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Case Report

Prolonged Albendazole Therapy in Non-Surgical Multi-Organ Hydatid Disease: Report of Two Cases

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Abstract

Hydatid disease (cystic echinococcosis) caused by *Echinococcus granulosus* is endemic in Iran, most commonly affecting the liver and lungs. Multiorgan involvement with multiple cysts is uncommon, and surgery may not be feasible or acceptable for all patients. We report two Iranian patients with multiorgan hydatid disease treated exclusively with long-term albendazole. The first was a 54-year-old woman with multiple hepatic and pulmonary cysts who declined surgery. Hepatic cysts showed imaging features suggestive of CE2, while several pulmonary cysts were ruptured. The second was a 43-year-old man with multiple pulmonary cysts who was not considered a surgical candidate. His cysts showed heterogeneous radiologic appearances, including lesions compatible with CE1 and lesions with a detached membrane (water-lily sign), suggestive of CE3a. Both patients received albendazole 400 mg twice daily, with follow-up for 18 and 24 months, respectively. Both demonstrated gradual clinical and radiologic improvement without significant adverse effects. These cases add to the limited evidence that prolonged albendazole monotherapy may be an option for selected non-surgical patients with multiorgan hydatid disease, particularly when close clinical and radiologic monitoring is feasible.

Introduction

Hydatid disease, also known as cystic echinococcosis, is a zoonotic parasitic infection caused by the larval stage of *Echinococcus granulosus* and, less frequently, *E. multilocularis*. It remains a major public health issue in regions where livestock farming and close human–animal contact are common (1, 2). The global distribution is variable, with

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prevalence reaching 2%–6% or higher in some endemic rural areas (3). The parasite is widespread in the Mediterranean basin, the Russian Federation, Central Asia, Australia, North and East Africa, and parts of South America. Prevalence increases with age and is higher in rural populations, with a slightly greater incidence in women (4, 5).

The liver is the most commonly affected organ in hydatid disease, accounting for the majority of cases (6, 7). Extrahepatic involvement may result from hematogenous dissemination or, less commonly, direct extension, affecting organs such as the lungs, spleen, central nervous system, and bones (6, 8, 9). Awareness of these presentations is important in endemic regions, where hydatid disease should be considered in the differential diagnosis of cystic lesions. Many cases remain asymptomatic and are incidentally detected on imaging (10, 11). Diagnosis relies on imaging modalities, including ultrasound, CT, and MRI, supported by serologic testing. Management is individualized according to cyst stage, size, location, and other clinical factors and may include albendazole therapy, percutaneous interventions, surgery, or active surveillance (12).

Multiorgan hydatid disease is rare and poses therapeutic challenges (13, 14). Although surgical treatment may be feasible in selected patients, multiorgan involvement can increase operative complexity and morbidity (15). Albendazole is an established component of medical therapy for cystic echinococcosis and may be used in selected patients who are not suitable for surgery or who have multiple cysts or multiorgan disease. In a 13-year experience from Iran, 119 patients received albendazole therapy, and patients who completed three treatment phases showed favorable radiologic outcomes (16). However, evidence for albendazole monotherapy remains limited and heterogeneous, with treatment response varying according to cyst characteristics, location, and duration of therapy.

Given the limited evidence regarding prolonged albendazole monotherapy in extensive multiorgan disease, we report two rare cases from an endemic region of Iran that were treated exclusively with albendazole and followed with clinical and radiologic monitoring.

Case Presentation

Case 1

A 54-year-old Iranian woman with hypertension presented with headache, dizziness, neck pain, weakness, fatigue, poor appetite, productive cough, and shortness of breath. Vital signs were stable, and pulmonary examination revealed mild inspiratory crackles; abdominal examination was unremarkable. Initial laboratory tests, including blood cultures, SARS-CoV-2 and influenza PCR tests, and viral hepatitis and HIV markers, were negative. CBC showed eosinophilia of 38%.

Chest CT revealed bilateral pulmonary cystic lesions, the largest measuring 40 mm (Fig. 1), as well as incidental hepatic cysts, the largest measuring 77 mm (Fig. 2). Abdominal ultrasound confirmed multiple hypoechoic hepatic cysts. Based on the available imaging features, the hepatic cysts were considered compatible with CE2, whereas the pulmonary lesions included ruptured/complicated cysts for which a definite WHO-IWGE stage could not be assigned. Differential diagnoses included bronchogenic cysts, lung abscess, bacterial or fungal pneumonia, aspergilloma, tuberculoma, solid tumors, granulomas, and hydatid disease. Serologic testing was not performed. The diagnosis was supported by characteristic radiologic findings, marked eosinophilia (38%), and relevant epidemiological features, including a history of consuming vegetables irrigated with wastewater and residence in suburban Tehran, which supported the diagnosis of concomitant pulmonary and hepatic hydatid cysts.

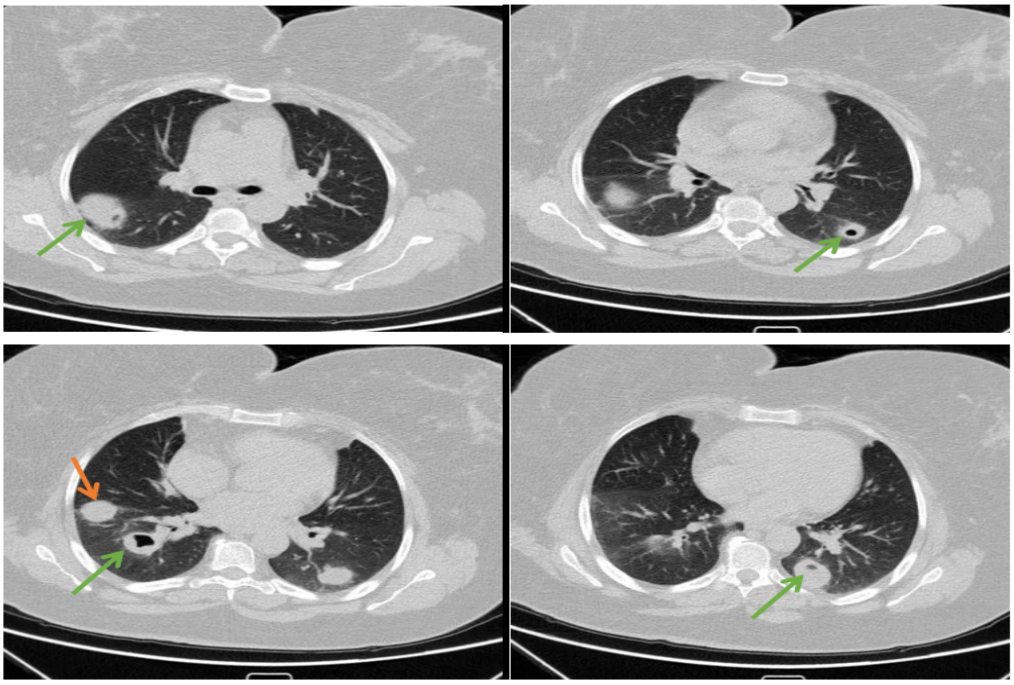


Fig. 1: Ruptured cysts characterized by air-fluid levels and a crescent sign (green arrows), alongside well-defined, homogenous, thin-walled cystic lesions (orange arrow)

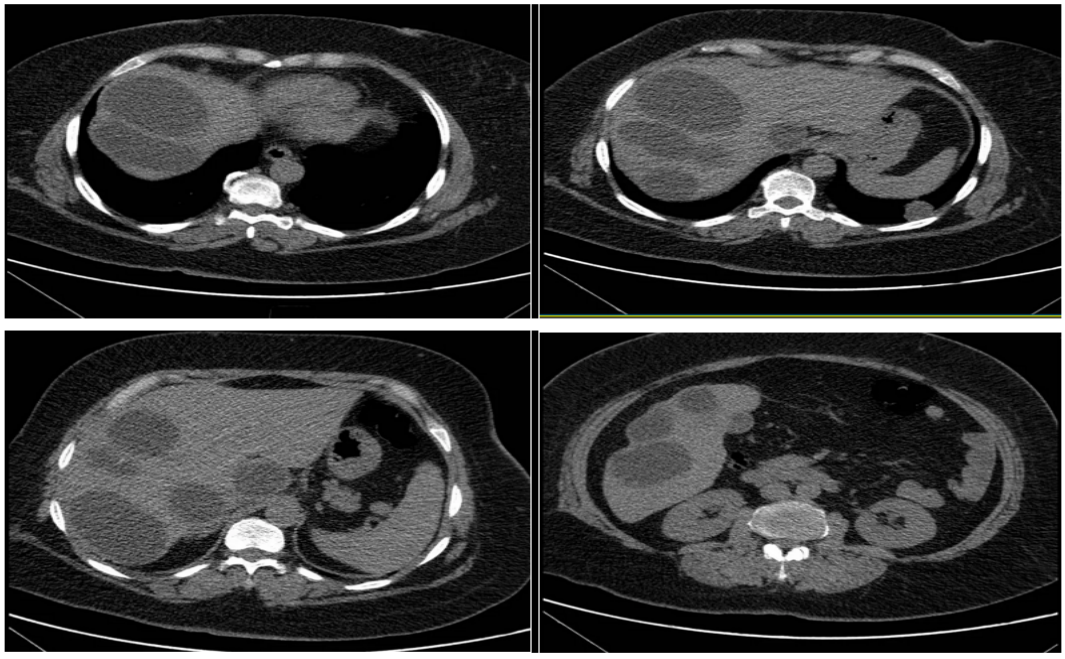


Fig. 2: Multiple thin-walled hepatic cysts without evidence of calcification, seen in the limited abdominal cuts of the patient's CT scan

Oral albendazole was initiated at a dose of 400 mg twice daily, and the patient was hospitalized for one week for monitoring. Brain CT and MRI showed no evidence of intracranial hydatid involvement. Her initial headache subsequently resolved, and no persistent neurologic symptoms were reported. She declined surgery and continued albendazole as an outpatient. Follow-up chest CT and liver ultrasonography demonstrated gradual radiologic improvement during treatment. The hepatic cysts gradually decreased in size and subsequently showed complete calcification on follow-up ultrasonography, consistent with inactive residual lesions. The pulmonary cysts subsequently showed imaging features consistent with rupture and involution. Hematologic and liver function tests remained within normal limits. Antiparasitic therapy was discontinued after substantial radiologic improvement and evidence of inactive residual lesions

Case 2

A 43-year-old previously healthy Iranian male who worked as a carpenter from suburban Tehran presented to the Infectious and Tropical Diseases Clinic at Shariati Hospital on Dec 19, 2023, with night sweats, productive cough, and approximately 20 kg unintentional weight loss over two months. He had previously been evaluated at a center affiliated with Imam Khomeini Hospital, where lung malignancy was suspected. Chest CT and bronchoscopy were performed, and bronchoscopic biopsy showed no evidence of malignancy. He was referred for further evaluation.

Given his stable clinical condition and an oxygen saturation of 97% on room air, outpatient management was considered appropriate. Due to persistent productive cough, chest imaging was repeated.

On the initial chest X-ray (Fig. 3) and CT scan obtained at Shariati Hospital, multiple lesions consistent with hydatid cysts were identified (Fig. 4). The pulmonary cysts showed heterogeneous radiologic appearances, with some lesions compatible with CE1 and others demonstrating a detached membrane (water-lily sign), suggestive of CE3a. Several cysts also showed features of rupture and were therefore described as ruptured/complicated cysts rather than assigned a definite CE stage. Serologic testing by enzyme immunoassay (EIA) was positive for hydatid IgG, while hydatid IgM was borderline. Liver ultrasonography revealed no hepatic involvement.



Fig. 3: Posteroanterior chest X-ray showing multiple nodules involving both lungs

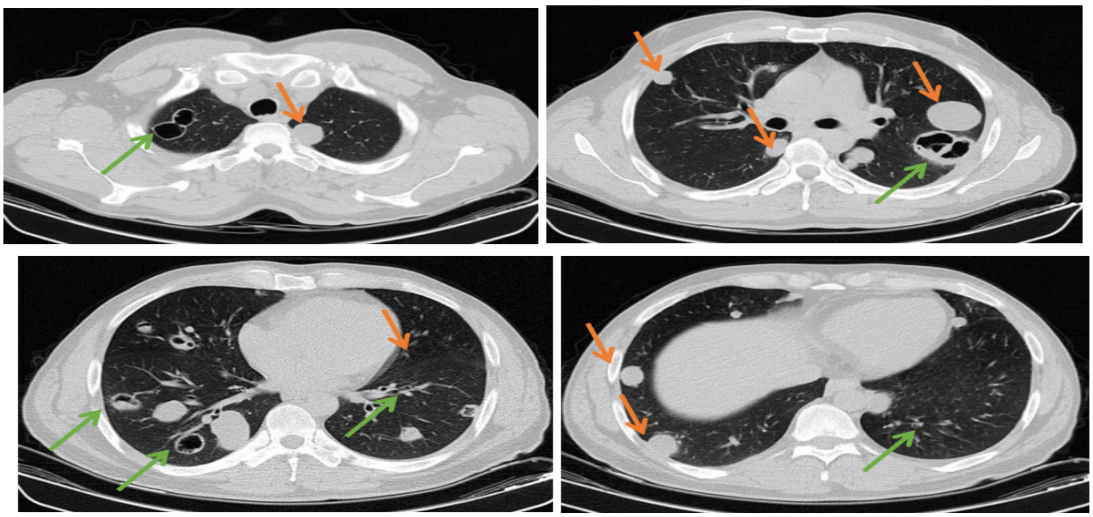


Fig. 4: Ruptured cysts characterized by air-fluid levels and a crescent sign (green arrows), alongside well-defined, homogenous, thin-walled cystic lesions (orange arrow)

Given the extensive and bilateral pulmonary involvement, surgical intervention was considered not feasible by the multidisciplinary team, and albendazole therapy was initiated. The patient was followed monthly, and treatment continued uninterrupted for two years. During follow-up, he reported occasional episodes of sudden coughing after strenuous activity, sometimes accompanied by salty taste and mild hemoptysis, which gradually resolved.

Follow-up CT scans showed reduction in the size and number of lung cysts (Fig. 5). The cysts progressively collapsed, and almost all of the lesions became markedly involuted, with only thin residual cyst walls remaining, indicating marked radiologic involution and inactivation. The patient gained approximately 19 kg and experienced marked clinical improvement, resuming normal daily and occupational activities.

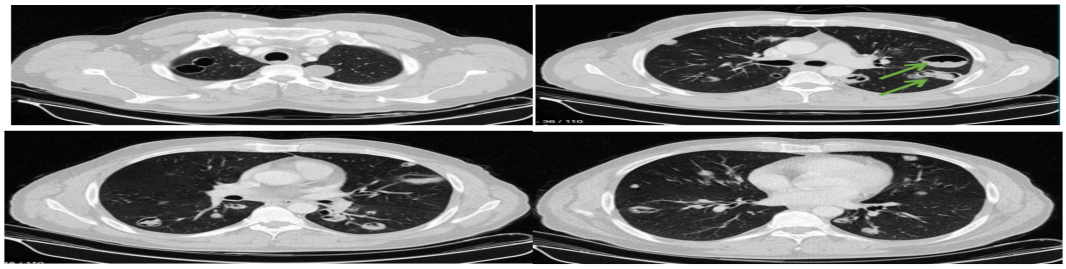


Fig. 5: Ruptured hydatid cyst "Water-Lily sign" (green arrows), reduction in size, thinning and collapse of the cyst walls, increased heterogeneity suggestive of inactivation of the pulmonary hydatid cysts

After two years of continuous therapy, given the significant clinical and radiologic improvement (Fig. 6, 7), albendazole treatment was discontinued



Fig. 6: Posteroanterior chest X-ray showing a reduction in the number and size of nodules

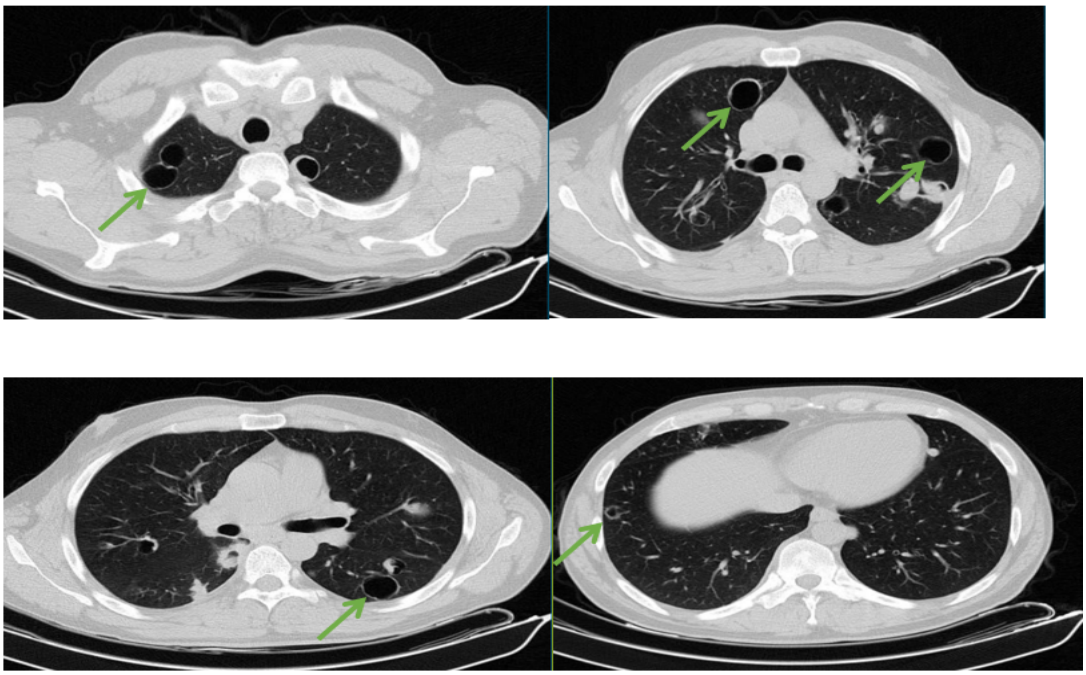


Fig. 7: Chest CT imaging revealed a thin-walled residual cavity after complete evacuation of the hydatid membrane (green arrows)

Treatment and Follow-up

Both patients received albendazole at a dose of 400 mg every 12 hours. Given the extensive disease and the decision to manage both patients without surgery, prolonged albendazole therapy was administered. Case 1 received treatment for 18 months and Case 2 for 24 months. the tablets were chewed and administered with a fat-rich meal to enhance absorption. Liver function tests were performed every three months and complete blood counts every six months throughout treatment. Treatment adherence was assessed through patient self-report during regular follow-up visits. No clinically significant abnormalities requiring treatment discontinuation were documented during follow-up.

Both patients demonstrated gradual clinical and radiologic improvement during prolonged albendazole therapy. Detailed changes in cyst number, size, WHO-IWGE classification, and radiologic status during follow-up are summarized in Table 1.

Table 1: Baseline and longitudinal radiologic characteristics of hydatid cysts during prolonged albendazole therapy

Patient	Organ	No. of cysts at baseline	No. of cysts at the end of treatment	Representative cysts	Baseline WHO-IWGE classification	Baseline size(mm)	Month 12 size (mm)	End of treatment	Final radiologic status
Case 1	Liver	8	4	Cyst 1	CE2-suggestive	48 ×77	45 ×64	46×56	Complete Calcification; CE5/inactive
				Cyst 2	CE2-suggestive	35 ×52	36 ×25	30×24	Complete Calcification;CE5/inactive
				Cyst 3	CE2-suggestive	31 ×40	33 ×25	33×21	Complete Calcification; CE5/inactive
Case 1	Lung	8	8	Largest cyst	Ruptured/complicated	44	-	-	Ruptured and completely emptied
Case 2	Lung	36	18	Cyst 1	Mixed CE1/CE3a-suggestive	42	-	-	Ruptured and completely emptied
				Cyst 2	Mixed CE1/CE3a-suggestive	36	-	-	Ruptured and completely emptied
				Cyst 3	Mixed CE1/CE3a-suggestive	34	-	-	Ruptured and completely emptied

After completion of albendazole therapy, both patients were followed for an additional six months. Follow-up was conducted through in-person visits and online consultations. No new symptoms suggestive of recurrence were reported during the post-treatment follow-up period. No additional imaging or post-treatment serologic testing was performed in either patient during this six-month period.

Ethics approval and consent to participate

Ethical approval was obtained from the Ethics Committee of Dr. Shariati Hospital (IR.TUMS.SHARIATI.REC.1404.134). The study adhered to the Declaration of Helsinki, and written informed consent was obtained from the both patients for publication of the case report and accompanying images.

Discussion

In selected patients with extensive or otherwise complex disease, medical therapy may be considered when surgery is not feasible or is declined. This is particularly relevant in endemic regions where surgical expertise might be limited or where patients are poor surgical candidates. In a 13-year experience from Iran involving 119 patients treated with albendazole, favorable radiologic outcomes were reported among those who completed the treatment protocol (16). These findings provide supportive, although limited, evidence for the use of albendazole in selected patients who cannot undergo surgery. In selected patients managed without surgery, albendazole therapy avoids the operative risks associated with surgical intervention, including cyst rupture, infection, and biliary complications. The optimal surgical strategy for complex cystic echinococcosis, particularly in patients with multiple cysts involving different organs, may be challenging and depends on cyst characteristics, location, and the feasibility of intervention (3). For patients with multiple cysts in multiple organs, a surgical approach would require either a high-risk single-stage multiorgan operation or multiple surgeries, both of associated with increased morbidity.

Medical therapy, on the other hand, can be administered on an outpatient basis and is cost-effective, making it especially suitable for resource-limited settings. However, there are limitations to this approach. Albendazole therapy requires long-term adherence and regular monitoring for side effects. Liver function abnormalities are relatively common, although they are rarely treatment-limiting, while occasional hematological changes affecting the white cells may be more serious (8). In addition, benzimidazoles are contraindicated in cysts at risk of rupture and in early pregnancy (14). Albendazole should be used cautiously in patients with chronic liver disease and is contraindicated in those with bone marrow suppression (4). Additionally, treatment response varies among cysts, and some lesions may require prolonged therapy or may show limited radiologic response. There is also a risk of recurrence after stopping treatment. Therefore, close follow-up with imaging (ultrasound or CT) and serology is essential.

In our two cases, prolonged albendazole monotherapy was associated with favorable radiologic outcomes despite the extensive multiorgan involvement. In Case 1, the hepatic cysts became completely calcified, while the pulmonary cysts showed imaging features of rupture and involution. In Case 2, the pulmonary cysts showed progressive collapse and marked radiologic involution during follow-up. These findings are consistent with the potential benefit of prolonged medical therapy in carefully selected non-surgical patients, although the response may vary according to cyst stage, location, and individual cyst characteristics.

These two cases suggest that albendazole monotherapy may represent a potential therapeutic option for the management of complex multiorgan hydatid disease, particularly in patients who are not suitable candidates for surgery. Surgical or percutaneous intervention remains appropriate for selected cysts according to their stage, size, location, and risk profile, whereas medical therapy may be considered in selected patients in whom intervention is not feasible or is declined (15). Further studies with larger cohorts are required to determine the optimal duration of therapy and to identify predictors of response to albendazole.

Conclusion

These cases suggest that prolonged albendazole monotherapy may be associated with radiologic improvement in selected patients with extensive or complex hydatid disease when surgery is not feasible or is declined by the patient. Radiologic improvement occurred gradually, including calcification of the

hepatic cysts and involution of the pulmonary cysts, without clinically significant adverse effects. Prolonged medical therapy may be considered in carefully selected non-surgical patients with favorable cyst characteristics and under close clinical and radiologic monitoring. Further large-scale studies are needed to determine the optimal treatment duration, predictors of response, and long-term outcomes.

Limitations

This study has several limitations. First, it is a case series with only two patients, which limits the generalizability of the findings. Second, the follow-up period, although sufficient to demonstrate radiologic improvement, may not capture long-term recurrence or late complications. Third, no control group was included, preventing direct comparison between albendazole monotherapy and surgical or combined treatment approaches. Finally, variations in cyst characteristics, patient comorbidities, and adherence to therapy may have influenced outcomes, and these factors were not fully standardized. Therefore, larger prospective studies are needed to confirm the efficacy, safety, and optimal duration of albendazole monotherapy in multiorgan hydatid disease.

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Conflict of interests

The authors declare no competing interests.

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