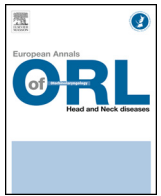




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Case report

Spontaneous arytenoid dislocation and crico-arytenoid instability

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ARTICLE INFO

ABSTRACT

Introduction: Arytenoid dislocation is a rare occurrence, for which the pathophysiology is still unclear.
Methods: We report here an unusual case of spontaneous arytenoid dislocation, which casts doubts on the prevailing classical theory of hemiarthrosis.
Results and conclusions: This case and a review of the literature suggest that arytenoid dislocation could be linked to congenital or acquired arytenoid instability, thus facilitating arytenoid dislocation after even minor trauma. Once the diagnosis is established, we recommend to first attempt reduction, followed by speech therapy, though underlying diseases should be researched.

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1. Introduction

Arytenoid dislocation is a rare occurrence [1,2] the mechanism of which is still unclear [1–8].

We report a rare case of spontaneous crico-arytenoid dislocation (CAD), which provides new insights for both mechanisms and treatment.

2. Case report

While drinking normally from a glass of water, a 34-year old man heard a “cracklem”, immediately followed by persistent cervical pain, dysphonia, and dysphagia. He came in within 2 hours of symptom onset, with important pain (VNRS: 6/10).

The patient was in excellent general condition, without any medical history. Nasal fiberoptic endoscopy with stroboscopy highlighted the right arytenoid malposition with diminished mobility of the vocal fold (Fig. 1), but contraction efforts were still visible. A CT-scan confirmed the diagnosis of arytenoid dislocation (Fig. 2).

The reduction of the dislocation was attempted under direct laryngoscopy under general anesthesia but failed despite several attempts (pushing the arytenoid from below and/or pulling it from above): when reduced, the arytenoid quickly dropped anteriorly in its previous position, without stability after reduction. After discussion with the patient, another attempt was performed the next day by another surgeon, with the same results. A rheumatologic check-

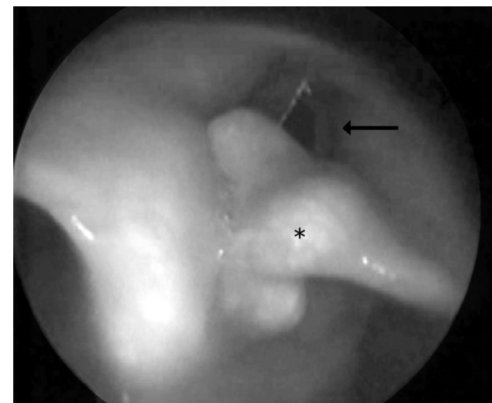


Fig. 1. Endoscopic view of the right dislocated arytenoids (*) in an abnormal position, tilted anteriorly and in external rotation, with an “hammock-like” appearance of the right vocal fold (arrow).

up found no abnormalities. After three months of voice therapy, the arytenoid remained fixed in the same position. Dysphonia was corrected by compensation by the contralateral vocal fold. Dysphagia improved but the patient still described some discomfort.

3. Discussion

The pathophysiology of crico-arytenoid dislocation (CAD) is unclear. Recently, body mass index was pointed out as an independent risk factor for postoperative arytenoid dislocation [3]. In a cadaveric study, Paulsen failed to obtain any CAD on 36 unfixed larynges, despite several attempts of intubation, extubation or

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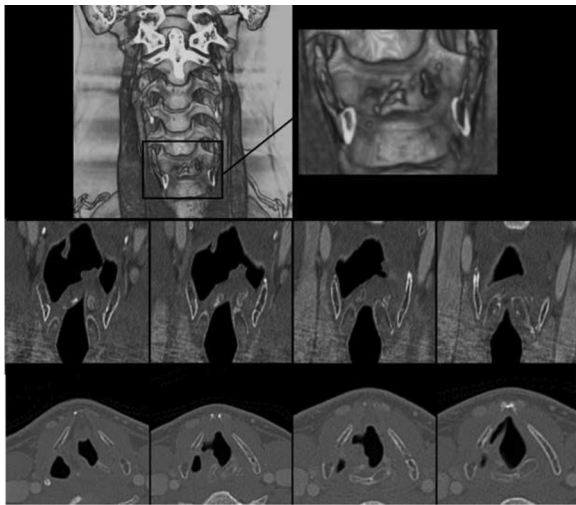


Fig. 2. CT-scan: axial & frontal view and 3D reconstruction showing the displacement and the dislocation of the right arytenoid.

intentional trauma with laryngoscopes [2]. The authors thus suggested that CAD is not directly caused by the trauma but results from the progressive constitution of a hemarthrosis of the cricoarytenoid joint. This hemarthrosis might lead to progressive fibrosis of the joint and subsequent definitive fixation. But our case is not consistent with this theory: the trauma was minor and without a subsequent hematoma. CT-scan and laryngoscopy (performed early after the onset of symptoms) did not find any tumefaction of the joint. Moreover, reduction remained possible, even if it was unstable. In the literature there are many cases of CAD caused by minor trauma: atraumatic tracheal intubations, transesophageal echocardiography, gastrointestinal endoscopy, or laryngeal mask insertion. In all these cases, the forces exerted on arytenoid are described as very subtle. Nerurkar described a case of CAD after a bout of coughing [4] and two other cases of spontaneous CAD are reported in the literature [5,6]. Unfortunately, for the first one, the diagnosis was too delayed to properly analyse the etiology of CAD [5]. The second one was a 82-year old man with acute hoarseness without a traumatic event [6]. These cases and this spontaneous dislocation actually evoke an analogy with glenohumeral instability: there could also be an individual susceptibility to cricoarytenoid joint instability. This might explain the impossibility of obtaining a prolonged reduction of the CAD in our case. Spontaneous reduction of CAD and successful recovery after voice therapy are sometimes possible [7], and is in complete contradiction with the hemarthrosis theory, which should theoretically lead to irreversible fixation of the joint. Recently, Friedman [8] failed to provoke CAD on six cadaveric human larynges with an apparatus simulating forcible collision with the arytenoid and applying high forces (5000 g) on the crico-arytenoid joint. Friedman et al. proposed that neurogenic causes might explain vocal fold motion impairment that has been considered to be due to CAD. They consider that high-placed endotracheal tube cuff may create a neuropathic injury to the adductor fibers of the recurrent laryngeal nerve [8].

But this hypothesis is barely compatible with spontaneous or post-endoscopic (transesophageal echocardiography, gastrointestinal endoscopy...) cases and does not explain pain in our case.

Actually, the inability to obtain CAD in Paulsen's and in Friedman's studies [2,8], despite significant efforts in this direction (resulting in fractures and joint injuries in Paulsen's study) may be due to the fact that none of the cadavers used showed any weakness that could lead to joint dislocation.

This vision of CAD has potential forensic implications. CAD may be more due to individual frailties rather than to technical errors. However, this does not detract from the need for early diagnosis and appropriate treatment.

Clinical suspicion relies on:

- traumatic context;
- intensity of pain and of dysphagia;
- preserved muscular motility.

This last criterion may be very subtle, but is determinant when muscular contractions are seen, and may be assessed in electromyography. Diagnosis can be confirmed by stroboscopy, CT-scan and/or 3D-CT [1]. Electromyography is the reference exam when available.

Once diagnosis is established, most authors agree that the best treatment is to attempt reduction of the dislocation (classically with direct laryngoscopy), followed by voice therapy. With early treatment, better results can usually be expected [1]. Attempting for reduction is all the more important if the physiopathology of the CAD is similar to glenohumeral dislocation. Finally, we also recommend systematically searching for an underlying disease, whether congenital (Marfan, Ehlers Danlos, MASS syndrome, etc. ...) or acquired (rheumatoid arthritis, infectious cricoarytenoid arthritis, etc. ...).

4. Conclusion

This case suggests that CAD might be more due to pre-existing crico-arytenoid instability rather than to the forces exerted on the articulation. This does not preclude a careful approach when exerting actions close to the cricoarytenoid joint.

Disclosure of interest

The authors declare that they have no competing interest.

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