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# Aneurysmal Bone Cysts of the Craniofacial Origin: A Systematic Review

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## Abstract

*Objective*. Aneurysmal bone cysts (ABCs) are blood-filled, locally destructive, benign bone tumors. Our objective was to conduct a systematic review outlining patient demographics, clinical characteristics, management, and outcomes of those with ABCs of the craniofacial bones.

*Data Sources*. Using PubMed, Cochrane, and Embase databases, 116 studies were included.

*Review Methods*. Following Preferred Reporting Items for Systematic Reviews and Meta-Analysis guidelines, a systematic review was conducted. Data including patient demographics, clinical characteristics, treatment strategies, and patient outcomes were collected.

*Results*. A total of 127 patients from 116 studies were identified. Age ranged from 8 months to 90 years, with a mean age of 19.0 years. The most commonly affected craniofacial locations were the mandible (n = 31, 24.4%), temporal bone (n = 21, 16.5%), and occipital bone (n = 14, 11.0%). The most common presenting symptoms included a nontender mass (n = 51, 40.2%), a tender mass (n = 31, 24.4%), and generalized headache (n = 30, 23.6%). Imaging modalities included computed tomography (CT) and magnetic resonance imaging (MRI) (n = 77, 60.6%), CT alone (n = 31, 24.4%), and MRI alone (n = 8, 6.2%). All patients underwent surgical resection, with 1 patient requiring adjuvant radiation in addition to surgery. In total, 121 patients were disease-free and symptom-free without evidence of recurrence (17.4-month mean follow-up, 5.4 months average time to first recurrence).

*Conclusion*. The current literature's characterization of ABCs in craniofacial bones is limited to case reports and case series. Given the rarity of these tumors, head and neck surgeons may rely on systematic reviews such as the present analysis to guide management.

## Keywords

aneurysmal bone cyst, craniofacial, otolaryngology, head and neck, cancer

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Aneurysmal bone cysts (ABCs) are blood-filled, locally destructive, benign cystic bone tumors that account for about 9.1% of all primary bone tumors.[1] They typically arise during the second decade of life, with an average age of onset at 13 years, and 90% of cases are diagnosed before the age of 30.[2] ABCs are slightly more prevalent in females than males.[2] While they predominantly arise in the metaphysis of long bones, such as the femur, tibia, fibula, and upper extremity, they may arise in a variety of locations such as the spine, sacrum, pelvis, foot, and fingers.[3] In these locations, ABCs may present with pain, swelling, or an associated pathological fracture of the bone.[3] However, there may be additional symptoms based on the location at which the tumor arises, such as neurological deficits secondary to spinal cord compression.[3] The variety of symptoms is especially worrisome for ABCs that arise in the skull bones, which account for 2% to 6% of total cases.[4,5] ABCs of craniofacial origin may present with a variety of symptoms, including masses, decreased hearing, worsening headaches, and visual disturbances, all of which depend on the craniofacial bone from which it arises.[5-7] Given the diversity of symptoms that may arise in ABCs of craniofacial origin, it is vital that physicians remain aware of the various nonspecific presentations of ABCs when assessing patients for craniofacial symptoms. The differential diagnoses for ABCs include but are not limited to chondroblastomas, fibrous dysplasia, giant cell tumors, telangiectatic osteosarcomas, and unicameral bone cysts.[8] Imaging can be extremely helpful in not only identifying the disease but also characterizing the extent of the disease.

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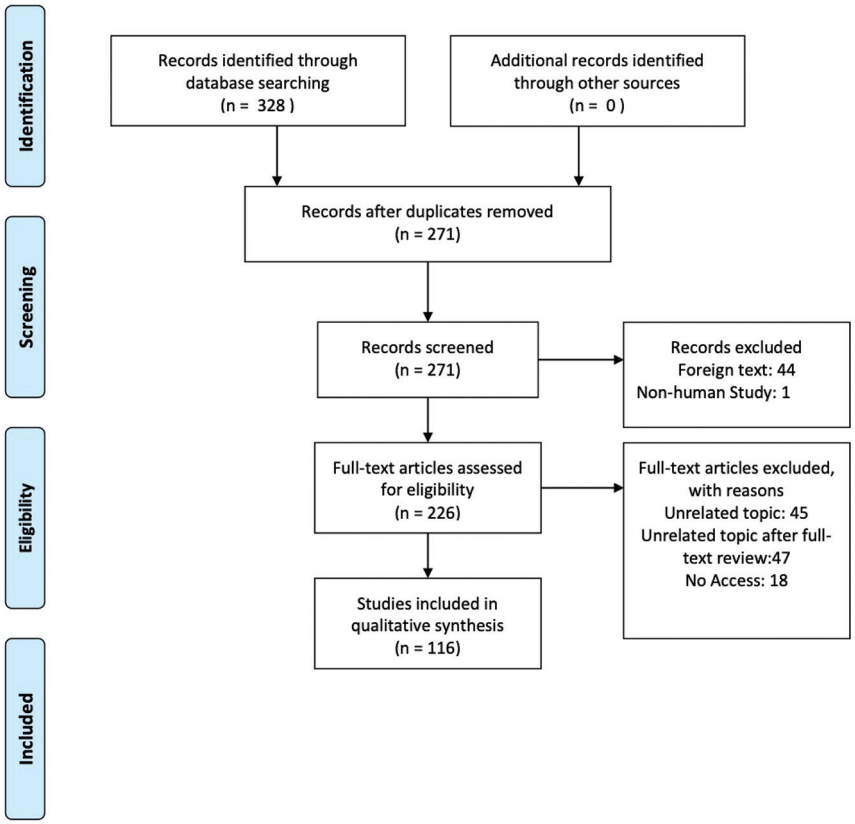


**Figure 1.** Article selection process based on Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) and the search algorithm employed.

Due to the rarity of ABCs of craniofacial origin, its characterization has primarily been limited to case reports and case series. To our knowledge, there has been no comprehensive systematic review that adequately summarizes the characteristics and management of ABCs in the craniofacial bones. To address this, our systematic review explores the patient demographics, tumor characteristics, manifestation, diagnosis, forms of management, and outcomes of ABCs in the craniofacial bones.

## Methods

This systematic review was performed following the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines. The authors (R.R. and J.A.) conducted a comprehensive literature search of EmBase, Cochrane Library, and PubMed to identify studies relating to ABCs in the craniofacial bones. The search was conducted in July 2019 and had major keywords related to ABCs of the craniofacial bones and its treatment, including the following: *aneurysmal bone cysts, craniofacial bones, drug therapy, chemotherapy, immunotherapy, surgery, and radiation therapy.* The authors did not place any date limitations while searching the databases. Studies were excluded if they were non-English, non-human subjects; review articles; or not within the scope of the selected topic. The authors also searched the bibliographies of included studies to identify any additional pertinent references. Studies that were unavailable to be initially accessed were requested by the authors. Discrepancies between the reviewers were resolved by a third reviewer (A.D.). **Figure 1** summarizes both the search and inclusion strategy used to locate pertinent studies.

The quality of the included studies and the risk of bias were evaluated using the Grades of Recommendation, Assessment, Development and Evaluation Group (GRADE) criteria, the Methodological Index for Non-Randomized Studies (MINORS) criteria, and the Oxford Center for Evidence-Based Medicine Levels of evidence.

### *Study Selection and Inclusion Criteria*

Titles and abstracts of retrieved studies were initially reviewed for inclusion by 2 authors (R.R. and J.A.). The studies must have discussed the ABC characteristics and management in order to be included. After screening of the title and abstract, the complete texts of the remaining articles were reviewed for relevance. Relevant studies should have discussed the tumor's presenting symptoms, characteristics, treatment modalities, and outcomes.

### *Data Collection*

At the conclusion of the screening process, 116 studies met the inclusion criteria and were analyzed for data extraction. For the included studies, patient demographics, not limited to

age and sex, were collected. In addition, the authors extracted the clinical characteristics of each case, including the tumor characteristics, manifestations, diagnostic modalities, management strategies, and outcomes. The outcomes collected include postoperative complications, follow-up time, and evidence of recurrence, if applicable.

## Results

A total of 127 patients from 116 studies on ABCs in the craniofacial bones were examined. Out of 127 included patients, sex was reported for 126 patients, with 54% male (n = 68) and 46% female (n = 58). The quality of evidence using the GRADE and MINORS criteria was low to moderate. The Oxford Levels of Evidence scores and quality of evidence are illustrated in Supplementary Table S1 (in the online version of the article). The mean age of patients was 19.0 years, ranging from 8 months to 90 years. **Table 1** illustrates a summary of the patient-level findings from included studies in this systematic review.

### *Tumor Location*

All of the studies examined in this review were characterized as ABCs of craniofacial origin. The 3 most commonly affected craniofacial locations were the mandible (n = 31, 24.4%), temporal bone (n = 21, 16.5%), and occipital bone (n = 14, 11.0%). Other less commonly involved locations included the frontal bone (n = 12, 9.4%), the sphenoid bone (n = 10, 7.9%), the maxilla (n = 9, 7.1%), skull base unspecified (n = 8, 6.3%), ethmoid bone (n = 6, 4.7%) the orbit (n = 6, 4.7%), the parietal bone (n = 5, 3.9%), and nasal bones (n = 5, 3.9%). **Table 2** depicts a comprehensive summary of the tumor locations.

### *Clinical Presentation*

Presenting symptoms were reported for all 127 patients. The 3 overall most common presenting symptoms of ABCs located in the craniofacial bones included a nontender mass (n = 51, 40.2%), a tender mass (n = 31, 24.4%), and headache (n = 30, 23.6%). Stratified by location, the most common presenting symptoms were mandible (nontender mass, 54.8%), temporal bone (nontender mass, 38.0%), occipital bone (headache, 64.2%), frontal bone (headache, 41.6%), sphenoid bone (headache, 60.0%), maxilla (nontender mass, 55.5%), skull base unspecified (decreased visual acuity, 37.5%), ethmoid bone (rhinorrhea, 33.3%), orbit (proptosis, 83.3%), parietal bone (nontender mass, 60.0%), and nasal region (nasal obstruction, 60.0%). **Table 1** illustrates a comprehensive summary of the presenting symptoms.

Furthermore, out of the 127 reported cases, 8 cases of ABCs (6.3%) were reported to arise secondary to fibrous dysplasia, 4 cases of ABCs (3.1%) were reported to be secondary to ossifying fibroma, 2 cases of ABCs (1.6%) were reported to arise secondary to osteoblastoma, and 1 case of ABC (0.8%) was reported to arise secondary to eosinophilic granuloma. A more complete summary of the etiologies can be found in the Supplementary Table S1 (in the online version of the article).

**Table 1.** Clinical Characteristics.

| Characteristic | Value |
|---|---|
| No. of subjects | 127 |
| Male, n (%) | 68 (54) |
| Female, n (%) | 58 (46) |
| Age, mean (range), y | 19.0 (0.6-90) |
| Location, n (%) | |
| Mandible | 31 (24.4) |
| Temporal | 21 (16.5) |
| Occipital | 14 (11.0) |
| Frontal | 12 (9.4) |
| Sphenoid | 10 (7.9) |
| Maxilla | 9 (7.1) |
| Skull base unspecified | 8 (6.3) |
| Ethmoid bone | 6 (4.7) |
| Orbit | 6 (4.7) |
| Parietal bone | 5 (3.9) |
| Nasal region | 5 (3.9) |
| Presenting symptoms, n (%) | |
| Nontender mass | 51 (40.2) |
| Tender mass | 31 (24.4) |
| Headache | 30 (23.6) |
| Vomiting | 7 (5.5) |
| Eye, n (%) | |
| Proptosis | 12 (9.4) |
| Decreased visual acuity | 10 (7.9) |
| Diplopia | 8 (6.3) |
| Ear, n (%) | |
| Hearing loss | 7 (5.5) |
| Tinnitus | 2 (1.6) |
| Nose, n (%) | |
| Nasal obstruction | 9 (7.1) |
| Anosmia | 4 (3.1) |
| Rhinorrhea | 3 (2.4) |

### *Imaging*

Imaging was reported for all 127 patients. The 3 most commonly used techniques were a combination of computed tomography (CT) scan and magnetic resonance imaging (MRI) (n = 77, 60.6%), CT alone (n = 31, 24.4%), and MRI alone (n = 8, 6.2%). Other radiological techniques included X-ray, bone scan, ultrasound, and angiography. Ansari et al[9] depicted an excellent CT scan and MRI images of an ABC arising in the temporal bone, while Breuer et al[10] depicted an excellent histological image of a mandibular ABC.

### *Management*

Management of ABCs included surgical intervention for all 127 cases. Of the specific surgeries reported, the 3 most common techniques included craniotomies (n = 38, 29.9%), rhinotomy (n = 4, 3.1%), and osteotomy (n = 3, 2.3%). The en bloc technique was reported in 8 cases (6.2%) while the curettage technique was reported in 7 cases (5.5%). Radiation was

only used in 1 case (0.7%) in conjunction with surgery. When reported, follow-up times ranged from 0.1 to 60 months, with an average of 17.4 months. A more detailed list of approaches and follow-up times can be found in **Table 3** and the Supplementary Table S1 (in the online version of the article).

### *Outcomes*

Most patients (n = 121, 95.3%) were disease-free without evidence of recurrence. Recurrence did occur in 4 cases averaging a follow-up time of 5.4 months, with times ranging from 1 to 12 months after the initial treatment. In addition, 2 patients continued to experience presenting symptoms despite no recurrence. Complications varied depending on presenting location and included bone deformity, compressed dura, subgaleal fluid collection, mandibular protrusion, residual fibrous dysplasia, and wound infections. None of the patients were reported deceased following treatment. A more detailed analysis of management and outcomes stratified by tumor location can be found in **Table 3**.

## Discussion

ABCs are blood-filled, locally destructive, benign cystic bone tumors that account for 9.1% of all primary bone tumors.[1] Interestingly, the name *aneurysmal bone cyst* has been proved to be a misnomer, as they are neither cystic nor aneurysmal.[3] Instead, these lesions form blood-filled cavities within bone that are lined by fibroblasts, giant cells, and trabecular bone.[11] Grossly, they appear as spongy, hemorrhagic masses engrossed in a thin shell of reactive bone.[3] ABCs have recently been associated with the specific translocation t(16;17) (q22;p13), which causes a gain-of-function mutation in TRE17/USP6 (ubiquitin-specific protease USP6 gene).[12] While they usually arise in the metaphysis of long bones such as the femur, tibia/fibula, and upper extremity, they may arise anywhere in the spine, sacrum, pelvis foot, and fingers.[3] They rarely arise in the craniofacial bones, accounting for only 2% to 6% of total cases.[4,5] Due to the scarcity of reports in the literature, there has yet to be a clear consensus on the management or clinical outcomes for ABCs arising in the craniofacial bones. The purpose of this systematic review is to review the relevant cases in the literature to allow for improved representations and management of these tumors.

### *Epidemiology/Patient Demographics*

The incidence of ABCs in the population is approximately 0.14 cases per 100,000 people per year.[13] As previously reported, ABCs are generally more prevalent in females compared to males.[13,14] However, our analysis of ABCs of the craniofacial bones revealed that tumors more commonly arose in males (54%) compared to females (46%). While ABCs are encountered in all decades of life, 75% to 90% of ABCs appear in the second decade of life, with 90% being diagnosed before the age of 30.[2,13] Our study supports these findings as 94 of 127 (74.0%) patients were 20 years or younger, and 15 of 127 (11.8%) patients were between ages 21 and 30 years. Given this, 109 of 127 (85.8%) patients were 30 years or younger at the time of diagnosis with ABCs in the craniofacial bones, which is consistent with the literature. However, our

**Table 2.** Clinical Characteristics by Location.[a]

| *Characteristics* | *Data* |
|---|---|
| No. of subjects | 127 |
| Male, n (%) | 68 (54) |
| Female, n (%) | 58 (46) |
| Mean age, y | 19.0 |
| *Presenting symptoms, n (%)* | |
| Mandible, n | 31 |
| Nontender mass | 17 (54.8) |
| Tender mass | 10 (32.3) |
| Not reported | 4 (12.9) |
| Temporal bone, n | 21 |
| Nontender mass | 8 (38.1) |
| Tender mass | 5 (23.8) |
| Headache | 6 (28.6) |
| Facial paralysis | 4 (19.0) |
| Hearing loss | 6 (28.6) |
| Ear pain | 1 (4.8) |
| Tinnitus | 1 (4.8) |
| Decreased visual acuity | 1 (4.8) |
| Vomiting | 3 (14.3) |
| Occipital bone, n | 14 |
| Nontender mass | 6 (42.9) |
| Tender mass | 4 (28.6) |
| Headache | 9 (64.3) |
| Decreased visual acuity | 1 (7.1) |
| Retinal hemorrhage | 1 (7.1) |
| Vomiting | 2 (14.3) |
| Frontal bone, n | 12 |
| Nontender mass | 4 (33.3) |
| Tender mass | 4 (33.3) |
| Headache | 5 (41.7) |
| Diplopia | 1 (8.3) |
| Proptosis | 1 (8.3) |
| Vomiting | 1 (8.3) |
| Sphenoid, n | 10 |
| Nontender mass | 2 (20.0) |
| Headache | 6 (60.0) |
| Decreased visual acuity | 3 (30.0) |
| Diplopia | 2 (20.0) |
| Proptosis | 2 (20.0) |
| Maxilla, n | 9 |
| Nontender mass | 5 (55.5) |
| Tender mass | 3 (33.3) |
| Headache | 1 (11.1) |
| Facial paralysis | 1 (11.1) |
| Diplopia | 1 (11.1) |
| Strabismus | 1 (11.1) |
| Nasal obstruction | 3 (44.4) |
| Skull base unspecified, n | 8 |
| Nontender mass | 2 (25.0) |
| Headache | 1 (12.5) |
| Decreased visual acuity | 3 (37.5) |

*(continued)*

analysis also revealed that patients with ABCs of craniofacial origin ranged from 8 months to 90 years of age, hence emphasizing that clinicians should be aware of the age predominance but not dismiss alarming symptoms as outliers do exist.

## Tumor Locations and Clinical Presentation

About 2% to 6% of ABCs arise in the craniofacial bones; therefore, the tumor location and clinical presentation of the tumor have not been well characterized in the literature.[4,5] Given that ABCs are most commonly seen in the metaphysis of long bones of the lower extremity and posterior spinal elements, their presenting symptoms may include pathologic fractures and neurological symptoms, respectively.[15-17] The most common presenting locations of ABCs in the craniofacial bones were the mandible, temporal bone, and occipital bone with a frequency of 24.4%, 16.5%, and 11.0%, respectively. However, our review documented ABCs can arise in a variety of craniofacial bones, including the sphenoid and nasal bones, among others. While ABCs are benign tumors of the musculoskeletal system, they may behave as locally aggressive neoplasms and present with a variety of symptoms. The clinical characteristics of ABCs in the craniofacial bones occur via the mass effect of the tumor. Our analysis showed that the 3 overall most common presenting symptoms included a nontender mass (n = 51, 40.2%), a tender mass (n = 31, 24.4%), and headache (n = 30, 23.6%). Given the heterogeneity of the symptoms, clinicians should maintain a high index of suspicion of ABCs depending on the exact location and presenting symptoms.

## Diagnosis and Differential Diagnoses

With the possible age range and nonspecific manifestations of ABCs of the craniofacial bones, their diagnosis may be difficult. Various imaging modalities are required for the preliminary identification of ABCs. X-rays are commonly the initial radiograph obtained followed by other modalities to increase sensitivity and specificity of ABC identification.[3,18] In our analysis, the most commonly ordered radiological technique was a combination of a CT scan and an MRI. Radiological characteristics include multilocular ''soap bubble'' lesions on X-ray, fluid-filled levels representing layering of different blood levels and internal septations on MRI, and lytic lesions with a thin outer residual rim of bone shell on CT.[6,18] Furthermore, the internal cysts of the lesion display low to medium intensity on T1 MRI and high intensity on T2 MRI, in addition to a hypointense rim, suggestive of a benign lesion.[6] However, none of these findings are pathognomonic for ABCs, and an incisional biopsy is required for accurate diagnosis and ruling out of other pathologies with similar imaging characteristic such as telangiectatic osteosarcoma.[3,18] Biopsy displays abundant red blood cells and pale brown hemosiderin pigments within the cyst-like spaces lined by fibroblasts, calcifications, osteoid, spindle cells, and scattered multinucleated giant cells.[19]

The differential diagnosis for ABCs is diverse, including unicameral bone tumor, giant cell tumor, telangiectatic osteosarcoma, and chondroblastoma, among others. The 3 most common juvenile craniofacial bony lesions are fibrous dysplasia, Langerhans histiocytosis X, and ABC.[20] In addition, the literature reports that approximately 70% of ABCs are primary and 30% are secondary to another underlying tumor.[21] It is important to distinguish primary ABCs from secondary ABCs in order to uncover potential underlying destructive bone tumors such as fibrous dysplasia, osteoblastoma, chondroblastoma, and giant cell tumor.[11,15,22] In our review, the most common type of secondary ABCs was due to fibrous dysplasia; hence, our analysis aligns with the current literature in that secondary ABCs are most likely due to underlying fibrous dysplasia. As previously mentioned, the definitive diagnosis and exclusion of other musculoskeletal neoplasms are through an incisional biopsy. Given the current literature's review and our present analysis, it is crucial for attending health care professionals to obtain radiological imaging in addition to an incisional biopsy for the definitive diagnosis of ABCs.

**Table 2.** (continued)

| *Characteristics* | *Data* |
|---|---|
| Proptosis | 2 (25.0) |
| Tinnitus | 1 (12.5) |
| Nasal obstruction | 2 (25.0) |
| Anosmia | 1 (12.5) |
| Hearing loss | 1 (12.5) |
| Ethmoid bone, n | 6 |
| Nontender mass | 2 (33.3) |
| Diplopia | 1 (16.6) |
| Decreased visual acuity | 1 (16.6) |
| Proptosis | 1 (16.6) |
| Headache | 1 (16.6) |
| Epistaxis | 2 (33.3) |
| Rhinorrhea | 2 (33.3) |
| Nasal obstruction | 1 (16.6) |
| Anosmia | 1 (16.6) |
| Orbit, n | 6 |
| Nontender mass | 1 (16.6) |
| Tender mass | 1 (16.6) |
| Proptosis | 5 (83.3) |
| Diplopia | 2 (33.3) |
| Parietal, n | 5 |
| Nontender mass | 3 (60.0) |
| Tender mass | 2 (40.0) |
| Headache | 1 (20.0) |
| Diplopia | 1 (20.0) |
| Nasal region, n | 5 |
| Nontender mass | 1 (20.0) |
| Tender mass | 2 (40.0) |
| Proptosis | 1 (20.0) |
| Nasal obstruction | 3 (60.0) |
| Anosmia | 2 (40.0) |
| Rhinorrhea | 1 (20.0) |

[a]Some aneurysmal bone cysts presented with multiple symptoms in the same region.

**Table 3.** Overview of ABC Cases by Location.

| Etiology | Studies | Patients | Age, mean (range), y | Treatment modality | Recurrence |
|---|---|---|---|---|---|
| Mandible | 25 | 31 | 19.0 (6-37) | Unspecified surgery ×17, curette ×5, parotidectomy ×3, condylectomy ×2, corticotomy ×2, mandibulectomy ×1, osteotomy ×1 | No recurrence ×30, recurrence ×1 |
| Temporal bone | 21 | 21 | 20.2 (4-61) | Unspecified surgery ×11, craniotomy ×6, mastoidectomy ×3, osteotomy ×1, radiation ×1, curette ×1 | No recurrence ×18, recurrence ×2, failure of symptoms to resolve ×1 |
| Occipital bone | 12 | 14 | 17.3 (2-54) | Unspecified surgery ×6, craniotomy ×6, cranioplasty ×2, curette ×1 | No recurrence ×14 |
| Frontal bone | 12 | 12 | 19.91 (11-73) | Craniotomy ×9, en bloc resection ×3, cranioplasty ×2, unspecified surgery ×1 | No recurrence ×12 |
| Sphenoid bone | 10 | 10 | 14.8 (0.83-28) | Unspecified surgery ×2, craniotomy ×5, maxillectomy ×1, osteotomy ×1, ethmoidectomy ×1 | No recurrence ×10 |
| Maxilla | 9 | 9 | 18.1 (5-45) | Unspecified surgery ×5, maxillectomy ×1, rhinotomy ×1, ethmoidectomy ×1, sphenoidotomy ×1 | No recurrence ×9 |
| Skull base unspecified | 8 | 8 | 16.5 (6.5-41) | Unspecified surgery ×1, craniotomy ×6, en bloc resection ×1 | No recurrence ×7, recurrence ×1 |
| Ethmoid bone | 6 | 6 | 34.3 (8-90) | Unspecified surgery ×3, craniotomy ×2, rhinotomy ×1 | No recurrence ×6 |
| Orbit | 6 | 6 | 16.3 (1-44) | Unspecified surgery ×1, craniotomy ×4, maxillectomy ×1, rhinotomy ×1 | No recurrence ×6 |
| Parietal bone | 5 | 5 | 17.0 (9-28) | Unspecified surgery ×4, lateral orbitotomy ×1, en bloc resection ×1 | No recurrence ×5 |
| Nasal region | 5 | 5 | 12.0 (5-23) | Rhinotomy ×1, endoscopic ×4 | No recurrence ×5 |

### *Management and Outcomes*

ABCs are currently managed through surgical intervention, mainly by resection and reconstruction of the defect using a bone graft and occasional curettage.[3] Other treatment modalities include en bloc resection or adjuvant radiotherapy, which are typically reserved for atypical cases in which excision may be difficult or in the case of ABC recurrence.[3] The findings in our current analysis were similar, as all of the patients with ABCs of craniofacial bones underwent surgical resection using various techniques. One patient (0.8%) in our review underwent adjuvant radiotherapy for an ABC of the temporal bone, with no evidence of recurrence.[9] The limited use of radiotherapy may be due to the sequelae of radiotherapy-induced sarcoma, which has been reported in the literature.[23] Additional therapies for the treatment of ABCs may include high-speed burr, argon beam coagulation, and arterial embolization.[3] In a study conducted by Lee et al,[20] the treatment of secondary ABCs in a craniofacial fibrous dysplasia included total resection of the ABC and cranioplasty in addition preoperative embolization of the feeding artery to decrease intraoperative bleeding.[24] In our analysis, 4 patients underwent preoperative embolization of the feeding artery.

The recurrence rate for ABCs is high, ranging from 10% to 50%.[15,16] Many factors may play into the recurrence rate, including histological features, patient ages, and management. Increased osteoid or fibromyxoid features on histology, younger age, and curettage and bone grafting alone are positively associated with recurrence of ABCs.[25,26] In addition, neither clinical features nor radiological findings were associated with recurrence rates.[26] However, recurrence rates for ABCs of craniofacial bones may differ and are not reported in the literature. In our analysis, only 4 of 127 (3.14%) patients had a recurrence with a mean follow-up time of 5.4 months. Based on the current evidence, we recommend that otolaryngologists monitor recurring symptoms in patients with a history of an ABC in the craniofacial bones, especially within the first 6-month period after treatment.

### *Limitations*

While the present study summarizes patient demographics, tumor characteristics, clinical manifestations, diagnosis, management, and outcomes ABCs in the craniofacial bones, there are several limitations inherent to the study design. As with all systematic reviews, there is the possibility that studies may have been missed during the initial identification of the pertinent studies. To reduce this, the authors reviewed the literature on multiple instances to confirm the inclusion of pertinent articles discussing ABCs of the craniofacial bones

only. Furthermore, likely to due to the rarity of these tumors, there are currently no randomized controlled trials on ABCs of the craniofacial bones in the literature to the best of our knowledge. Thus, the data for this study consists entirely of case reports and retrospective review case series. The latter study design may be slightly biased as it consists of multiple cases at a single institution. In addition, there is also a potential for publication bias, as case reports with unfavorable outcomes are less likely to be reported and published, which may account for the high recurrence-free data reported in this study. Despite this, systematic reviews such as the present study are integral to summarizing the patient demographics, tumor characteristics, manifestations, diagnosis, management, and outcomes for the limited patient population of ABCs in the craniofacial bones. Additional randomized controlled trials are needed to confirm the findings of this study. Given the rarity of these tumors, we should rely even more heavily on a systematic review such as the current analysis to guide management for these patients.

## Conclusion

Aneurysmal bone cysts are benign, cystic, locally destructive musculoskeletal neoplasms that less commonly arise in the craniofacial bones during the second decade of life. The 3 most common presenting locations of ABCs are the mandible, temporal bone, and occipital bone with various nonspecific symptoms, including a tender or nontender mass, headache, and vision loss. The diagnosis of ABCs is confirmed with excisional biopsy and resection, and other adjuvant therapies such as curettage may be needed to treat and prevent recurrence of ABCs in the craniofacial bones. Due to the low recurrence rates and high rates of disease-free survival, we recommend that clinicians keep a high index of suspicion for ABCs of craniofacial origin. Randomized control studies are needed to confirm the findings of this study in order to create standard treatment guidelines for ABCs in the craniofacial bones.

### Author Contributions

**Rafey Rehman**, primary author with significant contributions to the whole project, especially study acquisition, data analysis and interpretation of data, along with writing of the manuscript; **Antonio Dekhou**, significant contribution to screening process, data acquisition, and writing of the manuscript; **Muhammad Osto**, significant contribution to data analysis, along with writing, final revision, and formatting of the manuscript; **Jacob Agemy**, significant contribution to literature search, screening process, and data acquisition; **Amneah Chaaban**, significant contribution to literature search, screening process, and data acquisition; **Brian Yuhan**, drafted manuscript and reviewed versions of manuscript; **Eric Thorpe**, reviewed final versions of manuscript and helped draft the discussion.

### Disclosures

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### Supplemental Material

Additional supporting information is available at http://journals.sagepub.com/doi/suppl/10.1177/2473974X211052950

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