Sclerotherapy with polidocanol for treatment of aneurysmal bone cysts

Good results in 37 of 38 consecutive patients

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Background and purpose **Recent data suggest that percutaneous sclerotherapy is a safe alternative to surgery for treatment of aneurysmal bone cysts (ABCs). We present our experience of this method.**

Methods **We retrospectively analyzed data from 38 consecutive patients treated with repeated injections of polidocanol. Each injection consisted of 2–4 mg polidocanol per kg body weight. Radiological and clinical assessments were performed until healing.**

Results **All cycts except 1 healed after a median of 4 (1–11) injections. A lesion failed to heal in 1 patient, who was operated. 3 patients experienced minor local inflammatory reactions.**

Interpretation **Our results show that percutaneus sclerotherapy with polidocanol has high efficacy in the treatment of ABCs, with a low frequency of side effects. Our findings corroborate data presented in previous publications. We believe that the method will be especially valuable in ABCs of the pelvis and sacrum, where surgery is associated with considerable morbidity.**

Aneurysmal bone cysts (ABCs) are rare expansile osteolytic tumors with an annual incidence of 0.14 per 10^5; they are usually diagnosed at adolescence, and are equally rare in both sexes (Jaffe and Lichtenstein 1942, Leithner et al. 1999). Management includes combinations of embolization, curettage with or without bone grafting, cementing of the cavity, reconstructive surgery, and—most recently—sclerotherapy.

Sclerosants act by causing damage to the endothelium of vessels and starting a coagulation cascade that results in thrombosis. The use of polidocanol as an endovenous sclerosing agent to treat varicose veins dates from the 1960s, and has been recently shown to be effective in the treatment of ABCs (Jain et al. 2002). In a case series of 72 patients treated with percutaneous intralesional injections of polidocanol (Rastogi et al. 2006), a cure rate of 97% was reported. Similarly, in a prospective study, sclerotherapy was as effective as intralesional excision, but with less morbidity (Varshney et al. 2010).

Overall, sclerotherapy has emerged as a promising treatment that could eventually replace previous methods, which may be associated with considerable morbidity. However, it is not widely accepted and its efficacy remains to be verified in large series of patients. Here we report our experience of percutaneous sclerotherapy of ABCs since we started in 2007.

Patients and methods

From January 2007 to March 2012, all patients in whom radiology and cytology were consistent with ABC were treated with repeated injections of polidocanol. This has been the standard treatment, and no patients were excluded. The patients were retrospectively identified for this study.

38 patients (20 women) were identified, with a mean age of 16 (3–26) years (Table). Diagnosis was based on the radiological characteristics of the lesion (plain radiographs and MRI scans) and confirmed with fine-needle aspiration biopsy. Repeated percutaneous injection of 30 mg/mL polidocanol (2–4 mg polidocanol per kg body weight) under fluoroscopic or CT guidance and general anaesthesia was the mainstay of treatment. Children under 5 years of age often required general anesthesia. The lesion was punctured with a 18-G needle, and fluid was aspirated to verify proper positioning preceding infusion of the sclerosant. Aspiration of a significant amount of bloody fluid from the ABC cavity was interpreted as a sign of remaining active disease. 3 injections at intervals of 4 weeks was the most common schedule, and more injections

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Demographic data and tumor characteristics

Patient no.	Sex	Age at presentation	Size of the tumor (cm)	Location	No. of treatments
1	F	13	4	Femur	6
2	F	9	2	Humerus	3
3	F	17	2	Humerus	6
4	F	26	12	Ischium	3
5	M	17	3	Calcaneus	4
6	F	20	2	Calcaneus	1
7	M	19	4	Femur	4
8	M	20	2	Navicular	2
9	F	11	7	Fibula	4
10	F	25	5	Femur	4
11	M	6	3	Tibia	6
12	F	20	6	Calcaneus	3
13	M	7	3	Radius	6
14	M	14	9	Tibia	1
15	F	15	6	Pubic bone	2
16	M	20	2	Fibula	5
17	M	23	3	Ulna	4
18	F	17	3	Metatarsal	1
19	M	18	6	Humerus	6
20	M	19	6	Ischium	1
21	F	8	5	Fibula	2
22	M	5	5	Femur	4
23	M	16	9	Tibia	3
24	F	17	8	Femur	9
25	M	7	2	Radius	3
26	F	26	4	Calcaneus	3
27	F	15	10	Tibia	3
28	M	24	2	Phalanx hand	11
29	M	20	5	Sacrum	3
30	M	15	9	Humerus	3
31	F	13	4	Sacrum	4
32	F	30	4	Tibia	3
33	F	6	7	Fibula	4
34	F	24	2	Talus	3
35	M	12	6	Acetabulum	4
36	F	10	6	Tibia	4
37	F	12	8	Fibula	1
38	M	3	6	Humerus	3

were given if the lesion failed to heal. Occasionally, injections were targeted to specific loculi that were deemed to be more active. Even a large cyst with pathological fracture and cortical breach was treated as stated above.

The mean follow-up time was 17 (4–37) months. 1 patient was lost to follow-up after the third sclerothepy. Radiological assessment of the tumor (to detect signs of cortical sclerosis, reduction of the volume, and opacification of the cavity) combined with clinical assessment of the patient's symptoms (pain and swelling at the site of the lesion) were performed on an outpatient basis, usually 6–8 weeks after completion of a session of injections. Patients were allowed normal weight bearing and were followed until the lesion showed evidence of sclerosis and the symptoms subsided (defined as healing, which was the endpoint of treatment). Number of injections and amount of sclerosant, time to healing, efficacy, and adverse reactions were recorded.

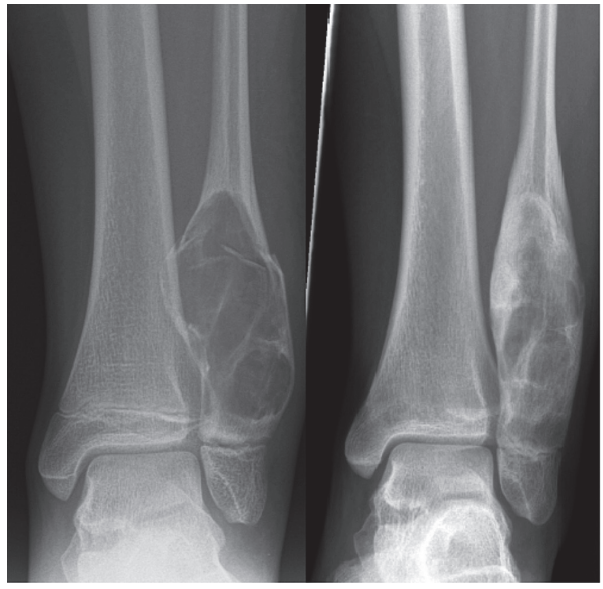

Figure 1. ABC in the distal fibula of a 12-year old female patient (case no. 1), showing consolidation after 4 injections of polidocanol.

Results

The median number of injections per patient was 4 (1–11). 5 patients were cured by a single injection of polidocanol, whereas 8 patients required more than 4 treatments. The amount of sclerosant varied from 30 mg to 300 mg per injection. The average cumulative dose of polidocanol per patient until healing was 450 (60–1,410) mg. The efficacy of the method was 97% (95% CI: 92–100). Only 1 patient had progressive disease despite repeated sclerotherapy for an ABC of the ulna, and proceeded to open surgery (curettage and filling of the cavity with polymethylmethacrylate), whereupon the symptoms subsided. There was 1 documented case of failure to successfully re-inject a lesion (second metatarsal) following 1 successful injection, but even so, healing was observed.

33 patients had no residual pain following their course of injections and had convincing radiological sclerosis; 2 patients had a minor amount of intermittent residual pain but the lesions were radiologically healed. 2 patients are still on routine follow-up and their symptoms have clearly improved. Representative cases are presented in Figures 1 and 2, depicting consolidation of the lesions after serial sclerotherapy.

There were no cases of anaphylaxis, major adverse reactions, infection, or local necrosis at the injection site. Minor local inflammatory reactions were observed in 3 patients (92% safety, 95% CI: 83–100) whereas 1 child required hospital admission overnight following injection due to severe pain and 2 patients required 1 week of simple oral non-opioid analgesia. A moderate flexion contracture was observed in 1 patient who was treated with repeated injections for an ABC with a pathological fracture in a digit. We did not observe any cases of growth arrest in our series, which included 15 children.

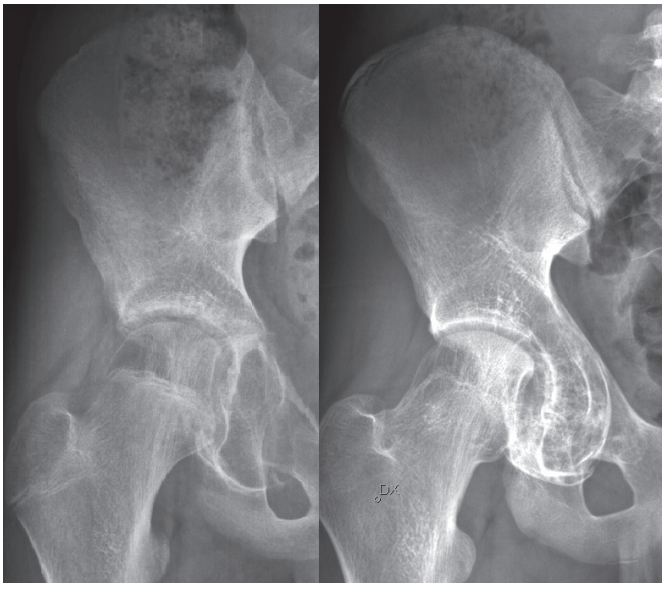

Figure 2. ABC in the acetabulum of a 19-year old man (case no. 2), which healed after 4 injections of polidocanol.

Discussion

The optimal treatment method for ABC is still being debated. Open curettage with or without bone grafting is a widely accepted mode of treatment, but it is accompanied by a high recurrence rate of approximately 30%, which can be reduced to 15% when a high-speed burr is used (reviewed by Varshney et al. 2010). Wide en-bloc resection gives excellent results in terms of local control, which approximates 100%. Yet, wide surgical margins are often not feasible, as the lesion can be close to neurovascular structures. Furthermore, extensive surgery is associated with considerable morbidity. Cumulative data suggest a growth disturbance rate of about 10% after various surgical procedures (Capanna et al. 1985, Green et al. 1997, Rizzo et al. 1999, Lampasi et al. 2007).

Radiation therapy has also been used, with local control rates that are comparable to those for intralesional therapy (Nobler et al. 1968, Feigenberg et al. 2001). However, a major concern is the risk of secondary malignancies, especially in the case of younger patients (Marcove et al. 1995). Other potential complications are the possibility of epiphyseal arrest and secondary deformity. The use of high-precision megavoltage radiotherapy and percutaneous radionuclide ablation has given better results, but is yet to be evaluated in larger series of patients (Feigenberg et al. 2001, Bush et al. 2010).

Embolization of the feeding arteries has also been suggested as an alternative, with good results reported (Amendola et al. 2013). However, the procedure is technically demanding and is not applicable to all cases, as ABCs often lack large afferent vessels. When used for the treatment of ABC of the spine, selective angiography is necessary to ensure that there is no risk of spinal cord ischemia. Thus, it is usually regarded as a supplement to surgery (Boriani et al. 2001).

Polidocanol sclerotherapy compares favorably with the above treatments. In our hands and also in previous reports (Rastogi et al. 2006, Varshney et al. 2010), it has an efficacy exceeding 90%. Furthermore, the treatment is simple and carries negligible risk of morbidity, there is no scar formation, and it can be reliably performed as a day-case surgery. The method is applicable to all cases, and does not require sophisticated technical equipment. Most importantly, sclerotherapy is effective in the case of lesions of the pelvis and sacrum that are difficult to treat surgically due to the risk of heavy bleeding and other major complications. In our series, all 6 patients who presented with ABC in this region healed uneventfully.

The need for multiple injections and prolonged treatment is an obvious disadvantage. Our data corroborate the view that multiple treatments are required for most patients (79–86% in the studies by Rastogi et al. 2006 and Varshney et al. 2010). However, prolonged follow-up is also the case for patients treated with surgery, radiotherapy, and embolization as healing of ABC is generally delayed. Varshney et al. (2010) reported growth disturbances in 4% of their patients treated with polidocanol. We did not observe any such case in our series.

The use of polidocanol is a definite advancement over previous sclerotherapy regimens that relied on alcoholic zein solutions, which were more toxic and had serious adverse effects after spill-out into nearby tissues (Falappa et al. 2002, Adamsbaum et al. 2003, Topouchian et al. 2004). There was one documented fatal outcome after injection in an ABC of the second cervical vertebra (Peraud et al. 2004). Indeed, we observed only minor transient inflammatory reactions, which is in line with previous studies (Rastogi et al. 2006, Varshney et al. 2010).

Sequential percutaneous administration of polidocanol is the standard treatment for ABC at our institution, as we have found it to be a safe, simple procedure with an excellent cure rate. All data that have accumulated thus far strongly favor sclerotherapy with polidocanol over surgery and suggest that it could become the treatment of choice for ABC.

OB, AH, and HB: patient treatment and follow-up. OB, PP, and PT: collection and analysis of data. PT, PP, AH, HB, and OB: preparation of the manuscript.

No competing interests declared.

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