Intraosseous pneumatocysts of the cervical spine: a report of four cases and review of literature

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Pneumatocysts are benign, gas-containing lesions, most commonly observed affecting the vertebrae, sacrum, and ileum. Most often, these lesions are asymptomatic and are detected incidentally during imaging examinations. Although once believed to be a rare lesion, recent studies suggest that it is more common than previously thought. We present four cases of pneumatocysts affecting the cervical vertebrae detected as incidental findings on cone beam computed tomography (CBCT). Given the increased use of CBCT in dentistry to evaluate the maxillofacial structures, it is likely that dentists will encounter this lesion on CBCT scans that encompass the superior cervical spine. Recognition of the pathognomonic features of this benign, innocuous lesion is important to avoid unnecessary investigations and causing alarm to the patient. We also present a comprehensive review of the literature on the demographic characteristics and clinical presentation of this relatively unknown lesion. (Oral Surg Oral Med Oral Pathol Oral Radiol 2015;119:e49-e54)

Cone beam computed tomography (CBCT) is used to image the dentomaxillofacial region for a variety of diagnostic tasks. Often, a region of the cervical spine is encompassed within the image volume, especially in medium−field-of-view (FOV) and large-FOV scans. Incidental findings noted on such CBCT scans affecting the cervical vertebrae include, but are not limited to, osteoarthritic changes, fusion and nonsegmentation of the vertebral bodies, and the presence of surgical hardware.[1]

Thus, dentists should recognize the presence of abnormalities in the cervical spine and, where appropriate, seek consultation or refer the patient to an appropriate provider for further management.

Intraosseous gas collection is considered an uncommon finding affecting the vertebral bodies.[2,3] It has been associated with various pathologic conditions that include gas-forming osteomyelitis, osteonecrosis, trauma, and surgery.[4] However, an innocuous and relatively more common cause of intraosseous gas collection in the vertebral bodies is the intraosseous pneumatocyst.[5] This is a relatively unknown cystic lesion that frequently affects the sacroiliac joint but is also observed within the cervical vertebrae. Manifestation in the cervical spine has been reported as uncommon,[6] with only 21 cases reported. However, a recent study suggests that this lesion is more common than previously believed,[7] which underscores the need for dentists to recognize this abnormality. We present four cases of intravertebral pneumatocysts, selected from our radiology archive. The record review to select cases was approved by the University of California Los Angeles (UCLA) Institutional Review Board. All four cases were detected as incidental findings on CBCT scans. To our knowledge, this represents the first report of this pathologic entity detected on CBCT scans acquired for dentomaxillofacial diagnosis.

CASE 1

A large-FOV CBCT scan of a 59-year-old male was submitted to our service for a radiologic consultation. The scan had been acquired to evaluate the posterior left mandible for implant placement. A corticated radiolucency with the attenuation of air was noted adjacent to the inferior border of the C3 vertebral body (Figure 1). The inferior endplate of the C3 vertebral body demonstrated thinned but intact cortical borders. Trabecular bone surrounding the lesion was slightly sclerotic. Degenerative changes were noted affecting the adjacent vertebral bodies, including sclerosis of the dens and osteophyte formation at the inferior aspect of C1.

CASE 2

A large-FOV CBCT scan was sent to our service for a radiologic evaluation. The patient was a partially edentulous 67-year-old woman, who was being

Statement of Clinical Relevance

The pathognomonic radiological features of vertebral pneumatocysts are presented, and their differentiation from other lesions described to allow clinicians to correctly recognize this entity.

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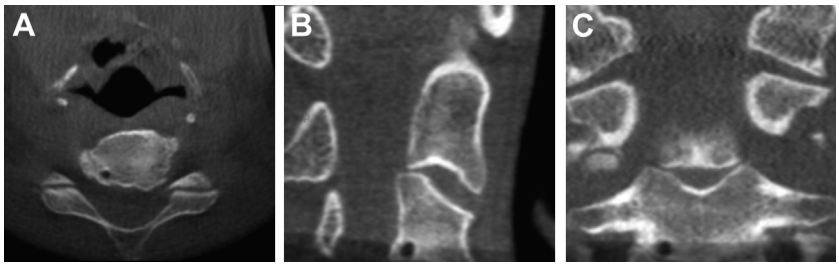

Fig. 1. Case 1. Axial (A), sagittal (B) and coronal (C) CBCT sections illustrating a well defined corticated radiolucency in the C3 vertebral body, with the attenuation characteristics of air.

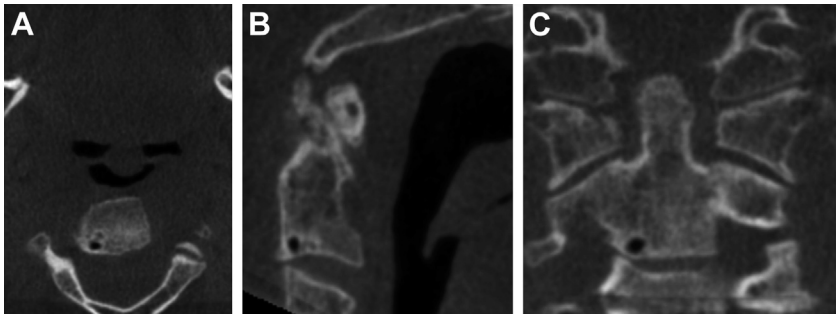

Fig. 2. Case 2. Axial (A), sagittal (B) and coronal (C) CBCT sections demonstrating a well defined radiolucency, with the attenuation of air, at the inferior margin of the C2 vertebral body, Note degenerative changes in both C1 and C2.

evaluated for potential implant placement. CBCT revealed a corticated radiolucency with the attenuation of air at the posterolateral aspect of the C2 vertebral body, immediately adjacent to its inferior border (Figure 2). The inferior endplate of the C2 vertebral body was thinned but intact. The surrounding trabecular bone was mildly sclerotic. The C1 and C2 vertebral bodies demonstrated degenerative changes, including large osteophyte formation at the inferior aspect of the anterior arch of the atlas.

CASE 3

A 60-year-old woman with left-sided mandibular hypoplasia and osteoarthritis of the left temporomandibular joint (TMJ) was evaluated for left-sided TMJ pain. A large-FOV CBCT scan showed a corticated radiolucency at the posterolateral aspect of the C3 vertebral body, immediately adjacent to its inferior border (Figure 3). Internally, the lesion mostly demonstrated an attenuation of air. However, the most inferior aspect of the lesion demonstrated attenuation characteristics of soft tissue. In this area, the inferior endplate of the C3 vertebral body was thinned but intact. The surrounding trabecular bone was of normal density and architecture. No osteophyte or cortical erosions of the adjacent vertebral bodies were observed. The intervertebral joint space between C3 and C4 was narrowed.

CASE 4

A 55-year-old woman with partial edentulism was evaluated for potential implant placement. A large-FOV CBCT volumetric scan showed multiple corticated radiolucencies, with attenuation of air, in the C4-C6 vertebral bodies (Figure 4). The lesions were localized at the endplates of the vertebral bodies, with significant thinning and probable disruption of the cortical border. The largest of the lesions was noted at the posterolateral aspect of the C4 vertebral body, immediately adjacent to its superior endplate. One of the lesions at the superior aspect of C4 appeared to communicate with the intervertebral joint space. Trabecular bone around some of the lesions was sclerotic. Extensive osteoarthritic changes affecting all of the imaged cervical vertebrae were noted, including large osteophytes at the anterior aspect of the C3-C5 vertebral bodies. Significant narrowing of the intervertebral joint space was seen between C5 and C6.

Review of literature

We reviewed the English language literature for previous reports of pneumatocysts of the cervical spine. We conducted a PubMed search using the search term "pneumatocyst." The resulting 30 papers were reviewed in detail. Papers reporting pneumatocysts occurring in the cervical spine were examined in more detail for clinical manifestations, radiographic features, and management. These papers are listed in Table I. In total, 17 individual case reports or case series describing cervical intraosseous pneumatocysts were reviewed. These articles were published from 1988 to 2013, mostly in journals of radiology, and yielded a total of 21 cases of cervical intraosseous pneumatocysts, to which we now add four additional cases.

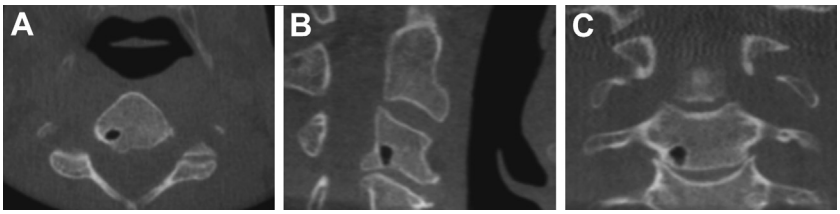

Fig. 3. Case 3. Axial (A), sagittal (B) and coronal (C) CBCT sections demonstrating a radiolucency at the inferior margin of the C3 vertebral body, Note thinning of the inferior margin of the vertebral body.

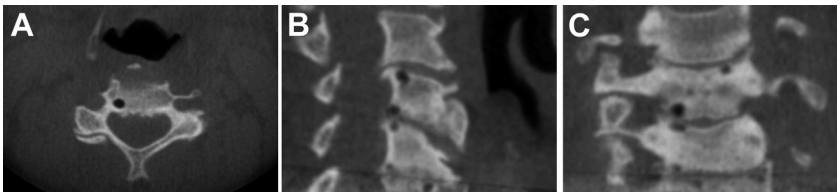

Fig. 4. Case 4. Axial (A) CBCT section illustrating a well-defined radiolucency at the posterolateral aspect of the C3 vertebral body. Sagittal (B) and coronal (C) CBCT sections demonstrating multiple radiolucencies in the vertebral bodies of C3, C4, and C5.

Only two studies examined the incidence of cervical vertebral pneumatocysts.[5,7] Of these, the study by Arslan et al. examined pneumatocysts in the entire spine, and the C-spine cases are reported in Table I. The study by Matsukubo et al.[7] was more extensive and analyzed cervical vertebrae exclusively. The latter study examined computed tomography (CT) scans of 500 patients, acquired for a variety of indications. They detected pneumatocysts in 213 patients. The findings of this study are summarized in Table II.

Among the 21 reported cases, the mean age of the patients was 57.3 years, with a range of 43 to 89 years. The ages of the patients in our four cases were also within this range (see Table I). In the Matsukubo et al. study,[7] the mean age of patients with intravertebral pneumatocysts was 63.4 years, with an age range reported as extending from the 40s to the 80s. Importantly, their study demonstrated an increase in incidence with age. Pneumatocysts were detected in 8% (7/86) of patients in their 40s or younger, whereas the incidence was 60% (40/66) in patients older than 80 years.

Of the total 21 cases of cervical vertebral pneumatocysts, 10 occurred in males and 9 in females. Gender information was not provided in two cases (see Table I). Gender information was also not provided in the study from Matsukubo et al.[7] Our cases presented here included three female patients and one male patient. Thus, based on currently available information, there does not appear to be an overt gender predilection.

Only 9 of the 21 reported cases specifically commented on communication with the intervertebral joint space—which was present in 2 of the 9 cases (22%) and absent in 7 of the 9 (78%) cases. One of our four cases showed a communication with the joint space. Interestingly, this finding was considerably higher in the Matsukubo et al. study (275/518 [53%]).[7] A high incidence of joint space communication is suggestive of an external origin of gas within the vertebral body. Given the importance of this finding from an etiologic standpoint, the discrepancy is noteworthy. It may be at least partially explained by the thinner reconstruction of the axial images. The Matsukubo et al.[7] study used axial slices of 1 mm thickness, whereas some of the cases reports here used 2- or 3-mm axial slices.[5] Thicker axial slices could have potentially obscured small discontinuities in the vertebral cortex due to superimposition of adjacent anatomy, resulting in fewer reports of joint space communication. In comparing these studies, it is also important to consider differences in reader sensitivities, especially as the radiographic manifestations of the lesion are better described.

Seventeen of the 21 cases (80%) case reports showed radiographic evidence of coexisting degenerative changes in the cervical spine (Table I). The remaining four cases did not provide any information regarding osteoarthritis. Associated degenerative changes of the cervical spine were reported at a similar frequency in the Matsukubo et al. study[7] (69%, n = 213). Three of our four cases showed degenerative changes in the cervical spine.

The majority of the pneumatocysts described occurred in the lower C-spine, both in the individual case reports as well as in the case series. The most frequently reported locations of the intravertebral pneumatocyst among case reports were C5 (48%) and C6 (24%). The same locations were also the most common sites in the Matsukubo et al. study,[7] C5 (26%) and C6 (35%), with slightly different frequencies reported. Three of our four cases were in the C2/C3. However, this most probably reflects the extent of C-spine coverage on the CBCT scans.

Table I. Characteristics of the 21 published cases of intraosseous pneumatocysts in the C-spine (the four cases from the current report are listed at the bottom of the table)

Site of IVP	*Age*	*Gender*	*Size* (mm)*	*Coexisting degenerative changes in the C-spine**	*Communication with joint space**	*Internal density on CT (HU)**	*Neck pain**	*Fluid**
C6[14]	NR	NR		Y				
C7[12]	52	M	10		Y	−850		
C5[22]	62	M		Y		−700		
C5[2]	63	F		Y	N			
C5[3]	57	M		Y				Y
C4/C5[5]	56	M		Y	N	−87 to −422		
C5[18]	56	F	6	Y	N	−890		
C5-C7[20]	69	F	7	Y	Y	−890		Y
C7[10]	58	F		Y	N	−580 to −950		
C6[10]	89	F		Y		−580 to −950		
C4[10]	53	F	10	Y		−580 to −950		Y
C5[10]	46	M		Y		−580 to −950		
C7[10]	43	M				−580 to −950		
C4[15]	65	M	12	Y		−890		
C5-C6[6]	55	M	6	Y		−890		
C6-C7[17]	48	F	8	Y	N	−900	Y	
C7[27]	51	F	13	Y			Y	
C5[21]	56	F	12	Y			Y	N
C5-C6[25]	59	M	9		N	−559	N	
C4[4]	51	M	10	Y	N	−970	Y	
C[26]	NR	NR						
Cases from current report								
C3	59	M	3.5	Y	N	NA	NR	N
C2	67	F	3	Y	N	NA	NR	N
C3	60	F	4.5	N	N	NA	NR	N
C4-C6	55	F	5	Y	Y	NA	NR	N

NR, not reported; *NA*, not applicable.
* Information on size and other features are listed where reported.

Table II. Characteristics of the 500 patients of the Matsukubo et al. study[7]

Patients with intravertebral pneumatocyst (IVP)	213
No. of patients with C-spine degenerative changes	148
Total no. of IVP	518
No. of IVP communicating with the joint space	275
Location	
C1	0
C2	10
C3	36
C4	73
C5	135
C6	181
C7	83

DISCUSSION

The pneumatocyst was first described as a pathologic entity by Weinberg and Schneider in 1982[8] and later named by Ramirez in 1984.[9] It is a benign gas-containing lesion, which most often affects the sacroiliac joint, and is less commonly observed in the spinal vertebrae.[10] Isolated cases of intraosseous pneumatocyst affecting other bones, such as the scapula, humerus, and clavicle, have also been reported.[11]

The pathogenesis of a pneumatocyst is not fully understood. One theory is that gas accumulation in the intervertebral disk, known as the *vacuum phenomenon*,[12,13] extends into the vertebral body through a defect in a degenerated end plate.[11,14] This hypothesis is consistent with the higher prevalence of these lesions in the older population, and its coexistence with degenerative joint disease. However, communication with the joint space is not radiographically observed in all cases. Microscopic communications not detected on CT might be present. The hypothesis seems less plausible in the case of lesions that occur in vertebral bodies with no evidence of local degenerative changes. Other proposed etiologies include spontaneous gas accumulation and secondary gas accumulation in a simple fluid-filled cyst.[15]

The most common cervical location for the intravertebral pneumatocyst is the lower cervical spine, namely, the C5-C6 vertebral bodies. This was reflected both in case reports as well as in the larger Matsukubo et al.[7] study. Being the site of lordosis inversion, the C5-C6 vertebral bodies may be more susceptible to degenerative changes. Outside of the cervical spine, the pneumatocyst is known to occur most frequently in the region of the sacroiliac joint.[16] Here, too, lesions are often accompanied by localized degenerative changes,

but less frequently than is seen in the cervical vertebrae.[11]

Intravertebral pneumatocysts typically do not cause any symptoms and are usually detected as incidental findings on CT imaging.[5,7] Although some patients do report pain in the region, the origin of the pain is often obscured by the coexistence of degenerative changes in the area.[6,17]

Until recently, the lesion has been described as rare, especially in the cervical spine.[15,18] However, two retrospective CT studies[5,7] have shown that the lesion is not as rare as once believed. One study[5] examined the cervical spine on CT scans from 89 patients and reported a prevalence of 9%. A second and larger study[7] of 500 CT scans showed a prevalence of 42%. The reason for the considerable difference between the two studies is not known but may reflect age differences in the two sample populations studied. The higher prevalence in the more recent reports may also reflect increasing recognition and knowledge of this lesion. Given the prevalence reported in the two recent studies, it is highly likely that dentists will encounter this lesion on CBCT scans that encompass the C-spine.

On CT and CBCT imaging, the pneumatocyst typically presents as a small, well-circumscribed, radiolucent lesion delineated by a sclerotic rim.[5,19] Communication with the intervertebral joint space and localized degenerative changes may be present.[16] On plain radiographs, the lesion has been mistaken for more invasive lesions, such as metastasis or multiple myeloma.[18,20] However, the gas-attenuating nature of the lesion on CT and CBCT is pathognomonic.[21] On magnetic resonance imaging (MRI), the presentation is less specific.[10] The lesion demonstrates hypointense signals on both T1- and T2-weighted images, without gadolinium enhancement.[18] Notably, bone lesions, such as enostosis, fibrous dysplasia, and blastic metastasis, also demonstrate similar MRI appearances.[10]

On CT and CBCT scans, the radiographic differential diagnosis may include degenerative changes, subchondral cysts, gas-forming osteomyelitis, simple bone cysts, and Schmorl nodes.[11,22] Subchondral cysts are areas of subarticular degeneration and contain granulation tissue. Similar to pneumatocysts, these lesions are typically accompanied by other signs of osteoarthritis, such as osteophytes and cortical erosions. Although radiolucent, these lesions have the attenuation characteristics of soft tissue features, in contrast to the air-attenuation features of pneumatocysts, which allows distinction of these two lesions. The term *Schmorl node* refers to a degenerative phenomenon in which protrusions of the intervertebral disk occur through the vertebral endplate and into the vertebral body.[23,24] The air-attenuation characteristics and regular borders are pathognomonic imaging features of the pneumatocyst and are usually sufficient to exclude these other diagnoses.

The natural course of the intravertebral pneumatocyst has been examined. In one report,[21] a lesion in the C5 vertebrae was monitored with serial CT imaging. Over the course of 3 years, no significant change in the radiographic appearance of lesion was evident. Two other case studies,[20,25] however, report progressive enlargement of a pneumatocyst when monitored over a period of weeks or months. In one study,[18] a pneumatocyst in the C5 vertebrae was examined over a 40-week period with serial imaging. At 14 weeks, the gas-attenuation characteristics had changed to a fluid-filled cyst. At 40 weeks, MRI showed contrast enhancement of the lesion, suggestive of the presence of granulation tissue. Despite these reports, the natural course of the intravertebral pneumatocyst remains unclear—the lesions may enlarge, remain stable, or even resolve into fluid-filled cysts.

Treatment is typically not required for the intravertebral pneumatocyst.[3,26] Sometimes, the lesions become large enough to occupy most of the vertebral body, in which case the possibility of vertebral fracture may be a concern.[27] In such cases, surgical intervention has been proposed.[21] At present, however, there are no documented cases of vertebral surgery to treat a pneumatocyst or a pathologic fracture at the site of a pre-existing lesion.

CONCLUSION

We have presented here four cases of intravertebral pneumatocysts, detected as incidental findings on CBCT scans obtained for dentomaxillofacial diagnostic indications. To our knowledge, this is the first description of these lesions observed on CBCT scans. This is also the first time the lesion has been reported in the dental literature. Familiarity with the demographic and radiographic characteristics of this lesion is of importance to dentists and oral and maxillofacial radiologists to avoid unnecessary investigation of this benign, innocuous lesion and to avoid causing alarm to the patients. Treatment of these lesions is not required. However, given the risk of enlargement of the lesion, follow-up radiographic examination is prudent.

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