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Original Article

Orthopaedics Section

Management of Unicameral Bone Cyst with Pathological Fracture of Proximal Humerus with Curettage, Fibular Grafting and Plate Osteosynthesis: A Retrospective Study

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ABSTRACT

Introduction: Unicameral Bone Cysts (UBC) are benign fluid filled lesions at metaphyseal regions of long bones of immature skeleton. Several treatment options exist but none have proven to be the best. Curettage helps to remove the cyst membrane responsible for lysis of bone and fibular grafting with plate osteosynthesis which gives adequate stability to the fracture and promotes healing of the UBC of proximal humerus with pathological fracture.

Aim: To assess functional outcome, pain relief, healing of the cyst and recurrence of lesion with curettage and fibular bone grafting with proximal humerus plating in UBC of proximal humerus with pathological fracture.

Materials and Methods: This was a retrospective study done using the medical records of 10 patients with UBC of proximal humerus with pathological fracture treated with curettage and fibular grafting and plate osteosynthesis at Nizams Institute of Medical Sciences, Hyderabad, Telangana, India, between January 2018 to December 2021. Patients were studied for functional outcome by Musculoskeletal Tumour Society (MSTS) score before surgery and one year after surgery. Healing of the cysts was evaluated by using a modified classification system by Chang et al., and pain relief was evaluated with pre and postoperative Visual Analogue Scale (VAS). Paired t-tests were used to compare the MSTS scores and VAS scores.

Results: Mean age of the study group was 15.6±2.3 years with range of 11-19 years. The MSTS score increased significantly after surgery from 11.6±2.06 to 27.3±1.6, p<0.0001. Cysts had healed completely in seven cases, partially in three cases and recurrence was seen in three cases at the end of one year. Mean VAS scores decreased significantly from 7.60±0.97 to 1.40±0.97 after the surgery. Among three cases with recurrence of UBC, two cases were diagnosed at eight months and one case at 12 months after surgery.

Conclusion: Treatment of UBC of proximal humerus with pathological fracture by curettage, fibular grafting and plate osteosynthesis gives good functional outcome and cyst healing rate. It is a safe and effective procedure without any complications.

Keywords: Benign bone lesions, Musculoskeletal tumor society score, Recurrence, Simple bone cyst

INTRODUCTION

The UBC or simple/solitary bone cysts are benign fluid filled cavities of bone that enlarge over time, resulting in thinning of the bone and pathological fractures. These are the most common benign lytic lesions in children and are assumed to occur due to the disturbance in local blood flow [1]. These cysts are commonly reported in the metaphyseal areas of long bones with open physis and most often in proximal humerus and proximal femur [2].

Incidentally detected, asymptomatic UBC may spontaneously resolve with skeletal maturity [3]. The UBC do present with pathological fractures due to cortical destabilisation. Once a pathological fracture occurs, healing of cysts is impaired and immobilisation, observation often leads to recurrent fractures in 62-82% of cases [4-6]. Therefore, some form of surgical treatment is essential for the cysts with pathological fractures to heal.

Pathological fractures due to UBC are more common in humerus than in femur [7]. This may be due to the non weight bearing nature of humerus bone and therefore significant pain is not felt by patients until the fracture. Thus, proximal humeral UBC's enlarge and weaken the cortex until they end up in a pathological fracture [8]. Various surgical options like percutaneous decompression with cannulated screws or pins, intralesional steroid injection or injection of bone marrow, curettage with or without bone grafting, intramedullary nailing or combination of curettage and bone graft substitutes were described [9-14]. Each of these treatment methods has variable success rates. Kokavec M et al., reported that only 3 out of 19 patients operated with curettage and bone grafting had recurrence, yielding an effective rate of 84.2% [15]. Traub F et al., reported 100% cure rate with curettage and bone grafting in their study with five years follow-up [16]. In a recent review by Noordin S et al., it is reported that curettage and bone grafting in UBCs has healing rates as low as 25-36% [17]. There is a controversy in the existing literature regarding healing rates and rate of recurrence following curettage and bone grafting for UBC. Most of the studies published were on curettage and bone grafting alone or curettage and bone grafting with intramedullary nails [18,19].

Present study highlights the importance of curettage and fibular bone grafting with proximal humerus plating in the management of displaced pathological fractures of proximal humerus owing to UBC in terms of functional outcome, healing of cyst, pain relief and recurrence of lesion.

MATERIALS AND METHODS

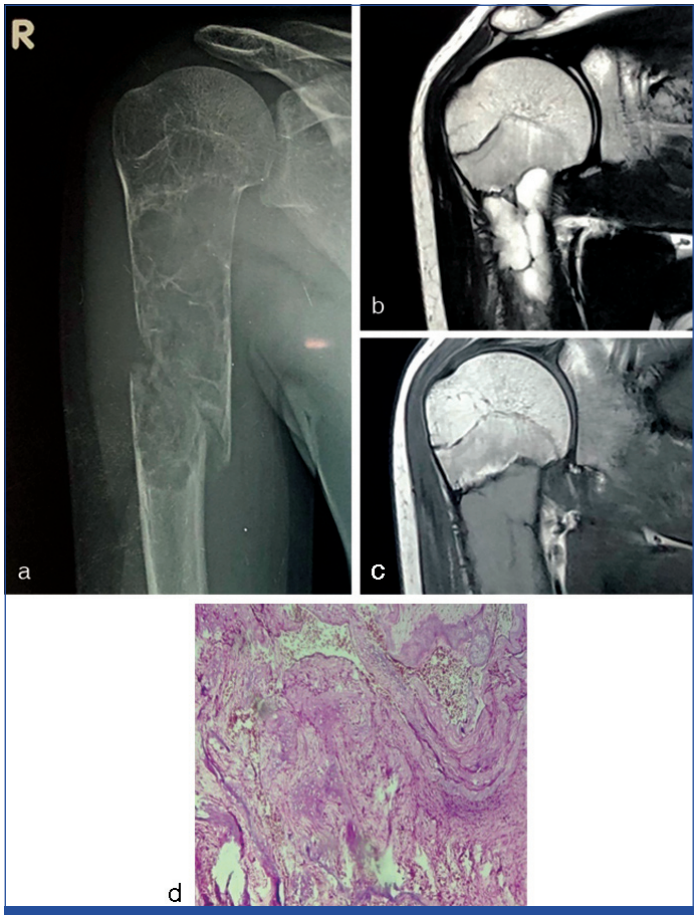
This was a retrospective study done by using medical records of 10 patients with pathological fracture of proximal humerus with UBC treated with curettage, fibular grafting and plate osteosynthesis at Nizams Institute of Medical Sciences, Hyderabad, Telangana, India, between January 2018 and December 2021. The analysis of the data was done in March 2022.

Inclusion criteria: Patients aged between 10-20 years gave consent, radiologically and histopathologically proven UBC of proximal humerus with displaced pathological fracture.

Exclusion criteria: Asymptomatic UBC of proximal humerus, UBC of proximal humerus without pathological fracture, UBC involving other than proximal humerus, recurrent UBC of proximal humerus.

Preoperative Assessment

A detailed history and clinical examination was carried out at the initial visit to assess signs and symptoms and anteroposterior and lateral X-ray views, Computed Tomography (CT) or Magnetic Resonance Imaging (MRI) scans [Table/Fig-1a-c] of the involved shoulder were recorded. Patients were then planned for core cut biopsy and once histopathological diagnosis [Table/Fig-1d] was confirmed patients were taken up for definitive surgery.



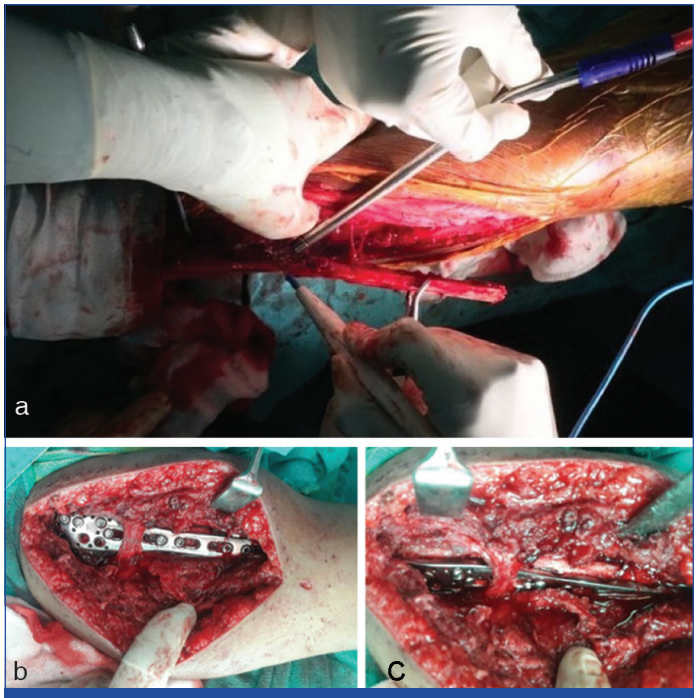
[Table/Fig-1]: a) Plain radiograph of a 18-years-old male showing unicameral bone cyst of proximal humerus; b,c) MRI image of the unicameral bone cyst of proximal homers; d) Histopathology showing features of unicameral bone cyst.

Surgical Technique

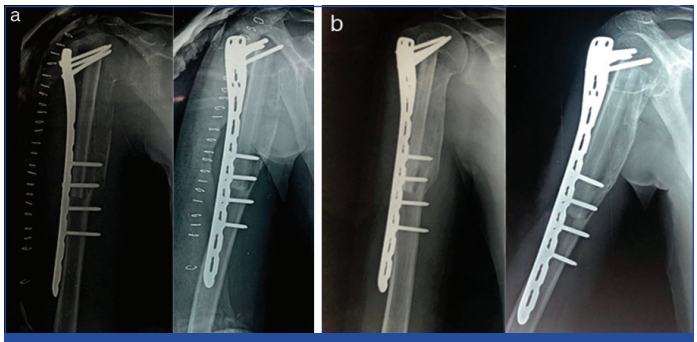
All the cases were operated by a senior orthopaedic surgeon under general anaesthesia. Proximal humerus was exposed through anterolateral approach and an adequate sized window was made over the proximal humerus. All the septae were opened and cyst was made into a single cavity and was thoroughly curetted. Curetted bone and membrane of the cyst cavity was sent for histopathological examination. Length of the cavity was measured after curetting the cyst and approximate sized fibular strut graft was harvested from ipsilateral leg [Table/Fig-2a] and was inserted into the distal medullary cavity. Fracture was reduced and a proximal humerus locking plate was applied [Table/Fig-2b,c]. Wound was closed in layers and a shoulder arm immobiliser was applied for four weeks. After four weeks, pendulum exercises were started and active shoulder abduction exercises were started after six weeks.

Postoperative Assessment

Patients were followed regularly for one month interval for first six months and then every three months until last follow-up [Table/Fig-3a,3b]. Patients were evaluated clinically and radiologically at every follow-up for functional outcome [Table/Fig-4], healing of cyst, pain and radiological signs of recurrence.



[Table/Fig-2]: a) Intraoperative picture showing the harvesting of a fibular graft; b,c) Intraoperative picture showing fibular grafting and plating after curettage.



[Table/Fig-3]: a) Postoperative radiograph of proximal humerus showing in situ fibular graft and surface plate; b) 12 months postoperative radiograph showing healed cyst.

[Table/Fig-4]: Shoulder abduction at 12 months after surgery.

The MSTS score was recorded at each follow-up and compared to preoperative score. The MSTS functional score measures outcomes in six categories including pain, function, and emotional acceptance for all patients. For upper extremity, hand positioning, manual dexterity and lifting ability were recorded [20]. Each parameter was scored 0 to 5 and combined for a possible total

Score	Pain	Function	Emotional	Hand positioning	Manual dexterity	Lifting ability
5	No pain	No restriction	Enthused	Unlimited	Unlimited	Normal load
4	Intermediate	Intermediate	Intermediate	Intermediate	Intermediate	Intermediate
3	Modest/Non disabling	Recreational restriction	Satisfied	Not above shoulder or no pronation/Supination	Loss of fine movements	Limited
2	Intermediate	Intermediate	Intermediate	Intermediate	Intermediate	Intermediate
1	Moderate/Disabling	Partial restriction	Accepts	Not above waist	Cannot pinch	Helping only
0	Severe disabling	Total restriction	Dislikes	None	Cannot grasp	Cannot help

[Table/Fig-5]: MSTS score for upper extremity [20].

score of 30. Intermediate values of 2 or 4 were assigned based on the examiner's judgment when achievement or performance falls between the specified values [Table/Fig-5]. For lifting ability, limited term indicates limitations in independent lifting. Helping term means that the patient cannot lift upper limb independently but useful in assisting the contra lateral upper limb in doing activities.

Healing of the cyst was evaluated by using a modification of a classification system first described by Neer CS et al., [21] and modified later on by Chang CH et al., [22]. According to this classification, operated UBC were classified into four types depending on healing response, 1) healed cyst- cyst filled by formation of new bone with or without small static, radiolucent areas less than 1 cm in size, 2) healing cyst- a static defect, radiolucent areas less than 50% of the diameter of the bone with enough cortical thickness to prevent fracture, 3) persistent cyst- Radiolucent areas greater than 50% of bone diameter with a thin cortical rim and no increase in cyst size, 4) recurrent cyst- cyst reappeared in a previously obliterated area or a residual radiolucent area has increased in size.

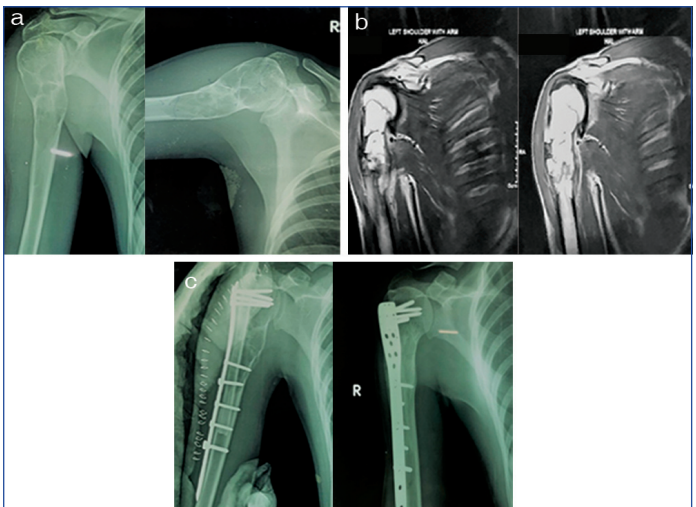
Pain relief was evaluated by using VAS that grades pain from 0-10 where score 0 is no pain and score 10 is worst pain [23]. The VAS score was recorded preoperatively and was compared to postoperative VAS score recorded at first follow-up visit.

STATISTICAL ANALYSIS

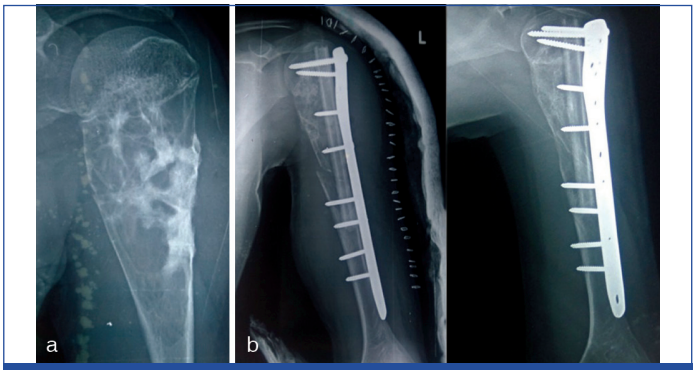
Statistical analysis was done with Statistical Package for the Social Sciences (SPSS) statistical software (version 22.0). Paired t-tests were used to compare the MSTS scores and VAS scores. The statistically significant difference level was set at $p<0.05$.

RESULTS

Mean age of the study group was 15.6±2.3 years with range of 11-19 years. Out of the 10 patients, six were males and four were females. All the patients were followed for at least 12 months with a mean follow-up period of 22.5±8.0 months (range:12-37 months) [Table/Fig-6a-c] and [Table/Fig-7a,b]. All the cases are summarised



[Table/Fig-6]: a) Radiographs of a 19-years-old female with unicameral bone cyst of proximal homers with pathological fracture; b) MRI image of the unicameral bone cyst of proximal humerus; c) Postoperative (left) and 12 months follow-up radiographs showing healed cyst.



[Table/Fig-7]: a) Radiograph of a 15-years-old male with unicameral bone cyst of proximal humerus; b) Postoperative and three months follow-up radiograph shows healing cyst.

in [Table/Fig-8]. The MSTS score has increased significantly from 11.6±2.06 to 27.3±1.6 one year after surgery ($p<0.0001$) [Table/Fig-9]. As per the described Neer CS et al., [21] and Chang CH et al., [22] classification, five patients had healed cysts, two patients had healing cysts and recurrent cysts were seen in three patients at the end of one year. Mean VAS scores before surgery was 7.60±0.97 and had decreased significantly after the surgery to 1.40±0.97 ($p<0.0001$). Among three cases with recurrence of UBC, two cases were diagnosed as early as eight months after the surgery and one case was diagnosed at 12 months [Table/Fig-10a,b]. There were no intraoperative complications in the studied subjects. One patient had superficial surgical site infection which was treated successfully with oral antibiotics and regular wound dressings.

DISCUSSION

The UBC in proximal humerus can be managed with multiple methods but there is no common consensus about the best treatment method. In 5-10% cases, spontaneous resolution of the cyst occurs [24]. But majority of the cases require some form of surgical intervention. Observation and regular follow-up is advised when the cyst is small and the risk of pathological fracture is minimal. Cases with undisplaced pathological fractures through the cyst were treated with immobilisation by cast application.

Surgical treatment methods known at present were aspiration of the cyst and injection of steroid, aspiration and marrow bone injection, curettage followed by bone grafting, Continuous decompression with elastic intramedullary nails and combination of above treatment methods [9-14]. Curettage and bone grafting is a common procedure routinely performed when there is a displaced pathological fracture due to UBC. Curettage helps in the surgical removal of the cyst membrane from which, the cyst fluid and destructive enzymes are secreted. It helps to connect the cyst with the adjacent bone marrow leading to the healing of UBC [19]. Bone grafting either autograft or allograft, hastens the healing process by acting as osteoconductive material.

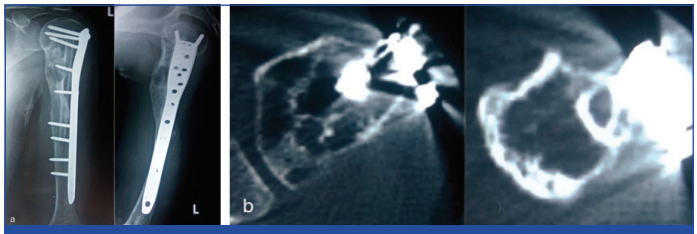
In this study, functional outcome measured by the improvement in MSTS score can be compared to study conducted by Zhang P et al., [25] ($p<0.001$). Mean MSTS score of 27.3 in present study was comparable to the average MSTS score of 28.5 by Erol B [19]. Both the studies mentioned above used elastic intramedullary nails for internal fixation after curettage when compared to this study wherein

S. No.	Age (Years)	Sex	Pathological fracture	Treatment	Follow-up	MSTS score (Preop)	MSTS score at one year	Radiographic healing	VAS (Preop)	VAS (Postop)	Recurrence
1	18	M	Yes	Curetage+Fibular Grafting+Plating	25 months	12	29	Completely healed	7	0	No
2	17	M	Yes	Curetage+Fibular Grafting+Plating	24 months	11	29	Completely healed	9	1	No
3	11	M	Yes	Curetage+Fibular Grafting	37 months	8	26	Partially healed	7	2	No
4	14	M	Yes	Curetage+Fibular Grafting+Plating	13 months	14	24	Recurred at 8 months	8	2	Yes
5	16	F	Yes	Curetage+Fibular Grafting+Plating	22 months	13	27	Completely healed	7	1	No
6	18	F	Yes	Curetage+Fibular Grafting+Plating	12 months	10	27	Recurred at 12 months	7	3	Yes
7	15	M	Yes	Curetage+Fibular Grafting+Plating	27 months	9	28	Recurred at 8 months	8	1	Yes
8	12	F	Yes	Curetage+Fibular Grafting	31 months	14	26	Partially healed	9	2	No
9	16	M	Yes	Curetage+Fibular Grafting+Plating	18 months	13	28	Completely healed	8	2	No
10	19	F	Yes	Curetage+Fibular Grafting+Plating	16 months	12	29	Completely healed	6	0	No

[Table/Fig-8]: Summary of 10 cases.

Variable	Preoperative	Postoperative	p-value
MSTS	11.6±2.06	27.3±1.6	<0.0001
VAS	7.60±0.97	1.40±0.97	<0.0001

[Table/Fig-9]: Comparison of preoperative and postoperative MSTS and VAS scores. Paired t test was used.
p-value <0.001 is considered to be highly significant



[Table/Fig-10]: a) Nine months follow-up radiograph showing recurrence of cyst; b) CT-Scan at nine months showing recurrence of the cyst.

proximal humerus locking plate as an internal fixation device was used in patients above 15 years of age. Plate was used to obtain anatomical reduction and rigid fixation of fibular graft and to allow early range of motion of shoulder. According to a recent study by Wang X et al., the function of the shoulder joint in the elastic nailing group was better than the control group when curettage and mixed bone grafting either with or without elastic intramedullary nailing was done using Neer Score [26].

Several authors compared curettage and bone grafting with steroid injections. Steroid injections have slightly higher healing rates compared to curettage and bone grafting but results were statistically not significant [27-29]. In a retrospective study by Bukva B et al., with 129 patients, curettage of intracystic membrane and implantation of bone graft was reported as a reliable procedure wherein complete healing could only be achieved with open surgery [30]. Study reported by Mik G et al., reported 55 patients who were treated with percutaneous decompression and bone grafting. In this series, 2nd and 3rd surgeries were required to achieve healing in 11 and 2 patients respectively [31]. In a case series of 20 patients reported by Hunt KJ et al, five patients needed second surgery and one patient needed third surgery to achieve healing [32]. In present study, three patients had recurrence and were operated with a second surgery.

In the study published by Zhang P et al., complete healing was reported in 14 cases and partial healing in four cases out of the total 18 cases without any recurrences [25]. In a similar study by Erol B complete and partial radiographic healing was achieved in 28 patients (82%) and six patients (18%), respectively and no recurrence was observed [19]. Similarly, in the study reported by Kokavec M et al., out of the 19 patients with bone cysts treated with curettage and bone grafting only three patients had recurrence during the follow-up yielding an effective rate of 84.2% [15]. Out of the 10 cases in present study, five cases had complete healing of cysts while two cases had healing cysts at one year. The effectiveness of the procedure was 70%. According to recent meta-analysis by Kadhim M et al., surgical curettage has a healing rate of 90% with the supplementation of autograft, allograft or any bone substitution material [33].

In the recent times, use of elastic intramedullary nailing was more popularised. Studies by Roposch A et al., Li W et al., and De Sanctis and Andreacchio A showed that the usage of elastic intramedullary nails has good healing rate and least rate of recurrence [34-36]. This is attributed to the continuous decompression of the cyst cavity by intramedullary nail. The drawback was a higher rate residual cysts. Combining curettage and bone grafting with intramedullary nails has shown good results with less recurrence rate and healing [19]. Fibular bone grafting in this study aids in continuous decompression akin to intramedullary nails.

Mean VAS scores in this study had decreased significantly after surgery from 7.60±0.97 to 1.40±0.97.This may be because of inclusion of the displaced pathological fractures, significant pain relief after the fracture reduction and fixation with plate. The average postoperative VAS score in this study was comparable to the reported score by Zhang P et al., with a significant difference p<0.05 [25].

In present study, three patients (30%) had recurrence after completion of the healing at the end of 12 months. Similarly, Oppenheim WL and Galleno H study reported a recurrence of 43.2% in 37 children with simple bone cysts who underwent curettage and bone grafting [28]. In an observational study by Zhang K et al., curettage and bone grafting without intramedullary nailing was done in 32 cases and 10 cases (32%) had recurrence [18].

Use of proximal humerus locking plate with fibular grafting in this study had an advantage of a stable fixation allowing early range of motion exercises and healing. However, comparison of intramedullary nails and plating in a single study would be more useful in determining a standard treatment approach for UBC of proximal humerus.

Limitation(s)

Small sample size, lack of control group, short follow-up and retrospective nature of study were the limitations of this study.

CONCLUSION(S)

Curettage and fibular grafting with proximal humerus locking plate osteosynthesis for displaced pathological fractures of proximal humerus with UBC has shown good results in terms of functional outcome, cyst healing rate, pain relief and the rate of recurrence.

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- Plagiarism X-checker: Apr 23, 2022
- Manual Googling: May 07, 2022
- iThenticate Software: May 23, 2022 (18%)

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AUTHOR DECLARATION:

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