The olfactory cleft localization of REAH seems to be more frequent than it was thought before. 4.5 Therefore, it is mandatory to think about this entity in all cases of diffuse nasal polyposis when, because of polypous tissue masses, it is not possible to distinguish where does the suspected mass come from: between the middle turbinate and the nasal septum or from the ostiomeatal complex proper. This is the main point to discover the real background of the disease. Moreover, misinterpretation of the REAH as chronic sinus inflammation and diffuse polyposis may result in an inappropriate treatment.

In our case, the preoperative situation strongly suggested esthesineuroblastoma, not polyposis at all. Because of the opacity between middle turbinate and nasal septum, we thought also about REAH in particular because we knew that Athre and Ducic⁹ found that REAH could be a locally aggressive process! They represent the patient with frontal sinus hamartoma with expansion to the posterior right orbital wall. The patient was cured with a frontal craniotomy and cranialization. This is exactly why we decided to remove the entire tumor, including both olfactory bulbs.

Regardless of the localization, the treatment of choice for hamartomas is complete local excision of the tumefaction. Conservative surgical resection seems to be curative, with no recurrences reported in variable follow-up periods. 4,9-12 In comparison with this, our surgical treatment was really a radical one.

CONCLUSIONS

Hamartomas of the nasal cavity and paranasal sinuses are rare. These lesions are characterized by an abnormal overgrowth of tissue elements indigenous to a particular area of the body. Hamartomas do not have unlimited growth potential nor do they have metastatic potential. Extensive progression of the disease to involve the orbit and intracranial vault is exceedingly rare but possible.

Conservative surgical resection of hamartomas is curative, and the literature describes no instances of recurrent, persistent, progressive, or metastatic disease.

Our case denies some literature statements regarding the progressiveness of this tumefaction because it was found to penetrate from the olfactory clefts to the endocranium. To our knowledge, our case is the first one reported with extension of the olfactory cleft REAH to the endocranium.

REFERENCES

- Albrecht E. Uber Hamartome. Verh Dtsch Ges Pathol 1904;7:153–157
- Delbrouck C, Aguilar SF, Choufan G, et al. Respiratory epithelial adenomatoid hamartoma associated with nasal polyposis. Am J Otolarvngol 2004;25:282–284
- Wenig BM, Heffner DK. Respiratory epithelial adenomatoid hamartomas of the sinonasal tract and nasopharynx: a clinicopathologic study of 31 cases. Ann Otol Rhinol Laryngol 1995;104:639–645
- Lima NB, Jankowski R, Georgel T, et al. Respiratory epithelial adenomatoid hamartoma must be suspected on CT-scan enlargement of the olfactory clefts. *Rhinology* 2006;44:264–269
- Cao ZW, GU ZW, Yang J, et al. Respiratory epithelial adenomatoid hamartoma of bilateral olfactory clefts associated with nasal polyposis: three cases report and literature review. *Auris Nasus Larynx* 2010;37:352–356
- Liang J, O'Malley BW Jr, Feldman M, et al. A case of respiratory epithelial adenomatoid hamartoma. Am J Otolaryngol 2007;27:277–279
- Mortuaire G, Pasquesoone X, Leroy X, et al. Respiratory epithelial adenomatoid hamartoma of the sinonasal tract. Eur Arch Otorhinolaryngol 2007;264:451–453

- Di Carlo R, Rinaldi R, Ottaviano G, et al. Respiratory epithelial adenomatoid hamartoma of the maxillary sinus: case report. *Acta Otorhinolaryngol Ital* 2006;26:225–227
- Athre R, Ducic Y. Frontal sinus hamartomas. Am J Otolaryngol 2005;26:419–421
- Endo R, Matsuda H, Takahashi M, et al. Respiratory epithelial adenomatoid hamartoma in the nasal cavity. Acta Otolaryngol 2002:122:398–400
- Himi Y, Yoshizaki T, Sato K, et al. Respiratory epithelial adenomatoid hamartoma of the maxillary sinus. *J Laryngol Otol* 2002;116:317–318
- Metselaar RM, Stel HV, Van der Baan S. Respiratory epithelial adenomatoid hamartoma in the nasopharynx. *J Laryngol Otol* 2005;119:476–478

Transient Cardiac Failure Due to Takotsubo Cardiomyopathy After Surgical Reduction of Nasal Fracture

Matteo Brucoli, MD, Francesco Arcuri, MD, Mariangela Giarda, MD, Arnaldo Benech, MD, PhD

Introduction: Takotsubo syndrome, also known as ampulla cardiomyopathy, broken heart syndrome, idiopathic apical ballooning syndrome, and stress-induced myocardial stunning, has been first described by Japanese authors in 1996 and subsequently specified in 2001; it derives from the resemblance between the ancient round-bottomed, narrow-necked Japanese fishing pots used to trap octopus in Asia and the end-systolic appearance of the left ventricle on ventriculography.

Clinical Report: We introduce the case of a woman who was involved in a traffic car crash and, subsequently, was admitted to the Maxillo-Facial Unit of the Novara Major Hospital with a diagnosis of nasal fracture. She underwent general anesthesia for the reduction of the fracture; after surgery, she developed acute chest pain, elevated cardiac biomarkers, ischemic electrocardiogram changes, and transient akinesis of the left ventricle without significant epicardial coronary artery disease. A diagnosis of takotsubo syndrome was made. Conclusions: This syndrome, which presents the same clinical features of a ventricular failure, is probably underdiagnosed, but after the introduction of sophisticated cardiac imaging and coronary intervention, more cases are identified and an unnecessary

From the Department of Maxillo-Facial Surgery, Azienda Ospedaliera Maggiore della Carità, University of Piemonte Orientale "Amedeo Avogadro," Novara, Italy.

thrombolytic therapy can be spared. This reversible condition, which

Received September 1, 2010.

Accepted for publication September 17, 2010.

Address correspondence and reprint requests to Francesco Arcuri, MD, S.C.D.U. di Chirurgia Maxillo-Facciale, Ospedale Maggiore della Carità, Corso Mazzini 18, 28100 Novara, Italy; E-mail: fraarcuri@libero.it

The authors report no conflicts of interest. Copyright © 2011 by Mutaz B. Habal, MD ISSN: 1049-2275

DOI: 10.1097/SCS.0b013e31822ea720

© 2011 Mutaz B. Habal, MD 1907

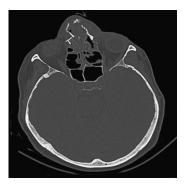


FIGURE 1. Axial computed tomographic scan demonstrating the nasal fracture.

is, to our knowledge, never mentioned after a craniomaxillofacial surgical procedure, should be considered in the diagnostic algorithm for all patients presenting with acute onset of chest pain, elevated cardiac biomarkers, and ischemic changes on the electrocardiogram after a general anesthesia.

Key Words: General anesthesia, takotsubo syndrome, nasal fracture

Takotsubo syndrome, also known as ampulla cardiomyopathy, broken heart syndrome, idiopathic apical ballooning syndrome, and stress-induced myocardial stunning, has been first described by Japanese authors¹ in 1996 and subsequently specified in 2001²; although most cases have been initially described in this country, nowadays there are reports from other parts of the world.^{5,6}

The name of this syndrome (*tako* means octopus and *tsubo* means pot) derives from the resemblance between the ancient round-bottomed, narrow-necked Japanese fishing pots used to trap octopus in Asia and the end-systolic appearance of the left ventricle on ventriculography.

This condition is characterized by a combination of the following: (1) acute psychologic/physical stress before the onset of chest pain, 2) disproportionately low release of cardiac enzymes with respect to the degree of the left ventricular dysfunction, 3) ischemic changes on the electrocardiogram (ECG), 4) no significant epicardial coronary artery disease, 5) transient akinesis or dyskinesia of the left ventricle, 6) apical ballooning with basal hyperkinesis on the left

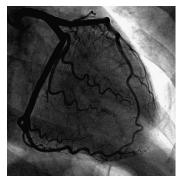


FIGURE 3. Diagnostic coronary angiogram demonstrating normal epicardial coronary arteries.

ventriculogram, 7) rapid resolution of the cardiac dysfunc tion, and 8) absence of recent head trauma, intracranial bleeding, pheochromocytoma, myocarditis, and hypertrophic cardiomyopathy.⁷

We introduce the case of a takotsubo syndrome in a patient who underwent general anesthesia for the reduction of a nasal fracture.

CLINICAL REPORT

On April 27, 2010, a 57-year-old white woman (American Society of Anesthesiologists status II, 56 kg) was involved a traffic car crash and, subsequently, was admitted to the Maxillo-Facial Unit of the Novara Major Hospital with a diagnosis of nasal fracture (Fig. 1). She had a history of hypertension, hepatitis, and bronchial asthma. She had no known drug allergies, and results of the preoperative laboratory tests were within the reference ranges; preoperative blood pressure and heart rate were 140/80 mm Hg and 74 beats/min, respectively.

General anesthesia was induced with propofol, and after the administration of rocuronium, the trachea was intubated with a 7-mm endotracheal tube; anesthesia was maintained with sevoflurane and fentanyl. Intraoperative monitoring consisted of ECG, noninvasive blood pressure measurement, and pulse oximetry. After a successful surgical reduction of the nasal fracture, the patient was extubated, but 2 hours after the end of the surgical procedure, she presented acute chest pain and hypertension; both conditions were rapidly controlled by esmolol and 2 consecutive sublingual administrations of nitroglycerine. Serial 12-lead ECG recordings demonstrated evolving anterolateral T wave inversion; postoperative cardiac enzymes

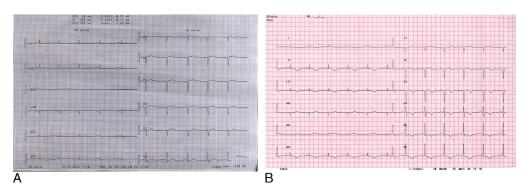


FIGURE 2. A and B, A 12-lead ECG showing evolution of widespread ST/T wave changes throughout the anterior cheat leads without Q waves including dynamic ST elevation (A) and T wave inversion (B).

1908 © 2011 Mutaz B. Habal, MD

showed elevation of troponin I and creatine kinase MB (1.76 and 8 ng/mL, respectively; reference, <0.01 and <5.00 ng/mL, respectively; Fig. 2). A transthoracic echocardiogram demonstrated severely impaired left ventricular systolic function with akinesia of all mid and apical segments, hyperkinetic basal contraction, and left ventricular ejection fraction of 35%.

Transfer to the cardiothoracic center took place with emergency diagnostic coronary angiography and cardiac magnetic resonance image demonstrating normal epicardial coronary arteries, apical akinesia with typical ballooning, and basal hyperkinesias without signs of ischemic lesions; diagnosis of takotsubo cardiomyopathy was made (Figs. 3–5). On postoperative day 3, a test transesophageal echocardiography showed normal ventricular function with no segmental wall motion abnormality; on the same day, the troponin I level decreased to 0.04 ng/mL. The patient's postoperative course was uneventful, and on the postoperative day 5, she was discharged to home with aspirin, β -blocker, and angiotensin-converting enzyme inhibitor. The patient has been reviewed in the outpatient clinic and she is well; a subsequent echocardiograph showed a normal volume status with normal wall motion and no mitral regurgitation.

DISCUSSION

Takotsubo is a rare disorder; in a recent study, the annual incidence of this syndrome in a Western population is calculated to be 0.00006%.⁷ Interestingly, in the Japanese population, the prevalence of this syndrome has been reported to be 1%.⁸ There might be a genetic component that has not been yet discovered; the mean age at onset of this syndrome is 62 to 75 years, and it frequently occurs in postmenopausal women.

The pathophysiology of this condition is debatable; numerous hypotheses have been introduced: (1) catecholamine-mediated cardiotoxicity, (2) coronary vasospasm, (3) microvascular dysfunction, (4) left ventricular outflow tract obstruction, and (5) cardiac autonomic imbalance. Catecholamine mediated cardiotoxicity is the most accepted mechanism; patients characteristically present with a preceding history of psychologic/physical stress leading to increased sympathetic activity with a direct catecholamine toxic effect on the cardiac myocytes.

Results are, however, conflicting; some studies report elevated catecholamine levels in takotsubo cardiomyopathy, whereas in others, catecholamine levels are normal; it is more likely that the pathogenesis of this syndrome is multifactorial. ¹¹ The overall prognosis of this condition is favorable; however, fatal complications such as cardiogenic shock and tachyarrhythmias can occur. Although the presence of a psychologic or physical stress is accepted as one of the

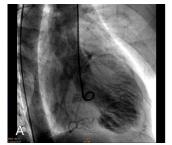




FIGURE 4. A and B, Contrast left ventriculogram showing akinesis of the apical region and hyperkinesis of the basal segments at end-systole (A) and end-diastole (B).

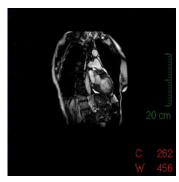


FIGURE 5. Cardiac magnetic resonance image showing left ventricular apical akinesis and basal hyperkinesis at end-systole without signs of ischemic lesions.

main features of the takotsubo syndrome, some cases have been reported to occur without any triggering effect.³

We theorize that stress caused by superficial anesthesia, insufficient analgesia, and surgical pain stimulus created a stressful event that caused a potentially life-threatening catecholamine release as basis of takotsubo syndrome in our patient. Because this condition leads to ventricular dysfunction with apical ballooning, the patients are potentially at a risk for ventricular thrombus formation and decisions regarding anticoagulation need to be discussed. Patients can also develop functional mitral regurgitation with hemodynamic failure; nowadays, no established guidelines exist.⁵

According to the literature, this syndrome has been frequently associated to emotional stress, sexual intercourse, and traumatic injury; it has been also described in patients with epileptic attacks, bronchial asthma exacerbations, dialysis, and during electrophysiological studies.

Wittstein et al¹² described 19 patients with ventricular dysfunction after sudden emotional stress who presented ECG, echocardiographic, and angiographic patterns similar to takotsubo cardiomyopathy; they identified a remarkable increase in catecholamine levels in such patients.

Tsuchihashi et al² described patients with transient apical ballooning associated with delivery, intubation, tracheotomy, lung biopsy, orthopedic surgery, colonectomy, and cholecystectomy.

It is evident that this syndrome is undistinguishable from the myocardial ischemia, and proper diagnosis is possible with angiographic evidence. The course is related to the entity of the left ventricular dysfunction, and signs of cardiac failure can be very severe, although the lesions are reversible.

The characteristic ECG features of takotsubo cardiomyopathy are nonspecific and include dynamic ST elevation (usually less than in acute anterior myocardial infarction) and/or T wave inversion typically throughout the anterior leads^{13,14}; our patient also had elevated levels of cardiac biomarkers.

Transthoracic echocardiography can identify regional wall abnormalities such as akinesia of the apex and/or the midportion of the left ventricle. Typically, the area of dysfunction usually involves a larger territory than that supplied by 1 epicardial coronary artery.

Diagnostic coronary angiography needs to be performed in all patients to exclude obstructive epicardial CAD; ventriculography may demonstrate the apical ballooning that occurs in addition to hypercontraction of the basal segments.¹⁵

Cardiac magnetic resonance imaging provides information regarding functional involvement, chamber dimensions, and presence of intramyocardial edema; it also allows the physicians to exclude infarction and inflammatory processes.¹⁶

© 2011 Mutaz B. Habal, MD 1909

CONCLUSIONS

This syndrome, which presents the same clinical features of a ventricular failure, is probably underdiagnosed, but after the introduction of sophisticated cardiac imaging and coronary intervention, more cases are identified, and an unnecessary thrombolytic therapy can be spared.

This reversible condition, which is, to our knowledge, never mentioned after a craniomaxillofacial surgical procedure, should be considered in the diagnostic algorithm for all patients presenting with acute onset of chest pain, elevated cardiac biomarkers, and ischemic changes on the ECG after a general anesthesia.

REFERENCES

- Satoh H, Tateishi H, Uchida T. Takotsubo-type cardiomyopathy due to multivessel spasm. In: Kodama K, Haze K, Hon M, eds. Clinical Aspects of Myocardial Injury: From Ischemia to Heart Failure. Tokyo, Japan: Kagakuhyouronsya Co, 1990:56–54
- Tsuchihashi K, Ueshima K, Uchida T, et al. Transient left ventricular apical ballooning without coronary artery stenosis: a novel heart syndrome mimicking acute myocardial infarction. Angina Pectoris-Myocardial Infarction Investigations in Japan. J Am Coll Cardiol 2001;38:11–18
- Kondo M, Matsuoka R, Araki M, et al. Assessment of clinical features in transient left ventricular apical ballooning. J Am Coll Cardiol 2003;41:737–742
- Dote K, Sato H, Tateishi H, et al. Myocardial stunning due to simultaneous multivessel coronary spasms: a review of 5 cases. *J Cardiol* 1991;21:203–214
- Kawai S, Kitabatake A, Tomoike H. Guidelines for diagnosis of takotsubo (ampulla) cardiomyopathy. Circ J 2007;71:990–992
- Gianni M, Dentali F, Grandi AM, et al. Apical ballooning syndrome or takotsubo cardiomyopathy: a systematic review. Eur Heart J 2006;27:1523–1529
- Donohue D, Movahed MR. Clinical characteristics, demographics and prognosis of transient left ventricular apical ballooning syndrome. Heart Fail Rev 2005;10:311–316
- Akashi YJ, Musha H, Kida K, et al. Reversible ventricular dysfunction takotsubo cardiomyopathy. Eur J Heart Fail 2005;7:1171–1176
- Akashi YJ, Barbaro G, Sakurai T, et al. Cardiac autonomic imbalance in patients with reversible ventricular dysfunction takotsubo cardiomyopathy. QJM 2007;100:335–343
- Iqbal MB, Moon JC, Guttmann OP, et al. Stress, emotion and the heart: tako-tsubo cardiomyopathy. *Postgrad Med J* 2006;82:29
- Dote K, Sato H, Tateishi H, et al. Myocardial stunning due to simultaneous multivessel coronary spasms. A review of 5 cases. J Cardiol 1991;21:203–214
- Wittstein IS, Thiemann DR, Lima JA, et al. Neurohumoral features of myocardial stunning due to sudden emotional stress. N Engl J Med 2005;352:539–548
- Mitsuma W, Kodama M, Ito M, et al. Serial electrocardiographic findings in women with takotsubo cardiomyopathy. Am J Cardiol 2007;100:106–109
- Kurisu S, Sato H, Kawagoe T, et al. Tako-tsubo-like left ventricular dysfunction with ST-segment elevation: a novel cardiac syndrome mimicking acute myocardial infarction. *Am Heart J* 2002;143: 448, 455
- Klinceva M, Widimský P, Pesl L, et al. Prevalence of stress-induced myocardial stunning (tako-tsubo cardiomyopathy) among patients undergoing emergency coronary angiography for suspected acute myocardial infarction. *Int J Cardiol* 2007;120:411–413
- Mitchell JH, Hadden TB, Wilson JM, et al. Clinical features and usefulness of cardiac magnetic resonance imaging in assessing

myocardial viability and prognosis in takotsubo cardiomyopathy (transient left ventricular apical ballooning syndrome). *Am J Cardiol* 2007;100:296–301

Maxillary Cementoblastoma in a Child

Fábio Wildson Gurgel Costa, DDS, MS,*
Karuza Maria Alves Pereira, DDS, PhD, MS,†
Marcelo Magalhães Dias, DDS, MS,‡
Márcia Cristina da Costa Miguel, DDS, PhD, MS,§
Maria Adriana Skeff de Paula Miranda, DDS, MS, ||
Eduardo Costa Studart Soares, DDS, PhD, MS¶

Abstract: Cementoblastoma is a rare benign tumor that almost always occurs in the premolar or molar region and more commonly in the mandible than in the maxilla. We present a unique incisor maxillary cementoblastoma in an 11-year-old child not previously described. To our knowledge, only 2 maxillary cases, both related to canine teeth, were described in the international literature. Thus, the aim of this article was to discuss the clinical presentation, diagnosis, and subsequent treatment of a patient with a cementoblastoma in the anterior maxillary region.

Key Words: Cementoblastoma, odontogenic tumors, maxilla

ementoblastoma is a rare benign tumor and comprises less than 1% to 6.2% of all odontogenic tumors. This lesion is more frequent in young patients, with about 50% of the cases arising in younger than 20 years. Approximately all benign cementoblastomas are intimately associated and partially enclosed to 1 or more roots of a single posterior mandibular erupted permanent tooth. However, a few articles concerning maxillary lesions have been published, most of them forthcoming in the posterior site.

We present a unique incisor maxillary cementoblastoma in a child not previously described. To date, only 2 maxillary cases, both related to canine teeth, were reported in the international literature. ^{4,5} Therefore, the aim of this work was to discuss the clinical

From the *Division of Stomatology, School of Dentistry, Federal University of Ceará Sobral Campus; †Division of Oral Pathology, Federal University of Ceará Sobral Campus, Ceará; ‡Division of Oral Rehabilitation, School of Dentistry, Catholic Faculty Rainha do Sertão, Quixadá; §Division of Oral Pathology, Federal University of Rio Grande do Norte, Natal; ||Division of Stomatology, School of Dentistry, Catholic Faculty Rainha do Sertão, Quixadá; and ¶Division of Oral Surgery, School of Dentistry, Federal University of Ceará, Ceará, Brazil.

Received August 19, 2010.

Accepted for publication September 25, 2010.

Address correspondence and reprint requests to Fábio Wildson Gurgel Costa, DDS, MS, Coordenação do Curso de Odontologia da Universidade Federal do Ceará Campus Sobral; Av. Comte. Maurocélio Rocha Pontes, s/n.º Derby, CEP 62.041-040, Sobral-Ce, Brazil; E-mail: fwildson@yahoo.com.br

The authors report no conflicts of interest. Copyright © 2011 by Mutaz B. Habal, MD

ISSN: 1049-2275

DOI: 10.1097/SCS.0b013e31822ea6e8

1910