Odontogenic myxoma: Report of three cases and retrospective review of literature in Indian population

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Abstract

Purpose: To review the clinical pattern and treatment modalities meted out to patients of odontogenic myxoma (OM) in Indian population in last 30 years and also to report 3 cases of OM. **Method:** A retrospective review of radiograph and histopathology of three cases of odontogenic myxoma treated between 2005 and 2011 was done. Immunohistochemical analysis was performed to examine the pattern of vimentin and NSE. Also a computerized literature search using Medline and Google scholar was conducted for published articles on OM in Indian population in last 30 years. **Result:** A total of 32 OM cases reviewed from 25 articles retrieved. Out of them 24 myxomas were present in maxilla, only 8 were in mandible and a single case was present in supraglottic region (larynx). Surgical procedure carried out was excision and curettage in 16 patients and resection with safe margin followed by reconstruction 13 patients. All the three reported cases were successfully treated by tumor enucleation and peripheral ostectomy with no recurrence after 3 to 7 years. **Conclusion:** Odontogenic myxoma is a rare odontogenic tumor with inconclusive clinical and radiographic features, hence histopathological examination is mandatory to confirm its diagnosis. Although the immunohistochemical analysis may help in diagnosis but plays no role in guiding treatment planning or predicting the rate of recurrence. Currently we lack data on number of reported OM cases in Indian population as author feels more patients must have been treated then reported.

Keywords: Indian population, maxilla, mandible, odontogenic myxoma, peripheral ostectomy

Introduction

The odontogenic myxoma (OM) of the jaws is a rare benign tumor characterized grossly by mucoid or gelatinous gray-whitish tissue that replaces the cancellous bone and expands the cortex. It is an asymptomatic lesion that shows an infiltrative growth pattern. It is invasive locally and has a high recurrence rate accounting 1-19% of all odontogenic tumors,[1] affecting mostly young patients in their second and third decade of life.[2] In general, the radiographic features are not pathognomonic of the lesion, but the most common presentation is a multilocular radiolucency.

The purpose of the study was to evaluate the clinical pattern and different treatment modalities meted out and assess their outcome published by the Indian authors in the last 30 years. The aim of this article was also to describe the three clinical cases diagnosed histopathologically as OM and illustrate their clinical, radiographic, histopathologic characteristics, and treatment with follow-up of 7 years.

Case Series

Medical records and radiographs of 3 patients with the diagnosed cases of myxoma were retrieved from the departmental archives of the Department of Oral Pathology and Oral and Maxillofacial Surgery Department of the Maulana Azad Institute of Dental Sciences, New Delhi. Radiological analysis was made on panoramic radiographs available in all cases. Immunohistochemical analysis was also done.

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Case report 1

A 7-year-old boy presented to Maxillofacial Surgery Department with the complaint of nontender swelling in right maxillary region since last 6 months. Swelling was beneath the right infraorbital region extending from lateral to ala of nose and going posteriorly till the arch of zygomatic bone. Intraorally, there was an expansion of buccal and palatal cortices from upper right canine to first molar on the same side. Overlying mucosa was of pink color, nonulcerated, and firm on palpation. The patient had squint in right eye since birth [Figure 1].

Computerized tomography (CT) scan revealed a homogenous mass in the right maxillary antrum region compressing medially, superiorly it was seen abutting floor of orbit, and inferiorly expanding maxillary alveolus from canine to second molar [Figure 2a and b]. Incisional biopsy under local anesthesia was planned, which revealed OM.

Immunohistochemical stains with neuron-specific enolase and vimentin were positive [Figures 3 and 4]. The lesion was completely excised with peripheral ostectomy through intraoral buccal vestibule approach considering the age of the patient, and Carnoy's solution was applied under general anesthesia. The patient is still under observation 4 years after surgery and there is no recurrence.

Case report 2

A 50-year-old female reported with complaint of a painless swelling in the anterior maxillary region for a period of 1-year. Initially, the swelling was small and showed a gradual increase to size of its present dimensions. The swelling extended from 13 to 22 causing displacement of teeth, exfoliation of 21, and obliteration of buccal vestibule [Figure 5].

Extraoral examination revealed diffuse swelling in the upper lip region protruding beyond it. The mucosa over the swelling was of the same color and texture as the surrounding mucosa with no local rise of temperature. On palpation, the swelling had firm consistency. Buccal and palatal cortices were expanded. The radiographic examination revealed a well demarcated multilocular radiolucent lesion with "tennis racket" appearance involving anterior maxilla. CT was performed which showed cortical distension in a buccal and lingual direction that had breached in some region. Incisional biopsy was performed and it revealed myxoma.

Taking into consideration the extent of tumor, anterior partial maxillectomy through intraoral approach using vestibular incision was done extending from 14 to 23 under general anesthesia. A surgical obturator was given to protect the wound. The final obturator was given after 6 months. Histopathological examination of excised specimen showed loose myxomatous tissue alternating with fibrous area and islands of residual bony trabeculae. The patient was followed up for 6 years and has remained disease free.

Case report 3

A 25-year-old young female reported to our department with complaint of appearance of painless lump intraorally in the left posterior region of lower jaw after extraction of left second molar, a month back in a private clinic. On examination, no extraoral swelling was present. Intraorally, a small exuberant growth protruding from the extraction socket of lower left second molar was seen, surrounding area was covered by normal appearing mucosa. A panoramic radiograph revealed the presence of a well demarcated, multilocular radiolucent lesion extending from first molar to third molar on the left side. A biopsy of the lesion was performed which revealed OM [Figure 6]. Aggressive curettage with extraction of 36 and 38 followed by peripheral ostectomy was done intraorally under local anaesthesia (LA), keeping in view the young age of female and literature showing less recurrence in mandible [Figure 7]. Histopathological examination showed unencapsulated tumor mass showing spindle- and stellate-shaped cells in loose connective tissue stroma. Odontogenic epithelial rests were not observed and are not essential for diagnosis [Figure 8]. The healing was uneventful with good amount of bone formation seen radiographically. The patient has been followed up for 7 years with no recurrence.

Literature review in Indian population

A computerized literature search using Medline and Google scholar was conducted for published articles on OM. MeSH phrase used in search were OM AND jaw; OM, maxilla AND mandible. The Boolean operator "AND" was used to combine and narrow the search. The full set of all these articles were thoroughly examined by the authors.

Most articles were case reports and retrospective case series. Results also contained various other literatures including pattern of other odontogenic lesions and case reports of OM in extra maxillofacial sites. The search was set to find the keywords in the title, abstract and text, result that appeared were of age range, site predilection, gender data, treatment pattern, and recurrence.

There was no specific article or meta-analysis which reviewed the literature regarding site, treatment pattern, and prognosis of the intervention exclusively in the Indian population. Search results were further refined by restricting search to articles published by Indian authors in the last 30 years. An additional hand search was also made to identify other published articles taking same parameters for online search.

A total of 32 cases of OM were described in 25 articles retrieved [Table 1]. Out of them, 24 myxomas were present in maxilla, only 8 were in mandible, and a single case was

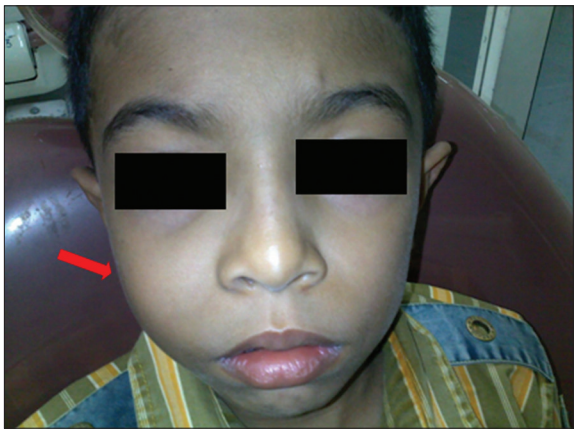

Figure 1: Extraoral photograph shows obliteration of right nasomaxillary groove and squinting of right eye

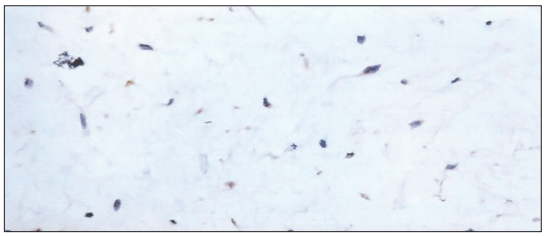

Figure 3: Photomicrograph of positive vimentin

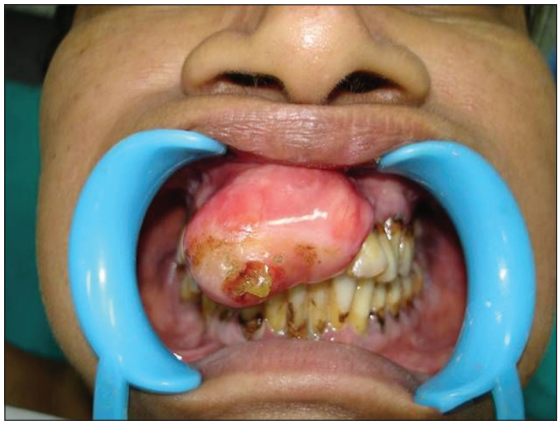

Figure 5: Intraoral view revealing buccal swelling causing exfoliation and displacement of upper anterior teeth

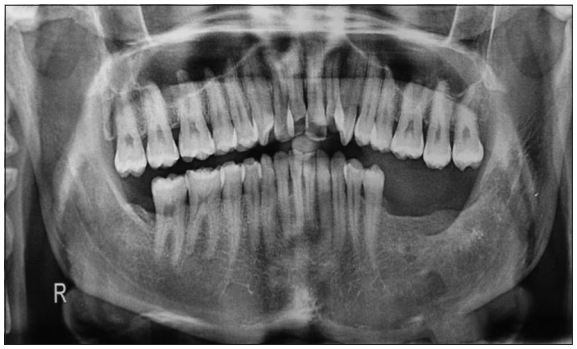

Figure 7: Panoramic radiograph of 5 years follow-up showing no recurrence

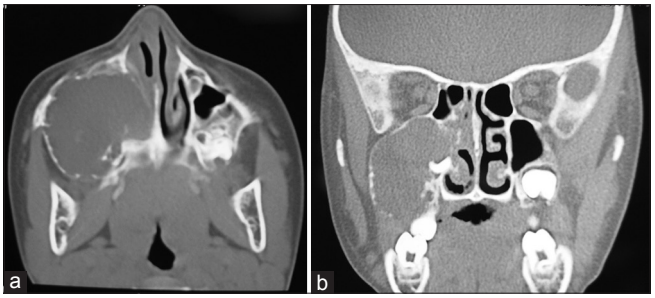

Figure 2: Computerized tomography scan. (a) Axial section – homogeneous mass obliterating the right maxillary antrum and displacing lateral nasal wall. Note the expansion of lesion in all direction. (b) Coronal section – tumor mass in contact with floor of orbit. Presence of septa can be appreciated

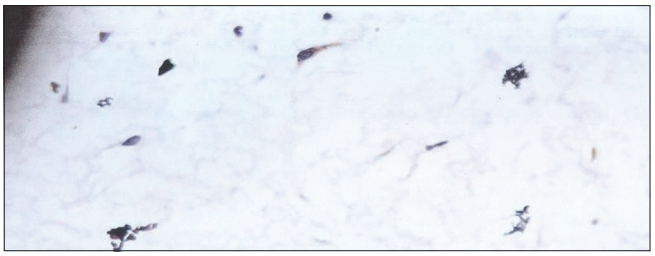

Figure 4: Photomicrograph showing positive expression of neuron-specific enolase

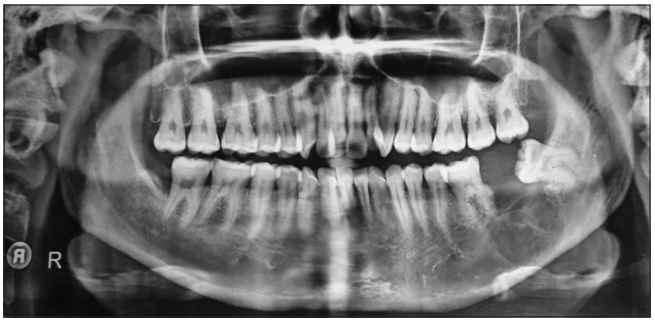

Figure 6: Panoramic radiograph showing a multilocular radiolucent area from lower left first molar to third molar

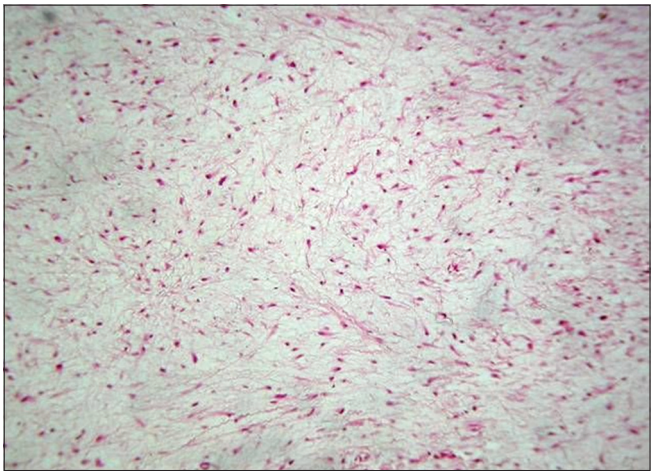

Figure 8: Tumor mass showing spindle- and stellate-shaped cells in loose, abundantly myxoid connective tissue stroma

present in supraglottic region (larynx). Average age of occurrence of OM is 25.5 years. A total of 6 patients were children ranging from 7 to 12 years of age. There were 16 males and 14 female patients, with no sex specified for two cases. Surgical procedure carried out consisted of excision and curettage in 16 patients and resection with safe margin followed by reconstruction was done in 13 patients. Reconstruction in 4 patients was done using iliac crest bone. Follow-up time was only given for 16 patients and out of these, 8 patients were followed up for 2 years or less, rest 8 patients had a follow-up time between 2 and 5 years. Out of which 2 patients who had undergone excision showed recurrence in their follow-up time of 1.5-3 years. They were successfully treated with resection and reconstruction.

Discussion

The WHO defines the myxoma (OM, myxofibroma) as a locally invasive neoplasm consisting of rounded and angular cells that lie in an abundant mucoid stroma.[1-3] Traditionally, the myxoma of the maxilla and mandible has been considered to be a neoplasm of odontogenic origin. Although the evidence is mainly circumstantial, support of an odontogenic origin has been perpetuated by (1) its almost exclusive occurrence in the tooth-bearing areas of the jaws; (2) its frequent occurrence in young individuals; (3) its common association with an unerupted tooth or a developmentally absent tooth; (4) its histological resemblance to dental mesenchyme, especially the dental papilla; and (5) the occasional presence of sparse amounts of odontogenic epithelium.[4-6]

Myxomas can occur anywhere in the jaws, but have a predilection for the molar and premolar regions of the mandible and maxilla.[1,7,8] However, Regezi and Sciubba (1993) reported equal distribution in the anterior and posterior regions of the maxilla.[8,9] In a study on bone tumors affecting the skeleton, McClure and Dahlin found only three cases of extragnathic myxomas.[10] Cortical expansion and perforation are common, and maxillary myxomas often extend into the sinus.

Kaffe *et al*. reviewed 164 OMs of the jaws and found that 75% occurred between the second and fourth decades (patient age range, 1–73 years; mean, 30). According to some authors, female: male ratio range from 1.5:1 to 4:1,[1,11,12] whereas others have found both sexes to be equally affected.[4,6] This is congruent with our cases in which two females in the range of second and fourth decade were affected and third case was a seven year old boy. It is believed to be uncommon in childhood with only about 7% occurring in the first decade.[10] There were 109 (66%) neoplasms in the mandible and 55 (34%) in the maxilla.[7]

The radiographic features of the OM are variable, ranging from small unilocular lesions to large multilocular neoplasms, which often displace teeth or less frequently resorb roots.[12,13,14] The multilocular trabecular pattern has been described as "honey comb," "soap bubble," "wispy," "spiderweb," and "tennis racket."[7,14,15] Most multilocular myxomas are > 4.0 cm; unilocular myxomas tend to be smaller.[7,11] In radiographic analysis of 21 cases, Peltola *et al*. found that multilocular were mostly located in the anterior and posterior region of jaws.[7] Only 5% of myxomas are associated with unerupted teeth.[11] It is also noted that lesion borders which have been found to be poorly defined or diffuse more often in maxillary lesions than mandibular lesions are better delineated on the cone-beam CT.[11,16] MacDonald-Jankowski *et al*.[17] related that CT was more likely to display a cortex perforation and pattern of septa into the lesion. Defining the tumor borders is essential for planning the extent of resection as the tumor is associated with high recurrence rate. Sumi *et al*.[18] described magnetic resonance imaging (MRI) images of OM as well defined, well enhanced mass lesion with high intensity on T2-weighted images (T2-WIs) and low intensity on T1-WI. However, Kawai *et al*.[19] reported homogeneous signal intensity on every pulse sequence. The lesion showed intermediate signal intensity on the T1-WI and T2-WIs, some discrepancy was seen in maxillary myxoma which he related to the viscosity of the mucoid substance or the protein density of the tumor.[19]

Histologically, the myxoma is bland in appearance and is composed of loosely arranged, evenly dispersed spindle-shaped, rounded, and stellate cells with a lightly eosinophilic cytoplasm in a mucoid rich (myxoid), intercellular matrix.[20,21]

Many stellate tumor cells have anastomosing, long tapering, and cytoplasmic processes. Although some degree of mild nuclear pleomorphism or hyperchromatism may exist, including an occasional mitosis or binucleate cell, there is no correlation with recurrence of the neoplasm.[20]

Myxomas are unencapsulated and pervade surrounding bone by expansion rather than cellular growth, possibly as a result of the large content of hyaluronic acid.[21]

Ultrastructural or immunohistochemical studies have shed some light on the histogenesis of the myxoma and the characteristics of the cells and matrix. In 2000, Jaeger *et al*.[22] reported the following findings in their literature review: (1) The tumor cells are secretory: (2) The cells are mesenchymal and express vimentin; (3) Positivity to S-100 protein and muscle-specific Actin has been variable; and (4) the matrix exhibits different proteins, mostly collagen types I and VI, tenascin, fibronectin, and proteoglycans. Ultrastructural examination of our case confirms the mesenchymal origin of the myxoma cells [Table 2]. Our findings were consistent with the results reported by Lombardi e*t al.* and Green e*t al*. with respect to vimentin.[23-25]

The treatment of OM varies from simple enucleation and curettage to more extensive radical surgical owing to

Table 1: All the 25 reviewed articles of odontogenic myxoma

Title	Author	Year	Number of case	Site	Age/sex	Treatment done	Follow-up	Recurrence
Maxillary odontogenic myxoma: A diagnostic pitfall on aspiration cytology	Kumar N, Jain S, Gupta S	2002	One	Right maxillary	23 years/male	Excision	16 month	No recurrence
Odontogenic myxoma of maxillary	Sivakumar, *et al*.	2008	One	Right posterior maxillary	30-year-old male	Partial maxillectomy		
Aspiration cytology of maxillary myxoma	Anand Kumar *et al*.	1993		Maxillary				
Fibro-myxoma of the maxillary	Prasad and Sharan	1983	One	Right maxillary	14 years/male	Excision	No follow-up	
Laryngeal myxoma	Baruah *et al*.	2001	One	Supraglottic region (larynx)	57/male	Excision	8 months	No recurrence
Fibromyxoma maxillary	Anupam Mishra, Naresh Bhatia, G. K. Shukla	2004	One	Right maxillary	30/female	Curettage	2 years	No recurrence
Fibromyxoma of maxillary	Prem Kakar and Sood	1969	One	Left maxillary	12 years/male	Curettage	No follow-up	
Myxoma of the jaw	Chatterji *et al*.	1985	3 cases	Right maxillary Right body of mandible	25 years/female 17 years/male 50 years/male	Excision Excision Segmental resection	No follow-up	
Odontogenic myxoma of the mandible	Manne *et al*.	2012	1 case	Right body of mandible	19 years/male	Segmental resection and reconstruction with microvascular iliac	30 months	No recurrence
Odontogenic myxoma of the maxillary: A report of rare case and review on histogenetic and diagnostic concepts	Shah *et al*.	2011	1 case	Right maxillary	37/female	Partial en block resection	6 months	
Recurrent odontogenic myxoma of maxillary: A diagnostic and operative dilemma	Jaswal	2008	1 case	Maxillary	32 years/male	Partial maxillectomy		
Odontogenic myxoma of the maxillary: A report of rare case and review of the literature	Singaraja	2010	1 case	Maxillary	7 years/male			
Odontogenic myxoma in a child: Diagnostic and treatment dilemmas	Sarode	2002		Mandible	10 years/male	Surgical excision of tumor mass with peripheral ostectomy		
Odontogenic myxoma: Report of two cases	Reddy *et al*.	2010	2 cases	Right postmaxillary Left side postmandible	25/male 26/female	Surgical excision with curettage Hemimandibulectomy and reconstruction with iliac crest graft	2 years follow-up	
Recurrent odontogenic myxofibroma of the mandible in a 12-year-old child: An illustrative case report	Mehrotra	2008	1 case	Ant mandible	12 years/male	Resection with continuity defect and reconstruction with iliac crest graft	3 years	No

contd...

Table 1: Contd...

Title	Author	Year	Number of case	Site	Age/sex	Treatment done	Follow-up	Recurrence
Peripheral myxoma of maxillary. A case report	Ramaraj	2003	1 case	Maxillary	35 years/female	Semi radical approach		
Odontogenic myxoma of the mandible: A case report	Sharma	2003	1	Mandible	23 years/female	*En bloc* resection and reconstruction with iliac graft	4 year	No
Myxoma of the jaw - a clinic-pathological study	Sooknundum	1986	5 case 4 cases 1 case	Maxillary Mandible	11-36 years/ female	Curettage	1.5-3 years	Recurrence in 2 patient followed by hemimandibulectomy and curettage in maxillary
Fibromyxoma of maxillary	Kabir	1985	1 case	Right maxillary	40 years/male	Total maxillectomy on right side		
Odontogenic myxoma of maxillary: A case report	Gowda	2011	1 case	Left maxillary on palate	25/female	Curettage	4 months	No
Odontogenic fibromyxoma of maxillary: A case report	Ishita Gupta	2010	1 case	Left premolar maxillary	30 years/female	Segmental maxillectomy with safe margins		
Maxillary odontogenic myxoma: A rarity	Meghanand T Nayak	2011	1 case	Right maxillary	11 years/male	En bloc resection of maxillary		
Om a case report	Preeti Sethi	2011		Right maxillary	25 years/male	Lesion along with the associated bone resected	5 months	
Histological spectrum of myxoma	Monika Mendiratta	2012	1 case	Right mandible	33 years/male	Excision		
Exophytic growth	Rameshwari Singhal	2012		Left maxillary	19 years/female	Excision	6 months	

high recurrence rate around 25% seen typically during the first 2 years after removal.[23] Pahl *et al*. attributed recurrence to the insidious local invasion into cancellous bone beyond radiographically visible margins and absence of encapsulation.[26] Localization of tumor, particularly in pediatric population is an important factor in determining the excision's extent to follow a less radical approach in pediatric population to limit transfacial access, surgical morbidity, and potential growth disturbance. This is supported by Rotenberg *et al*.[27] who suggested conservative resection (narrow resection margin or 1 clear tissue plane) for myxomas of the pediatric maxilla. Furthermore, King *et al*. recommended the use of liquid nitrogen cryotherapy as an adjunct procedure in pediatric patients.[28]

Conclusion

OM is a rare odontogenic tumor with inconclusive clinical and radiographic features, hence histopathological examination is mandatory to confirm its diagnosis. It was seen that although the immunohistochemical analysis may help in diagnosis, it does not provide surgeon any information which can guide in treatment planning or can predict the rate of recurrence.

Table 2: Expression of vimentin and NSE in all three cases

	Case 1	Case 2	Case 3
Vimentin	+	+	+
NSE	+	+	+

NSE: Neuron-specific enolase

However, detailed examination of MRI and CT scans of patients with OM is needed so as to make it helpful in better and effective management of OM since the lesion presents a high potential for recurrence.

Currently, we lack data on the number of reported cases of OM in Indian population as author feels more number of patients must have been treated then reported. In addition, there is deficiency of information regarding recurrence rates in OMs, especially concerning different surgical approaches.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published

and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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