Background
Charcot neuroarthropathy is a systemic disease that generates pathological changes in the musculoskeletal system, causing instability, dislocations, and deformities. Charcot neuroarthropathy of the knee, due to either diabetes mellitus or syringomyelia, is anecdotally reported with the epidemic of the diseases. However, idiopathic sensory peripheral neuropathy can inflict osteoarticular structures directly, inducing a dysfunctional Charcot neuroarthropathy. An early diagnosis and effective relief of the symptomatic deformity is essential for the treatment.

Case presentation
We report the case of a patient with idiopathic sensory peripheral neuropathy who presented with a swelling right knee, as well as distorted and painless gait disorder, diagnosed as Charcot neuroarthropathy of the knee. Partial weight bearing with a hinged knee brace was used to correct the abnormal alignment and gait posture, and bisphosphonates were prescribed to decrease pathological bone resorption. Although the alignment and Knee Society Score got a gradual deterioration, the combination of orthosis and pharmacy could alleviate the symptom to a certain extent.

Conclusion
The diagnosis of Charcot neuroarthropathy of the knee is rare that requiring early diagnosis. The presence of features, including painlessness, numbness, and deformed arthropathy following chronic-onset algesthesia loss should be taken carefully.

Keywords:
Charcot arthropathy, Idiopathic sensory peripheral neuropathy, Knee
axonal damage. Charcot-Marie-Tooth disease was excluded due to his normal motor function. There was no history of diabetes mellitus and neurosyphilis. He also had no family history of inherited diseases.

On physical examination, a valgus distortion of the knee was noted, with significantly restricted ranges of flexion (105°) and extension (10°). Inspired by a recent study [9], we used the system of Knee Society Score (KSS) to evaluate the function. The knee score was 46 and the function score was 40 on admission. There was a slight deep tenderness over the superolateral aspect of the patella and the lateral femoral condyle. Despite gross deformity and distinct instability, he complained of no pain during daily activities and sleep. Paracentesis yielded bloody effusions, both negative for biochemical examinations and microbial culture. Laboratory studies showed slight elevation in alkaline phosphatase (ALP) levels (147 U/L; reference: 34–104 U/L) at admission, reflecting abnormal absorption of bone metabolism. Infection was excluded due to the normal ranges of leukocyte count (4.7 × 10⁹/L; reference: 3.5 × 10⁹–9.5 × 10⁹/L), C-reactive protein (10.36 mg/L; reference: 0.00–10.00 mg/L), and erythrocyte sedimentation rate (12 mm/h; reference: 0–21 mm/h) along with negative microbiological and cytological examination of the repeated fluid aspiration. The coagulation and syphilis tests were normal without evidence of hemophilic arthritis and syphilis-induced Charcot arthropathy. Plain radiographs and CT scans showed apparent widening of the joint space and multiple abrasions of the lateral femoral condyle, with abnormal debris close to the posterior plateau (Figs. 2, 3). Stress radiography was not conducted due to the high risk of pathologic fractures and incompetency of pain complaint. Plain radiographs, captured under supine and neutral position, showed the femorotibial angle (FTA) was 165°. Magnetic resonance imaging revealed a wide impairment of the anterior cruciate ligament, lateral collateral ligament, and meniscus lateralis with massive joint effusions (Fig. 4). Idiopathic sensory peripheral neuropathy-induced Charcot neuroarthropathy was diagnosed in collaboration with neurologists.

Partial weight bearing with a hinged knee brace was advocated to correct the abnormal alignment and gait posture, followed by a gradual increase in flexion and extension over a 3-month period for clinical improvement. However, the significant disability determined the joint motion should be protected by the orthosis. Mild swelling of the knee was found in the lateral compartment, but there was no tenderness or redness. Additionally, he was administered physical therapy and rehabilitation along with bisphosphonates to decrease pathological bone resorption. ALP was dynamically monitored and found to be maintained between 108~134 U/L (reference: 50–136 U/L). Plain radiographs at the 12-month follow-up showed the valgus knee with the FTA of 159°, with a slight deterioration restricted in the lateral compartment (Fig. 5). The knee was reevaluated with KSS, showing the knee score dropped to 41 and the function score maintained 40. The range of motion was 10°~105°, reflecting the flexion and extension in the sagittal plane was not affected. Considering the potential risks of arthroplasty and inherent disadvantages of arthrodesis, protected weightbearing with the orthosis was recommended as the current treatment of choice [10].

Discussion and conclusions
Historically, the 2 dominant pathophysiological causes of Charcot neuroarthropathy included the neurotraumatic (abnormal sensory innervation, repeated microtrauma, and unregulated inflammatory cascade) and neurovascular theories (hypervascular region in subchondral bone, vascular shunting, and subsequent osteopenia) [1, 3]. Peripheral neuropathy caused by diabetes mellitus is the main cause of Charcot neuroarthropathy of the knee. While the idiopathic sensory peripheral neuropathy of the knee. While the idiopathic sensory peripheral neuropathy in our case can cause chronic-onset loss of algesia, inducing hypalgesia, numbness, and deformed arthropathy.
The primary degeneration of the dorsal root ganglion and trigeminal ganglion sensory neurons can cause gait ataxia, proprioceptive sensory loss, unnoticed damage, as well as repetitive microtrauma to the weight-bearing joint, contributing to Charcot neuroarthropathy [11, 12]. If not diagnosed and intervened promptly, it may result in severe complications, such as fractures and joint dislocations.

Charcot neuroarthropathy can occur secondary to syringomyelia, syphilis, diabetic neuropathy, alcoholic peripheral neuropathy, and idiopathic neuropathy. Diabetes is the most common etiology. Charcot neuroarthropathy is considered a major risk factor for below-the-knee amputation among the diabetics [1], while upper extremity can also be affected as a result of syringomyelia [5]. The diagnosis of Charcot neuroarthropathy is based primarily on thorough history and physical examination, including features, such as loss of protective sensation, presence of ulceration, and warm, swollen, erythematous foot and/or ankle. A combination of autonomic neuropathy, motor neuropathy, and sensory neuropathy results in osteopenia, joint instability, and increased loads on joints. The signs and symptoms of idiopathic sensory peripheral neuropathy-induced Charcot neuroarthropathy are consistent with non-length dependent nerve-fiber degeneration. The loss of sensory neurons in the dorsal root ganglia induce degeneration of short and long peripheral axons and central sensory projections in the posterior columns. Idiopathic sensory peripheral neuropathy induced Charcot neuroarthropathy, unlike others, is a rare disorder with unique characteristics, including deformities of fingertips and asymmetrical sensory disturbances.

Neuropathic arthropathy can be classified into four stages using standard radiology (prodromal, developmental,
Identification of Charcot neuroarthropathy of the knee in Stage 0 and I have important therapeutic implications. The immobilization and non-weight bearing of the knee may prevent progression of skeletal destruction and deformity. Antiresorptive therapy, especially with bisphosphonates, has been revealed to have a modest effect on Charcot neuroarthropathy [13], inhibiting excessive osteoclast activation and proinflammatory cytokine response [14]. Total knee arthroplasty (TKA) and arthrodesis should be employed only in the reconstruction or coalescence stages [15], similar to a salvage operative treatment. Although TKA is satisfactory for most patients [16], the complications due to fragile bone and ligamentous laxity still exists [17], including periprosthetic fracture, aseptic loosening, instability, and infection [18]. Knee arthrodesis is accompanied by major functional limitations, resulting in the restriction of knee motion. Bisphosphonates were prescribed due to the nature of progressive destruction of Charcot neuroarthropathy. Bisphosphonates were beneficial to control the level of ALP, which was monitored through the observation. However, with a brief literature review, we found most of previous studies did not detect any biomarkers associated with bone absorption [5, 6, 9, 19–22]. Although Charcot neuroarthropathy is not a bone metabolism disorder in nature, a dynamic monitor of serum biomarkers can provide substantial reference to modulate bone homeostasis and alleviate skeletal destruction. The activity of osteoclast and underlying inflammation should be evidenced by essential mechanistic investigations in the future [1].

To the best of our knowledge, this is the first case of Charcot neuroarthropathy due to idiopathic sensory peripheral neuropathy. The patient had an impressive history of multiple fingertips abscission and left leg amputation following painless ulceration. Nerve conduction studies confirmed the presence of idiopathic sensory peripheral neuropathy. The swelling of the right knee and painless gait disorder were the primary clinical features, consistent

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**Fig. 4** Magnetic resonance imaging reveals a wide disruption of anterior cruciate ligament, lateral collateral ligament and meniscus lateralis (a, b), with significantly increased effusions. The lateral femoral condyle is almost entirely destructed while the medial condyle is uninvolved (c)

**Fig. 5** Anteroposterior (a) and lateral (b) plain radiographs of the knee at the 12-month follow-up showed the lesion was restricted in the lateral compartment. The femorotibial angle has progressed to 159°
with the diagnosis of neuropathic arthropathy of the knee. Moreover, only the lateral compartment of the knee was significantly involved, with an obvious valgus deformity, detected promptly before total joint destruction. Our case had satisfactory clinical outcomes from periods of nonoperative management in Stage I.

In summary, the diagnosis of Charcot neuroarthropathy of the knee is rare and is considered as the accumulated effect of peripheral neuropathy. Although an early diagnosis of Charcot arthropathy cannot alter natural course of the disease, it is beneficial to relieve symptoms and prevent severe complications. The presence of features, including painlessness, numbness, and deformed arthropathy following chronic-onset algesthesia loss increases the likelihood of idiopathic sensory peripheral neuropathy-induced Charcot neuroarthropathy.

Abbreviations
ALP: Alkaline phosphatase; FTA: Femorotibial angle; KSS: Knee Society Score; TKA: Total knee arthroplasty

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Authors’ contributions
Q-HY and PH collected and analyzed the clinical data of the patient. Y-SG, and C-QZ undertook patient rehabilitation. All authors cared for the patient and approved the final manuscript.

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Availability of data and materials
All data generated or analyzed during this study are included in this published article.

Ethics approval and consent to participate
All procedures were part of the standard medical care, and the need for ethics approval and consent to participate was waived.

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and any accompanying images. The proof of consent to publish from study participants can be requested at any time.

Competing interests
The authors declare that they have no competing interests.

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