient was eventually removed and the right diagnose was confirmed via histopathology after operation, it is important to differentiate spinal enchondroma and tuberculosis to avoid pateint to received their unnecessary anti-tuberculosis drugs treatment. In fact, to distinguish those two diseases is also difficult when the enchondroma located in vertebra for the similar radiological appearances. So, the preoperative biopsy should be performed.

Treatment

This spinal lesion should be removed decidely in following situations:when the diagnose of enchondroma is suspected for its possibility of sarcomatous degeneration;when signs of nerve root or spinal cord compression were existed[11]. As suggested by other author, the purpose of surgery were to establish a accurate diagnosis, prevent malignant transformation, and preserve neurologic function[8,13]. The treatment method of enchondroma include excision and curettage. Curettage has been demonstrated effective to the enchondroma located in phalanges. However, the chioce of treatment of spinal enchondroma is excision since the curettage may increase the risk of local recuurence[7,14]. This conclusion could be further confirmed in our data. There was one case who was performed via curettage developed into recurrence ultimately, while the other 9 cases treated by excision gained the good prognosis.

An appropriate approach play a key role in achieving complete resection[15]. Jing Guo et al. used posterior approach and observed a good results in their case report[5]. In present study, anterior approach was performed in 3cases, posterior aproach in 6 cases and posterior combined with anterior approach in 2 cases. We believe that the approach was decided by the location of the lesion. Posterior approach was typically used for the lesions located in middle and posterior columns, while the anterior approache were required when the tumor was growth arising from anterior colum. Although the antero-posterior approach is difficult to perform,it is still adopted to the lumbosacral tumors where anterior reconstruction was inadequate to provide spinal stability.

Prognosis

After reviewing of the previous literature, we found that most of the cases achieved an ideal outcome with no pain, no neurological symptoms and improvement of numbness and motor weakness[5,8,9]. Similar outcome was also observed in our follow-up. So it is seems that this benign tumor is so mild that we can cure them easily.However, the following two cases have sounded alarms for us. Case 9 suffered from recurrence after 6 mouths postoperatively with a recurrence rate of 10%(1/10,the patient send from other hospital for her recurrence was excluded), which was consistent of the privious study[7]. The reason of

recurrence can be attribute to the inadequate resection. Although there was no recurrence after a second surgery, the lower extremity paralysis had no improvement for the invasion by the recurrent tumor.Case 2 was send to our hospital for the recurrence of the sacral enchondroma. Preoprative radiological examination showed that the tumor was very large and closed to the iliac vessels. We explained the surgical risk in detail,but the patient still required surgical treatment for the killing pain.After abundant preoperative plan, internal iliac artery ligation was performed via anterior approach and tumor removed via posterior approach. Unfortunately, the iliac artery was ruptured and the patient died from hemorrhagic shock.

Conclusion

To establish the diagnosis of spinal enchondroma is not always a simple job for the absence of specific clinical characteristics. The optimal treatment is total resection.Although spinal enchondroma is benign, a few cases can develop into local recurrence. Sacral enchondroma should be more vigilant for its fatal complications.

Declarations

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Availability of data and materials

All data used in the study are available at the request of editors and reviewers.

Authors' contributions

DHL and HJT conceived and designed the study and edited the final manuscript. DHL and YL collected the data, performed a literature review, and produced the draft manuscript. HJT, CSL, XBL, KL and XXC contributed to the pathological part of the review and helped with the writing of the manuscript. All authors read and approved the final manuscript

Competing interests

The authors declare that they have no competing interests.

Ethics approval and consent to participate

All patients consented to the data of their conditions being used in this study. The study was approved by the Ethics Committee of Minzu Affiliated Hospital of Guangxi Medical University and the First Affiliated Hospital of Guangxi Medical University

Consent for publication

All patients consented to the publishing of the results of this study.

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Figures

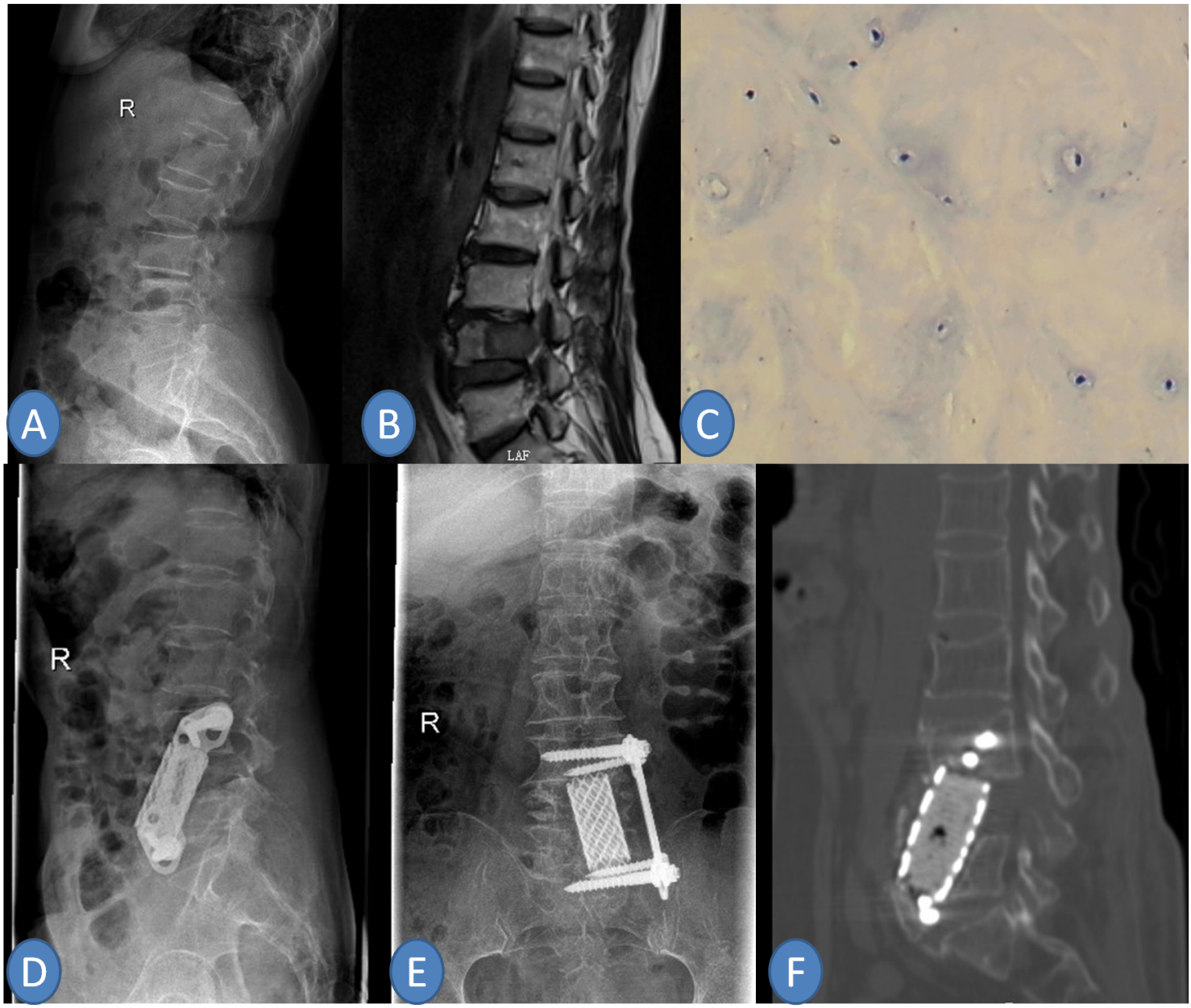

Figure 1

(Case 1). A 63-years-old woman complained her lumbago and numbness on lower limbs for one year. No significant abnormal can be obversed in plain film (A). Preoperative magnetic resonance imaging(B) revealed well-circumscribed extradural mass. Histological examination(C) showed mature hyaline cartilage with nests of benign-appearing chondrocytes (hematoxylin-eosin stain, original

magnification×100). No sign of relapse was detected in plain radiograph(D□E) and computed tomography scan(F) in six mouths following-up.

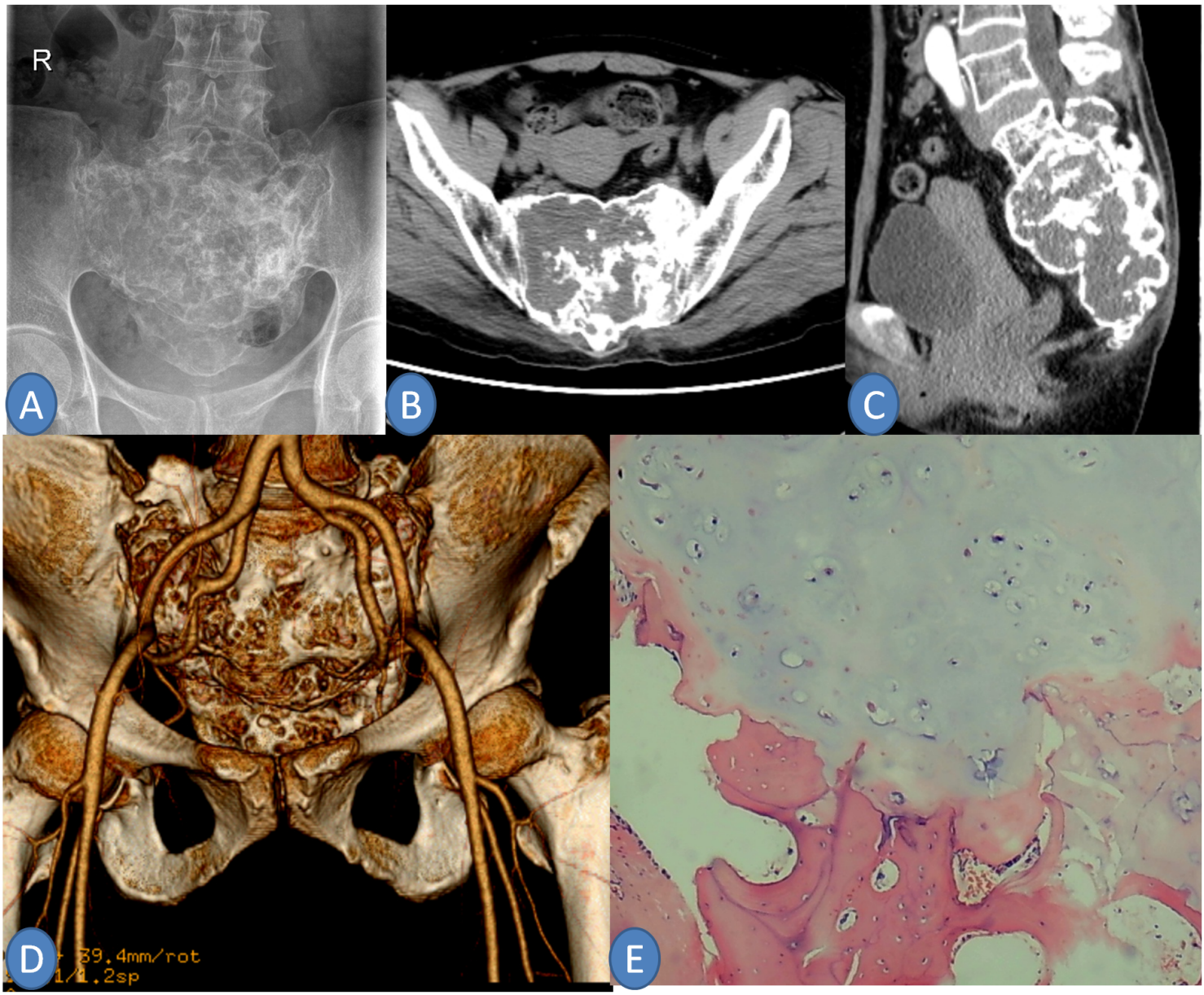

Figure 2

(case 9) A 49-years-old woman was sent to our institution for local recurrence and complained severe pain. Preoperative X-ray(A) and CT(B□C) demostrated the recurrence of tumor. Computed tomography angiography (D) revealed the tumor was very closed to iliac vessels. Internal iliac artery ligation was performed via anterior approach and tumor removed via posterior approach. Unfortunately, the iliac artery was ruptured and the patient died from hemorrhagic shock. Postoperative pathological examination(E) confirmed the recurrence of enchondroma (hematoxylin-eosin stain, original magnification×40).